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Stakeholder perspectives on optimal follow-up care: An interdisciplinary economic and psychological investigation into gynaecological cancer

Timmis, Laura

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**Stakeholder perspectives on optimal
follow-up care: An interdisciplinary
economic and psychological
investigation into gynaecological
cancer**

Laura Timmis

**Thesis submitted to Bangor University in fulfilment of the
requirements for the degree of Doctor of Philosophy**

**Centre for Health Economics and Medicines Evaluation
Bangor Institute for Health and Medical Research
Bangor University**

“It's like putting a car in for its MOT and it's fine. And then it, a wheel falls off”

*(Pat, diagnosed with endometrial cancer
4-5 years post treatment)*

Thesis Summary

Aims

This thesis employs an interdisciplinary health economic and health psychology perspective to explore and understand patients, their informal caregivers and Health Care Professionals preferences for gynaecology cancer follow-up care, with the overarching aim of improving the efficiency of care provision.

Methods and Results

A systematic literature review (Chapter 2: n=8) identified that patients had different preferences for care to meet their need for reassurance: hospital follow-up, Specialist Nurse led telephone follow-up, General Practitioner led follow-up and/or self-management. Health Care Professionals valued multidisciplinary models of care and the self-management model of care given the need for a changing model of care. No studies of informal caregivers' care preferences were identified. A qualitative study of patients (n=17) and their informal caregivers' (n=7) preferences for care, based on their experience of care (Chapter 3), identified that follow-up was valued because of the reassurance it provides. For patients, 'access to an expert', 'procedures' and 'holistic care' provided reassurance. 'System failures' and 'low self-confidence' prevented reassurance. For informal caregivers, 'person centred information', which was met through the provision of 'person centred access to care', provided reassurance. The attributes generated from the systematic literature review (Chapter 2), qualitative study (Chapter 3) and other methods were compared (Chapter 4) for the purpose of designing a discrete choice experiment (Chapter 4-5). Attributes generated were dependant on the methods used to identify them (Chapter 4). A stated preference discrete choice experiment (Chapter 5) identified that patients (n=77), their informal caregivers (n=26) and Health Care Professionals (n=67) had different relative preferences, and were prepared to make differing trade-off's for the model of gynaecology cancer follow-up preferred.

Conclusion

This interdisciplinary thesis makes multiple novel contributions to health economics methodology and policy, raising key questions for evidence based decision making for the prudent changing model of care, within resource scarcity.

Acknowledgements

“Storms make trees grow deeper roots”

James this thesis is dedicated to me and you. Thank you for your heartfelt understanding and patience, and for running the house, financially and practically, as I dedicate all my non-parenting moments to finishing this thesis. You certainly meet the requirements of informal caregiver! Now it's my turn to wash up...

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proof reader. To my late Gran who always believed in me. Thank you. This has been of particular comfort and given me strength towards the end of this journey.

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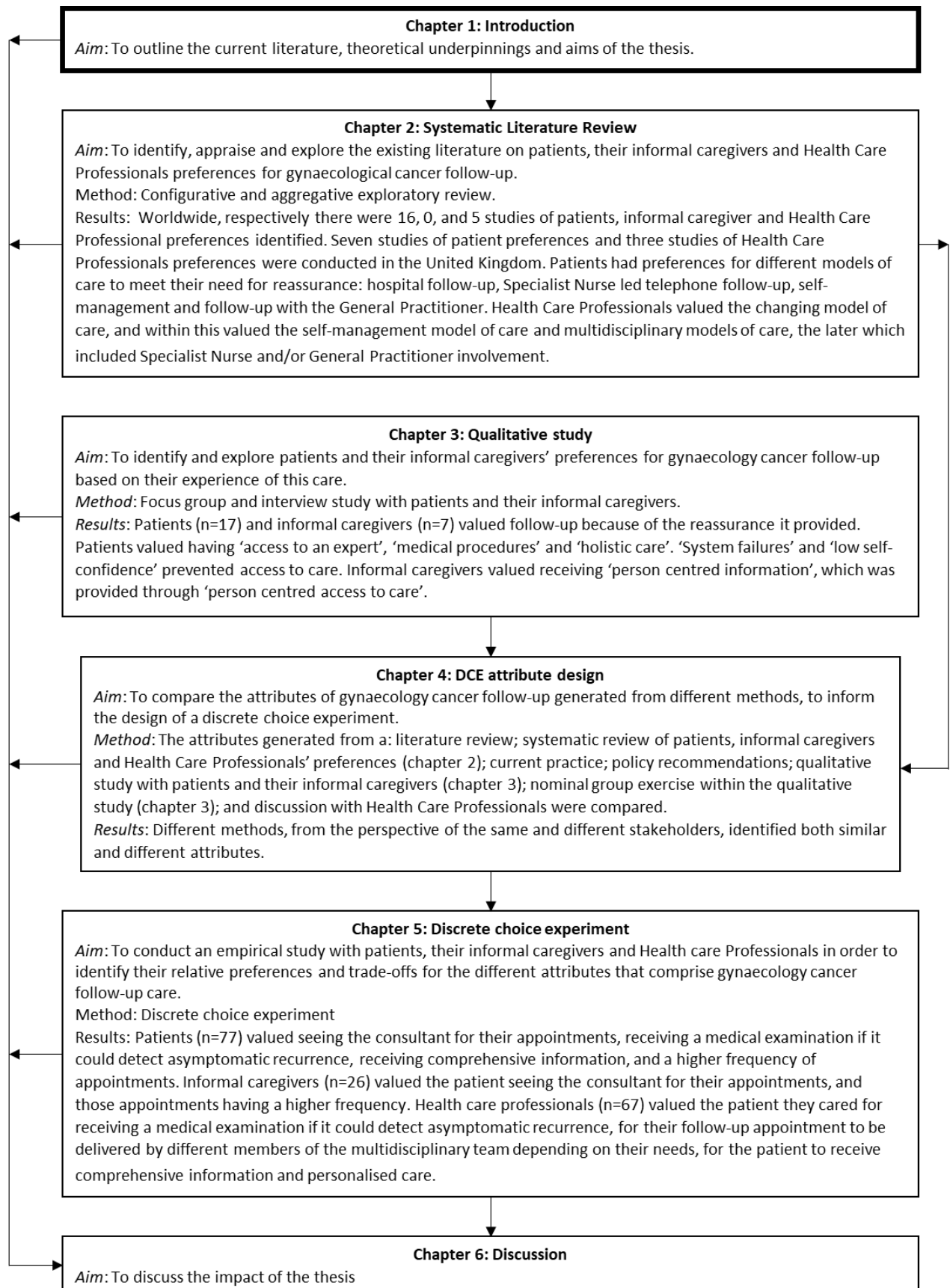
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Chapter 1:

Thesis introduction and background



1.1 Chapter Summary

This chapter provides an overview of gynaecology cancer, follow-up care, the need for a changing model of care, and alternative models of follow-up care to meet this need. The importance of understanding the preferences of patients, their informal caregivers and Health Care Professionals within follow-up care is then explored. The value of taking a health economic and health psychology approach is given before an overview of the thesis, the aims of the thesis, the aim of each chapter and dissemination are discussed.

1.2 Overview of gynaecology cancer follow-up care

1.2.1 *Gynaecological cancer incidence and 5 year survival and recurrence data*

In the United Kingdom, there were respectively 8,984, 7,270, 3126 and 1,339 new cases of endometrial, ovarian, cervical and vulvar cancer in 2015. For patients diagnosed in England and Wales in the year 2010-2011, the 5 year survival rate for endometrial, ovarian and cervical cancer was 79%, 46.2%, and 67.4% respectively. For patients diagnosed in England between 2009 and 2013, the 5 year survival rate for vulvar cancer was 63.6%. This figure is not available for Wales (<https://www.cancerresearchuk.org/health-professional/cancerstatistics/statistics-by-cancer-type>; Personal communication, 2019).

On an individual level survival depends on the type of cancer, the cancer stage (how advanced it is when it is diagnosed), the grade of the cancer (the aggressiveness of the cancer cells and how similar or different the cancer cells are to healthy cells in that area), the outcome of treatment(s), the patients general health and the presence of any co-morbidities (<https://www.cancerresearchuk.org/health-professional/cancer-statistics/statistics-by-cancer-type>). Research shows that patients' needs in the follow-up care period differ by diagnosis and treatment received (Morrison et al., 2012; Salani, 2013).

1.2.2 *Treatment for gynaecological cancer*

The type and extent of treatment given to patients is dependent on the type, stage and grade of the cancer, the patients' general health and treatment preferences, as well as their postcode (post-code lottery: Target Ovarian Cancer, 2015; <https://www.cancerresearchuk.org/about-cancer/womens-cancer>). Options for treatment

following a diagnosis of gynaecological cancer include surgery and/or chemotherapy and/or radiotherapy including brachytherapy (internal radiotherapy). Patients may receive chemotherapy before and/or after surgery (<https://www.cancerresearchuk.org/about-cancer/womens-cancer>).

1.2.3 Effects of diagnosis and treatment

Long term effects of gynaecology cancer are dependent in part on the exact diagnosis received (site of gynaecological cancer, stage of disease and grade), treatment received (surgery, chemotherapy, radiotherapy and brachytherapy including the more precise details of this for example, chemotherapy drug administered), as well as the patients' premorbid or concurrent physical and psychological wellbeing (Salani., 2013). However, many effects of the cancer, its diagnosis and treatment are universal. Physical symptoms may include: symptoms of menopause (including hot flushes, irritability and vaginal dryness); lymphedema (swelling and pain) in the legs; and peripheral neuropathy (sensory, motor, automatic or mono neuropathy, for example, numbness or loss of bowel control). Psychological symptoms may include: psychological distress (including anxiety, depression and fear of cancer recurrence); and sexual health dysfunction. This description is not exhaustive, but a summary of common and shared symptoms regardless of precise gynaecology cancer diagnosis and treatment (Hodgkinson et al., 2007; Salani., 2013). Despite these possible side effects, most patients do not report a long term impact of diagnosis and treatment on their quality of life (Chan et al., 2001; Dahl, Wittrup, Væggemose, Petersen, & Blaakaer, 2013; Wenzel et al., 2002).

1.2.4 Follow-up care

Post treatment, patients traditionally have regular follow-up appointments in secondary care with the aim of: detecting possible disease recurrence under the assumption that early detection may increase survival; managing physical and psychological effects of treatment; providing patients with reassurance and collecting data for the purpose of research (Hall & Rustin, 2011; Rustin et al, 2010). In the literature, the definition of follow-up care varies. For the purpose of this thesis, follow-up care was defined in collaboration with study collaborators Mr Simon Leeson (Consultant Gynaecologist and Oncologist, BCUHB) and Professor Nicholas Stuart (Consultant Oncologist, BCUHB and Bangor University). The definition of follow-up care was:

Care delivered from three months post-treatment, provided by the National Health Service (NHS) and offered to all patients treated with curative intent (dependant or not dependant on risk of recurrence).

1.2.5 Guidance for the delivery of follow-up care

Despite the rationale, there is no NICE guidance or medical consensus and limited evidence as to what form or frequency of gynaecological cancer after care is most effective in detecting a recurrence of cancer before the presence of symptoms (asymptomatic recurrence). The aim of asymptomatic detection of cancer recurrence is to increase overall survival due to earlier onset of secondary treatment (Clarke et al., 2014; Elit et al., 2009; Fung-Kee-Fung et al., 2006; Lanceley et al., 2013; Leeson et al., 2017; Nama, Nordin & Bryant, 2013; Rustin et al., 2010). This results in a postcode lottery of follow-up care provision (Leeson, Stuart, Sylvestre, Hall & Whitaker, 2013).

Recent guidance for the follow-up of gynaecological cancer in the United Kingdom has been published (British Gynaecological Cancer Society and Royal College of Obstetrics and Gynaecologists, 2014; Fotopoulou et al., 2017; Sundar et al., 2017). However, the guidance varies in their evidence base and comprehensiveness. Guidance for the follow-up of endometrial cancer is comprehensive recommending a personalised stratified approach (Sundar et al., 2017). Guidance for the follow-up of ovarian and vulvar cancer is brief and takes a traditional non-personalised approach (British Gynaecological Cancer Society and Royal College of Obstetrics and Gynaecologists, 2014; Fotopoulou et al., 2017). There is currently no national guidance for the follow-up of cervical cancer.

1.2.6 *Current model of care*

The traditional model of follow-up appointments has an approximate annual cost of £9.5 million to the NHS, with a unit cost of £116 (for a consultant led, non-admitted, face-to-face attendance at a gynaecology oncology follow-up clinic: NHS improvement, 2018). However, within North Wales, a trial exploring standard hospital follow-up compared to personalised Specialist Nurse led follow-up, identified a unit cost of £52.99 per traditional follow-up appointment (Morrison et al., 2018). However, the authors do not report variation in this estimate. These different costs for the traditional model of follow-up might be explained by

different models of care being delivered across the United Kingdom. In 2012 an audit of the model of follow-up care delivered in the United Kingdom was conducted (Leeson et al., 2013). Findings identified that follow-up varied by the mode of care (face-to-face; telephone; patient initiated; or a combination of these), the Health Care Professional that delivered care (hospital Doctor; Specialist Nurse; General Practitioner), the medical investigations given (examination; ultrasound; CA125 testing; other blood tests; CT scan, MRI; cytology; colposcopy; vulvoscopy), the clinic these appointments were delivered in (single speciality clinics or combined clinics, for example surgery and medical oncology), rapid re-access if concerns (ranging from 2-4 weeks, however, not all patients were given information with the contact details for the clinic to gain re-access if required), and the duration of care (ranging from 2 years to 5 years: Leeson et al, 2013). The frequency of care delivered was not explored in this audit. Nevertheless, this audit was conducted before the development of the aforementioned guidance (British Gynaecological Cancer Society and Royal College of Obstetrics and Gynaecologists, 2014; Fotopoulou et al., 2017; Sundar et al., 2017), so a more up to date audit may yield different results. It is known that the self-management model of care/patient initiated care is delivered within some health boards (see alternative models of care: University Hospitals of Leicester NHS trust, 2016; University Hospital Southampton NHS foundation trust, 2014).

1.2.7 Does the current model of follow-up meet the aims of follow-up?

Recurrence of gynaecology cancer is often symptomatic, and is most likely to occur in the two years following treatment (Kerr-Wilson & McCrum, 1995). Therefore, patients with symptoms of potential recurrence often present in between scheduled appointments, and are scheduled a timely appointment for further investigation (Jefford et al., 2013; Jeppesen et al, 2017; Leeson et al., 2017; Leeson et al, 2013; Lim, Howells & Evans, 2004; Oonk et al., 2003; Vistad, Cvancarova & Salvesen, 2017). However, the provision of scheduled follow-up appointments can lead some patients to wait until their next appointment to present with potential symptoms of recurrence, thus delaying the onset of further possible treatment (Olaitan et al., 2001). Specifically:

1.2.7.1 Endometrial cancer

A systematic literature review exploring detection of recurrence and survival in endometrial cancer identified that in a pooled data sample of 2922 patients, 2.6% patients (n=77, 95% CI 74-81) were diagnosed with symptomatic recurrence, and 0.8% patients (n=23, 95% CI, 19-26) were diagnosed with asymptomatic recurrence (Fung-Kee-Fung et al., 2006). It was reported that 68% to 100% of recurrences were detected in the first three years post-treatment. Interestingly the authors report that there were no statistically significant differences in survival between patients whose recurrence was detected symptomatically compared to asymptotically, or by model of follow-up received (comprised of appointments over different frequencies which entailed different tests, with (Fung-Kee-Fung et al., 2006). This finding questions the rationale for the follow-up of endometrial cancer. A Cochrane review of endometrial cancer follow-up is currently being conducted (Aslam et al., 2016).

1.2.7.2 Ovarian cancer

A Cochrane systematic literature review (Clarke, Galaal, Bryant, Niak., 2014) to evaluate ovarian cancer follow-up identified one study (Rustin et al., 2010). This randomised trial (Rustin et al., 2010) demonstrated that in a sample of 529 participants, asymptomatic detection of recurrence of ovarian cancer through monitoring CA125 levels, resulted in earlier detection of recurrence and so earlier onset of secondary treatment, but had no effect on survival and a reduced effect on quality of life compared to symptomatic detection of recurrence (Rustin et al., 2010). As treatment for a recurrence of ovarian cancer has shown to not have a significant effect on survival, patients may not be treated for an identified recurrence until symptomatic, thus questioning the rational for ovarian cancer follow-up (Geurts et al., 2012; Leeson et al., 2017).

1.2.7.3 Cervical cancer

A Cochrane systematic literature review to evaluate cervical cancer follow-up identified no randomised controlled trails (Lanceley, Fiander, McCormack & Bryant, 2013). A systematic literature review that included prospective studies identified 17 studies comprised of 5510 patients diagnosed with stage I-IV disease (Elit et al., 2009). In individual studies, between 8%-26% of patients experienced a disease recurrence. Of the individual studies that reported survival by symptomatic and asymptomatic detection of recurrence, survival following

symptomatic detection of recurrence ranged from 8–38 months and survival following asymptomatic detection of recurrence ranged from 8–42 months (for one of these studies the median survival following asymptomatic detection could not be reached due to the study period ending: Zola et al., 2007; Elit et al., 2009).

An audit conducted in Wales of 291 patients post-treatment for early stage cervical cancer, demonstrated that 53/291 (18%) were diagnosed with a recurrence (Lim, Howells & Evans, 2004).

- For 6 patients it could not be identified how the recurrence was identified
- Seven of these recurrences were identified in the follow-up clinic
 - Of these, 2 patients were asymptomatic and 5 symptomatic
- 40 patients were diagnosed through re-presenting at the clinic through self-referral or referral by another Health Care Professional (and so symptomatic)

Therefore only 2/291 (0.7%) of patients experienced recurrence which was detected when asymptomatic.

- Median survival following diagnosis of symptomatic self-presentation in between scheduled appointments had a median survival of 26 months (n=40 patients)
- Recurrence detected based on symptomatic presentation at scheduled follow-up appointment had a median survival of 37.5 months (n=5 patients)
- Asymptomatic detection of recurrence had a mean survival of 8.3 months (n=2 patients)

It is reported that these differences were not statistically significant (the figures were not reported), but this could be attributed to the small sample sizes (Lim, et al., 2004). Nevertheless, the rationale for cervical cancer follow-up in meeting the aims of gynaecology cancer follow-up is questioned.

1.2.7.4 Vulvar cancer

Recurrence of vulvar cancer has received less research attention of a robust nature. A literature review reported that recurrence of vulvar cancer ranged from 12-37%, with the main variance in recurrence reported to be due to the size of the cancer and whether it had spread to the lymph nodes (Coulter & Gleeson, 2003). However, A cross-sectional study of hospital data for 238 patients diagnosed with stage I-IV vulvar carcinoma, identified that

65/238 (27%) of patients experienced a recurrence over a mean of 63 months (range of months since treatment 6 – 149 months: Oonk et al., 2003).

- Twenty three of these recurrences were detected based on symptomatic re-presentation at the clinic between scheduled appointments.
- Forty two of these recurrences were detected at the scheduled follow-up appointment.
 - Twenty one of these patients were symptomatic and 21 asymptomatic.

The authors report no difference in mean survival between patients diagnosed with recurrence at their scheduled appointments compared to if they re-presented at the clinic with symptoms ($P=0.22$). However, do not report survival by symptomatic and asymptomatic detection (Oonk et al., 2003).

Psychologically, the traditional model of follow-up care has been shown to lead to high levels of anxiety amongst some patients before and following the follow-up appointment (Greimel, Lahousen, Dorfer, Lambauer & Lang, 2011; Kew, Galaal, & Maderville, 2009). A cross-sectional study identified that anticipatory anxiety is severe in some patients (14%) whilst in others it is moderate (20%) or low (66%: Greimel et al., 2011).

Gynaecological cancer follow-up patients often live with a fear of recurrence, and value follow-up care because of the medical reassurance it provides (Brandenberg, Berendsen, & de Bock., 2017; Bradley, Pitts, Redman & Calvert, 1999; Lydon, Beaver, Newbury & Wray, 2009; Oshima et al, 2011; Sekse, Raaheim, Blaaka & Gjengedal, 2010: see Chapter 2). A qualitative study with sixteen gynaecology cancer patients at least five years post treatment, identified that a fear of cancer recurrence, and thus possible loss of life, was present throughout the follow-up period and beyond. It was reported that women were constantly watching their body, vigilant to signs and symptoms which may indicate a possible cancer recurrence (Sekse et al., 2010). Findings from a focus group study confirmed that post treatment patients were living with uncertainty in part due to a perceived risk of recurrence, and these women valued follow-up appointments because of the reassurance provided in absence of disease detection (Oshima et al., 2011).

Oshima et al (2011), participant number 15: "I don't have any particular problems. During every visit, I feel relieved knowing that everything is all right."

A cross-sectional study identified the prevalence of perceived risk of recurrence before a follow-up appointment in a sample of 210 patients diagnosed with gynaecology cancer in Austria (Greimel et al., 2011). It was identified that pre-appointment, 10% (n not given) of participants experienced a severe fear of recurrence, 22% a moderate fear of recurrence, 30% a little fear of recurrence, whilst 38% did not experience any fear of recurrence (Greimel et al., 2011).

Patients also report unmet needs in the follow-up care period (Beesley et al., 2008; Hodgkinson et al, 2007; Miller, Pittman & Strong, 2003). A cross-sectional study with 802 patients who were between three months and five years post diagnosis, identified that 43% (n not given) of women had one high or moderate unmet need, as assessed using the supportive care needs survey short form (SCN-SF, Beesley et al., 2008; McElduff et al., 2004). In support of the findings regarding the saliency of a fear of cancer recurrence, this study identified that the most common high or moderate unmet need, was a need for support due to fear of the cancer spreading (n=123, 17%), followed by concerns about people close to them (n=109, 15%), and uncertainty about the future (n=104, 14%). Interestingly, patients treated with chemotherapy were more likely than those not treated with chemotherapy to report to have higher levels of unmet needs regarding physical/daily living, psychological needs and needs regarding the health system/information (Beesley et al., 2008). The authors measured the presence of radiotherapy to explore the association between this treatment and needs, but did not report these results.

In light of the above, it is not surprising that some patients are open to, or have a preference for alternative models of care (Cox et al., 2008; Lydon et al., 2009: see Chapter 2 for further discussion).

The efficacy of the traditional model of follow-up in meeting the aims of follow-up, combined with the financial cost to the NHS, raises the question of opportunity cost. Opportunity cost in this context, is defined as the benefits forgone from these resources being invested in gynaecology cancer follow-up, compared to the next best alternative (Morris et al., 2007).

It is proposed that the aim of gynaecology cancer follow-up should be modified, ensuring that it has an evidenced based purpose, with this modification being transparent to patients, their informal caregivers and Health Care Professionals (Lajer et al., 2010). It is recommended that this model of care is flexible to meet patients' individual physical and psychological needs (Leeson et al., 2017). In support of these research led recommendations, there is an increased policy focus on the need to improve patients' outcomes in the follow-up period, stratifying care to meet patients' individual needs (Wales Cancer Network, 2016). These recommendations are in line with the Welsh NHS strategy for prudent health care (Welsh Government and NHS Wales, 2016).

Prudent health care

Within Wales, there is a policy focus on prudent healthcare with an aim to ensure that the care the NHS provides adds value, improves patients' outcomes and is sustainable within the finite resources available, and ever infinite needs (Welsh Government and NHS Wales, 2016). Prudent healthcare applies to all aspects of NHS care in Wales: service design, management and delivery. There are four principles which guide prudent health care:

1. The public, patients and Health Care Professionals are co-producers within the aim to achieve health and well-being.
2. Care provided for those with the greatest need first, with the effective use of healthcare resources and skills.
3. To provide only the care needed, not more care, not less care, and to do no harm.
4. To reduce variation in care delivery through the development of evidenced based practice, consistently and transparently adhered to.

Prudent health care calls for action throughout Wales to:

- 1) Reduce unnecessary care and ensure patients are able to make informed decisions about the care they receive.
- 2) Change the outpatient model of care, so that patients can receive specialist advice within primary care.
- 3) Integrate healthcare and public services, "to provide the right care, in the right place, at the right time".

(Welsh Government and NHS Wales, 2016)

It is clear from the above that prudent health care is not currently delivered within gynaecological cancer services within the United Kingdom, and specifically, Wales where this is a governing strategy focus.

1.3 Alternative models of follow-up care

Alternative models of care include follow-up:

- Delivered face-to-face by the gynaecology cancer Specialist Nurse.
- Appointments delivered on the telephone by the gynaecology cancer Specialist Nurse.
- Delivered by a General Practitioner.
- Self-management or patient-initiated care (where patients are informed of signs of recurrence and directly request an appointment with secondary care if they experience any signs of recurrence and/or have any concerns).
- The provision of a health and wellbeing clinic and holistic needs assessments, leading to a care plan, onward referral and treatment summary.

(Howell et al., 2012; Lewis et al., 2009a; Lewis et al 2009b; Lewis et al., 2009c; Morrison et al., 2018; Sandsund et al., 2017; National Cancer Survivorship Initiative, 2013)

The National Cancer Survivorship Initiative (2013), argue for a stratified approach whereby there are three levels of care, assigned based on the patients' needs. Level one is comprised of supported self-management and open access to the clinic; level two is comprised of shared care between the patient and secondary care clinician; and level three is comprised of multidisciplinary team led complex case management (National Cancer Survivorship Initiative, 2013). It is reported that a stratified approach to cancer follow-up could improve the efficiency of cancer care (defined as the use of health care resources in a way that obtains the best value for money: Williams, 1988), enabling reinvestment of resources from the un-necessary follow-up of healthy patients to more intense services to meet the needs of patients that require more support post-treatment (Allirajah, 2010; Leeson et al., 2017; National Cancer Survivorship Initiative, 2013).

There is increasing research into the effectiveness and cost-effectiveness of alternative models of gynaecology cancer follow-up care (Beaver et al., 2017; Brothers, Easley, Salani &

Andersen, 2013; Lanceley et al., 2016; Morrison et al., 2018). A randomised nested cross-sectional study exploring the provision of survivorship care plans compared to standard care, identified no significant difference between patients' perception of care, and perceived helpfulness of the information received (Brothers et al., 2013). It must be noted that gynaecology cancer follow-up patients were recruited within one year of treatment completion to receive this model of follow-up, and only received one survivorship care plan at one appointment (Brothers et al., 2013).

A randomised control trial explored the provision of personalised versus standard follow-up in ovarian, fallopian tube and peritoneal cancer follow-up patients (Lanceley et al., 2016). In this study the personalised model of care was comprised of a holistic needs assessment followed by flexible contact (face-to-face or telephone), managed by the Specialist Nurse. However, the authors state that the intervention may also have included the provision of medical tests and face-to-face appointments with the Physician. Standard care was comprised of three monthly face-to-face appointments with the Physician and if the patient presented with symptoms, medical tests. Results identified a positive effect of personalised follow-up on quality of life and patient satisfaction with care, and a reduced cost of care, however, delayed expression of symptoms of recurrence was also reported. The effect of this on survival is unknown (Lanceley et al., 2016).

A randomised trial (Beaver et al., 2017) and randomised feasibility trial (Morrison et al., 2018), demonstrated different results regarding Specialist Nurse delivered telephone follow-up compared to hospital follow-up. A feasibility trial comprised of personalised Specialist Nurse led telephone follow-up (based on responses to validated needs assessment measures) at 3 and 6 months post treatment; a booklet with comprehensive information; and onward referrals where necessary, demonstrated an improved effect on quality of life and at a reduced cost of care to the NHS compared to standard care. Standard care was comprised of a three and six month Consultant led face-to-face appointment including a physical examination (Morrison et al., 2018). However, a randomised control trial comprised of a telephone intervention delivered by the Specialist Nurse had no effect on quality of life or satisfaction with care, but increased satisfaction with information provided in the appointment, improved communication between the patient and Health Care Professional, and increased perception of the Health Care Professional's knowledge of them, compared to standard care (Beaver et al., 2017). In this study standard care was comprised of a face-to-

face appointment with the Doctor or Specialist nurse and included a physical examination (Beaver et al., 2017). Overall patients in the intervention arm received more frequent care of a longer duration than standard care, resulting in no difference in the cost to the NHS (Dixon et al., 2018). It must be noted that the feasibility trial was conducted at the start of follow-up care, so patients had not received an alternate model of care (Morrison et al., 2018), unlike in the later trial (Beaver et al., 2017). In both trials of Specialist Nurse led telephone follow-up, patients reported either themselves or their informal caregivers wanting the patient to receive care only by a Consultant, as a key reason for not consenting to take part in the trial (Beaver et al., 2017; Morrison et al., 2018). This raises the importance of preferences for care informing research trial development (Chalmers et al., 2013). It is increasingly recognised that health care and cancer care more specifically, as well as prudent health care in Wales, requires a collaboration between patients, their informal caregivers and Health Care Professionals (Deshields et al, 2012; Welsh Government and NHS Wales, 2016). Therefore the preferences of these stakeholders need to be understood.

1.4 Preferences

1.4.1 Preference of patients

There is a difference between patients' preferences for care and the health care services that are delivered (Mulley, Trimble & Elwyn, 2012), and research that is prioritised (Chalmers et al., 2013). There is an increasing emphasis on the provision of patient centred services to meet the individual needs of patients and so improve patients' outcomes within health care generally (Coalition for Collaborative Care, 2014; The Kings Fund, 2010) and more specifically, within cancer services in Wales (Wales Cancer Network, 2016; Welsh Government and NHS Wales, 2016) so that patients receive:

"...the care they need (and no less), and the care they want (and no more)"

(Mulley et al., 2012, pg 2)

Therefore the 'right care' to be delivered is dependent on both the patients' medical diagnosis and their preferences for care (Mulley et al., 2012). Understanding patients' preferences would improve patients outcomes, improve the ethics of the health care system and health

care policy (by only delivering services that patients want to receive), increase the confidence of Health Care Professionals (with the knowledge that they are treating patients how they wish to be treated), and reduce inefficiencies in current health care delivery (by no longer delivering services that patients do not want to receive), thus allowing the re-allocation of funding without a large effect on patients outcomes (Dixon, 2012; Mulley et al., 2012). Interestingly, because of this, when care is aligned with patients' preferences, patients consume less care, resulting in a reduced cost to the health service (Wanless, 2002; Wennberg, Marr, Lang, Malley & Bennett, 2010). Without understanding patients' preferences for services, policy makers and commissioners are making decisions about the allocation of resources and delivery of services without a true understanding of the demand for a service (Mulley et al., 2012). Preference misdiagnosis (the misdiagnosis of patients' preferences for care) is discussed as being equivalent to medical misdiagnosis in its effect on both patients' outcomes and the efficiency of care, but differs in that preference misdiagnosis is a silent (Mulley et al., 2012). Care aligned to patient preferences is similar to preference-sensitive health care, but differs in that preference-sensitive care refers to decision making where the risks and benefits of different treatment options are known (Dartmouth Atlas Project, 2007).

“Preference-sensitive care comprises treatments that involve significant tradeoffs affecting the patient’s quality and/or length of life. Decisions about these interventions – whether to have them or not, which ones to have – ought to reflect patients’ personal values and preferences, and ought to be made only after patients have enough information to make an informed choice”

(Dartmouth Atlas Project, 2007, pg1)

In this decision making context, there is currently insufficient evidence regarding the benefits and risks of different models of gynaecology cancer follow-up.

1.4.2 Preference of informal caregivers

Informal caregiving is defined as the act of providing care to promote the welfare of a family member (or friend), above and beyond the care they would normally provide as part of their

role, for example when challenges to their health are experienced (Pearlin, Mullon, Semple & Skaff, 1990; Revenson et al., 2016). Informal caregivers often have no choice as whether or not they provide care, but instead the illness of a family member thrusts them into this role, often with little warning or preparation time (National Alliance for caregiving and AARP, 2015). Within this role informal caregivers may provide support with activities of daily living for example washing and assistance walking, emotional support, and/or practical support with hospital visits (Hodgen, Greenfield, Nugus & Kierman, 2013; Williams, Tisch, Dixon & McCorckle, 2013; Wolff & Roter, 2011). It is consistently reported in the literature that the presence of spousal support affects the patients' quality of life (Courten, Stevens, Crebolder & Philipsen, 1996; Leung, Pachana & Deirdre, 2014; Luszczynska, Pawlowska, Cieslak, Knoll & Scholz, 2013).

As a result of having this role and the individual tasks undertaken, there may be a significant positive and/or negative effect on the informal caregivers' own physical and psychological health outcomes (Romito, Goldzweig, Cormio, Hagedoorn, & Andersen, 2013; Stafford & Judd, 2010). The outcomes of caregiving may continue after the need for caregiving has ended (Kim, Spillers & Hall, 2012). Because informal caregivers are not the direct recipient of a patient's care, and due to the possible impact of caregiving, informal caregivers have been referred to as an invisible patient (Brodaty & Donkin, 2009). Because of the role and impact of caregiving there is a policy focus on understanding and involving informal caregivers as well as patients in the decision making for patients' care (Department of Health, 2012). The preferences of informal caregivers have been shown to both be concordant and discordant to the preferences of patients, for the patients' care (Shin et al., 2013).

1.4.3 Preference of Health Care Professionals

In accordance with the principal/agent theory (Buchanan, 1988), patients trust Health Care Professionals to make the best decisions regarding their treatment and care, under the assumption that as the Health Care Professional, they are the expert (agency relationship). The perfect agency relationship is one in which the agent (Health Care Professional) makes the decision that the patient (principal) would regarding their care, if they had the same level of expertise as the agent (Health Care Professional: Buchanan, 1988). However, a systematic literature review exploring concordance and discordance between patients and Health Care Professionals' preferences as measured in a discrete choice experiment, has demonstrated

that patients and their Health Care Professionals' preferences for care are often discordant to varying degrees (Harrison, Milbers, Hudson & Bansback, 2017), despite Health Care Professional's believing that they are in-tune with their patients' preferences (Mulley et al., 2012). Even when there is guidance for the delivery of follow-up care, some Health Care Professionals deviate from this in accordance with their own preferences for care provision (van Hezewijk et al., 2011). Given the above, there is a need to understand Health Care Professional's preferences for patients care, particularly as patients consume less care where the provision of care matches their preferences (Stacey et al., 2011).

1.5 The application of health economics and health psychology to understanding preferences for care

Health economics and health psychology are both concerned with understanding, explaining and predicting health behaviour (Hanoch & Gummerum, 2008). Health economics is concerned with the allocation of finite resources when needs are infinite, with an overarching aim of improving the efficiency and equity (defined as the fairness in the distribution of health care resources), of health care service provision (Morris et al., 2007). Whilst efficiency and equity are both aims, there is a trade-off between efficiency and equity, they cannot be equal goals (Brazier et al., 2007; Morris et al., 2007). Health economics addresses three questions within this, 1) the decision about which services to produce, 2) how these services should be produced, and 3) who should receive these services (Kernick, 2003). Health psychology takes a wider biopsychosocial approach to understanding the biological, psychological (personal, cognitive, emotional, behavioural) and social (including contextual and cultural) factors involved in illness causation, prevention and management, and in the maintenance and promotion of health. Health psychology aims to use this understanding to improve health care systems and health care policy (Engel, 1980; Engel, 1977; Matarazzo, 1982; Morrison & Bennett, 2016). Bringing health economics and health psychology together in a series of studies to explore preferences for gynaecology cancer follow-up should add to understanding of preferences and the underlying illness representations, experiences, attitudes and beliefs governing these. This interdisciplinary approach will add to the methodological and policy contributions that this thesis can make.

1.5.1 Health Economics

1.5.1.1 Market failure

From a health economic perspective, many goods are traded under a competitive market, where there is equilibrium between the demand and supply of goods (Brazier et al, 2007). However, a market system fails in health care for three reasons, 1) lack of certainty, 2) lack of information symmetry, 3) externalities (Arrow, 1963; Culyer, 1971). Within health care, there is a lack of certainty regarding a patient's/future patient's demand for future health care as when an individual becomes ill, and the health care they will need when they become ill, cannot be predicted. Additionally, the benefits gained from a health service are not equal between individuals, so the benefit of a health service cannot be predicted with certainty (Arrow, 1963). Within health care, there is also a lack of information symmetry between the consumer (patient) and provider of care (Health Care Professional). Generally, even if patients have asked questions and researched the disease and treatment options (as they are increasingly likely to in this age of online medical repositories and social media), they are still likely to be relatively uninformed about the disease they have, the options for treatment and potential outcomes compared to the Health Care Professional. Therefore, Health Care Professionals are required to act as both the agent (making the same treatment decisions that the patient would if they were fully informed) and supplier of health care. As well as the above posing concerns regarding the agency relationship, and the respective ability of Health Care Professionals to identify and measure demand and treat these demands, it can lead to supplier induced demand (Arrow, 1963). Conversely, Health Care Professionals are generally uninformed of patients' preferences for care (Mulley et al., 2012). If Health Care Professionals were accurately informed and acted on patients preferences, and if patients were accurately informed about the disease they have, options for treatment and outcomes, health care would work much more closely to the perfect market (Mulley et al., 2012). A market system also fails due to external factors, for example, when individuals choose not to consume the amount of care they need, for whatever reason (Culyer, 1971).

As a market system fails in health care it requires government intervention. In the United Kingdom government intervention is through the management and delivery of health care through the provision of the NHS. It is the aim of the NHS to provide equitable access to meet the health care needs of the population, based on the clinical need of the individual and not their ability to pay, with no monetary cost (<https://www.nhs.uk/using-the-nhs/about-the->

nhs/principles-and-values/). There are finite resources and infinite health care needs. Therefore trade-offs need to occur with the aim of increasing efficiency and/or equity (Brazier et al., 2007). The allocation of resources to one person, or one service, takes away the resources that could be allocated to the next person or service, and those benefits. This is known as opportunity cost. Economic evaluation importantly provides evidence to aid policy makers and health care commissioners with their decision making for the allocation of scarce resources where needs are infinite (Brazier et al., 2007).

1.5.1.2 Demand for health care

Within health economics, demand is defined as a health care want, whilst a health care need is defined as the capacity to benefit from health care, as assessed by the Health Care Professional. Therefore, there may be a health care need but no demand, or a demand but no health care need. In accordance with economic theory, demand for health care is determined by: the price of the good; an individual's income; the cost of other goods; individual taste and societal influence; the size of the population and its composition (Morris et al., 2017). Grossman's model of demand distinguishes between health care as an input, and health as an output, measured in terms of utility. In accordance with this model, health care (as well as other factors) is demanded because it improves health, with individuals seeking to maximise their health within their available budget (Grossman, 1972).

1.5.1.3 Discrete choice experiments

Discrete choice experiments are a method of stated preference evaluation, and enable exploration of the value of a service and the trade-off's that consumers are prepared to make for their preferred service, for example, model of care, when there is no competitive market for this service (Ryan 2004; Ryan, Gerard and Amaya-Amaya, 2008). This is of key importance and benefit to policy makers and health care commissioners, and as such it is recommended that the National Institute for Clinical and Health Excellence (NICE) use discrete choice experiments as a way of identifying preferences for health care (Hanemann & Kanninen, 1999; Ryan, 2004). Discrete choice experiments are underpinned by consumer choice theory, Lancaster's theory of value (Lancaster, 1966) and random utility theory (McFadden, 1974).

In the late 19th century economics started to acknowledge the perceived value of goods and services, where the perceived value was defined by how useful the good or service was

(its utility). In accordance with consumer choice theory, it is assumed that individuals are rational decision makers whom aim to maximise their utility for good health, and make decisions that they perceive will maximise their health (Grossman, 1972). Therefore when presented with different bundles of attributes, individuals will assign utility to each off the bundles they are presented with, and will make a choice for which bundle of attributes provides them with the most utility within their budget, with choice being a reflection of preferences (Lancaster, 1966). However, discrete choice theory differs to consumer choice theory for three reasons.

- 1) It assumed that the bundle of attributes define the utility that it provides, and so a discrete change in the bundle of attributes can cause a preference switch from one bundle of attributes to another (Lancaster, 1966).
- 2) The choice between a service is defined by alternatives that are finite and mutually exclusive (the respondent is only able to make a choice for one bundle of attributes, described by the attributes and levels that comprise it), as opposed to being able to purchase/choose an infinite number of services. For example, making a choice between 'Follow-up A' and 'Follow-up B'.
- 3) As opposed to being deterministic, individual choice behaviour is random as it is intrinsically probabilistic (Random utility theory: McFadden, 1974). Therefore utility and choice is in part due to an observable function (utility caused by the bundle of attributes and its alternatives), and partly due to a random component (as not all factors affecting an individuals' preferences can be measured and observed: Manski, 1977).

In discrete choice experiments participants are presented with a series of pairwise choices, and for each pairwise choice, are asked to make a decision for which hypothetical model of care they have a preference for, for example 'Follow-up Care A' or 'Follow-up Care B'. The hypothetical models of care are defined by the attributes (and specific levels) that comprise it. For example, Health Care Professional seen (with attribute levels: Consultant; Registrar; Specialist Nurse; General Practitioner), content of care (with attribute levels: examinations and tests; psychological support) and duration of care (with attribute levels: 2 yearly; 5 yearly). Participants' responses provide information on: 1) whether an attribute is important to participants, 2) the relative importance of attributes, 3) the preferences ordering within each

attribute, 4) the trade-off between attributes that participants are prepared to make to have a one unit increase in another attribute level (also known as marginal rate of substitution: MRS), and 5) the overall utility of a service (Ryan, 1996).

1.5.2 Health Psychology

Health psychologists take a different approach to decision making, using social cognitive models to understand health beliefs and health behaviours, and models of illness appraisal to understand illness cognitions and coping response, at both the macro (for example, social norms) and micro level (for example, attitude). Models of health behaviour and illness appraisals emerged inductively from the findings in Chapter 3, and as such are described in the context and content of the qualitative study.

This thesis takes an interdisciplinary health economic and health psychology approach to valuing preferences for care. The studies reported in this thesis are embedded in health economic theory but acknowledge emerging underlying psychological factors in understanding preferences for gynaecology cancer follow-up.

1.6 Thesis Overview

Follow-up is given to gynaecological cancer patients following treatment, with the aim to detect possible recurrence; manage any physical and psychological treatment effects, reassure patients and collect data for the purpose of research (Hall & Rustin, 2011; Rustin et al, 2010). Traditionally, follow-up appointments are delivered in secondary care for a given duration and are comprised of gynaecological examinations and possible tests, with the exact model of follow-up differing based on local protocol (Hall & Rustin, 2011; Leeson et al, 2013). This traditional model of follow-up care has a cost of approximately £9.5 million to the NHS each year (NHS improvement, 2018). There are no NICE guidelines, limited evidence and medical consensus regarding the follow-up care delivered post gynaecology cancer treatment. The efficacy of the traditional model of care has been questioned in its ability to detect recurrence in the absence of symptoms, and then increase survival following asymptomatic detection (Rustin et al., 2010), as well as meet patients' needs (Hodgkinson et al., 2007; Miller et al 2003). Recurrence of endometrial, ovarian and cervical cancer is typically symptomatic

and patients are generally seen within two weeks of symptom presentation (Fung-Kee-Fung et al., 2006; Guerts et al., 2012; Jeppesen et al., 2017; Leeson et al., 2013; Lim et al., 2004; Rustin et al., 2010; Vistad et al., 2012). There is a need for a prudent changing model of aftercare to meet patients biopsychosocial needs (Leeson et al., 2017; National Cancer Survivorship Initiative, 2013; Welsh Government and NHS Wales, 2016), with increased focus on patients preferences informing the allocation of health care resources (Mulley et al., 2012). In addition there is growing recognition of the importance of understanding informal caregivers' preferences in health care decision making given the role and impact of caregiving upon care recipient (and caregivers) outcomes (Department of Health, 2012). It is recognised that the preferences of Health Care Professionals is central to the model of care delivered (Buchanan, 1988; van Hezewijk et al., 2011). Therefore this thesis explores using mixed methods, the preferences of patients, their informal caregivers and Health Care Professionals for gynaecology cancer follow-up care, with the aim of enhancing understanding and informing policy change. An interdisciplinary health economic and health psychology approach is taken, to ensure that preferences and the underlying experiences governing these are identified and understood. Knowledge of preferences and why these preferences are held will be of value to policy makers and health care commissioners in the design and application of a changing model of gynaecology cancer follow-up care.

1.7 Thesis Aims

The aim of the thesis is to understand patients, their informal caregivers and Health Care Professionals' preferences for gynaecology cancer follow-up care.

1.7.1 Chapter 2

Primary Aim: To systematically identify, extract, appraise and explore the existing literature on patients, their informal caregiver's and Health Care Professionals preferences for gynaecological cancer follow-up care.

Secondary Aim: To identify gaps in knowledge where further research is needed, and if a prior discrete choice experiment into patients and their informal caregiver's preferences for gynaecological cancer follow-up care has not been conducted, use the attributes and attribute levels valued by patients, their informal caregivers and Health Care Professionals to aid the design of a subsequent discrete choice experiment.

1.7.2 Chapter 3

Primary Aim: To identify and explore patients and their informal caregivers' preferences for gynaecology cancer follow-up, based on their experience of this care.

Secondary Aim: To use the attributes and attribute levels identified to aid the design of a subsequent discrete choice experiment.

1.7.3 Chapter 4

Aim: To compare the attributes of gynaecology cancer follow-up generated from different methods, to inform the design of a discrete choice experiment.

1.7.4 Chapter 5

Aim: To conduct an empirical study with patients, their informal caregivers and Health care Professionals in order to identify their relative preferences and trade-offs for the different attributes that comprise gynaecology cancer follow-up care.

1.8 Dissemination

As part of this PhD four empirical chapters were written (chapters 2-5). The discrete choice experiment has been submitted for publication to the British Journal of Cancer (Chapter 5). The systematic literature review (chapter 2), qualitative study (chapter 3), methodological comparison of attribute generation (chapter 4) and discrete choice experiment (chapter 5) and are in preparation for submission for publication. The thesis protocol was presented at the European Society of Gynaecology Oncology conference in Turin, Italy in 2014. The systematic review protocol was published by the Centre for Reviews and Dissemination (2015). The qualitative study was presented in a poster at the International Congress of Behavioural Medicine in Chile, 2018. The methodological comparison of attribute generation was presented at the Health Economics Study Group meeting in Manchester, 2016. On completion of this PhD studentship, the results will be disseminated to the gynaecology oncology teams both locally and nationally.

1.8.1 Thesis as a whole

Budd, L. J., Morrison, V., Stuart, N. S. A., Leeson, S. C., Whitaker, R., Williams, N. H., Yeo, S. T., Aslam, R., & Edwards, R. T. (2014, September). *Patients' and Carers' Perspectives and Preferences for Gynaecological Cancer Follow-up After Treatment in Wales: A Health Economics Perspective*. Paper presented at the European Society of Gynaecological Oncology Conference, Turin, Italy. (Oral presentation)

Budd, L., Edwards, R. T., Whitaker, R., Stuart, N., Leeson, S., Williams, N. (2014, May). *Patients' and Carers' Perspectives on Follow-up After Treatment for Gynaecological Cancer in Wales: A Health Economics Analysis*. Paper presented at the Changing prospects for cancer, Salford, UK. (Poster presentation)

Budd, L. J., Morrison, V., Stuart, N. S. A., Leeson, S. C., Williams, N., Whitaker, R., & Edwards, R. T. (2014, April). Patient's and Carer's Perspectives and Preferences for Gynaecological Cancer Follow-Up After Treatment Care: A Health Economics Perspective. Paper presented at The Welsh Health Economic Group Meeting (WHEG), Machynlleth, UK. (Oral presentation).

Budd, L. (2014, October). *PhD student case study*. Tenovus Cancer Care Local VIP event, Bangor, UK. (Oral presentation).

1.8.2 Systematic literature review

Budd, L., Morrison, V., Seale, T., Edwards, R. T (2015). Patients, carers and health care professionals' preferences for gynaecological cancer follow-up care: a mixed method systematic review (Protocol). *PROSPERO 2015*: CRD42015016230. Available from http://www.crd.york.ac.uk/PROSPERO/display_record.asp?ID=CRD42015016230

Budd, L., Edwards, RT., Stuart, N., Leeson, S., & Morrison, V. (2015, May). *Patients' and Carers' Perspectives for Gynaecological Cancer Follow-up Care: Results from a qualitative study and systematic literature review*. Paper presented at the South Wales Gynaecology Oncology Meeting, Swansea, UK (oral presentation).

1.8.3 Qualitative study

Budd, L., Morrison, V., Stuart, N, Leeson, S., & Edwards, R. T. (2015). *Patients' and Carers' Perspectives for Gynaecological Cancer Follow-up Care: Provisional results from a qualitative study*. Paper presented at the College of Health and Behavioural Sciences postgraduate conference, Bangor, UK. (Oral presentation)

Budd, L., Morrison, V., Stuart, N., Leeson, S., Whitaker, R., Williams, N., & Edwards R. T. (2014, November). *The qualitative design of a discrete choice experiment to explore patients and their informal caregivers preferences for gynaecological cancer follow-up care: a focus group study*. Paper presented at the National Cancer Research Institute conference, Liverpool, UK. (Poster presentation)

Budd, L., Edwards, R. T., Stuart, N., Leeson, S., & Morrison, V (2015, May). *Patients' and Carers' Perspectives for Gynaecological Cancer Follow-up Care: Results from a qualitative study and systematic literature review*. Paper presented at the South Wales Gynaecology Oncology Meeting, Swansea, UK. (Oral presentation).

Timmis, L. J., Edwards, R. T., Stuart, N., & Morrison, V. (2016, March). *Patients and informal caregivers experience of gynaecology cancer follow-up care*. Paper presented at the British Psychology Oncology Society conference, Cambridge, UK. (Poster presentation).

Timmis, L., Edwards, R.T. & Morrison, V. (2015, November). *How to choose what is good for you – provisional results of analysis*. Paper presented at the Annual Gynaecological cancer education evening, Llandudno, UK. (Oral Presentation)

Timmis, L., Edwards, R. T., Stuart, N., Leeson, S., & Morrison, V. (2015, September). *Care following treatment for gynaecological cancer: What is important to BCUHB patients and their informal caregivers? (Provisional results)*. Paper presented at the Bangor University and Betsi Cadwaladr University Health Board Research showcase, Bangor, UK. (Poster presentation).

Timmis, L., Morrison, V., Stuart, N., Leeson, S., & Morrison, V. (2015, August). *Patients' and Carers' Perspectives for Gynaecological Cancer Follow-up Care: Provisional results from a qualitative study*. Paper presented at the College of Health and Behavioural Sciences Post graduate school conference, Bangor, UK. (Oral presentation)

Morrison, V., Edwards, R. T & Timmis, L. (2018). "To know that there is a light at the end of the tunnel": Patients preferences for gynaecological cancer follow-up care. Paper presented at the International Congress of Behavioral Medicine, Santiago, Chile. (Poster Presentation).

1.8.4 Methodological comparison of attribute generation

Timmis, L. J., Morrison, V., & Edwards, R. T. (2016, January). *Attribute and attribute level development in discrete choice experiments: do the methods used effect the final choice of attributes and levels?* Paper presented at the Health Economics Study Group, Manchester, UK (presentation and discussion of paper).

1.8.5 Discrete choice experiment

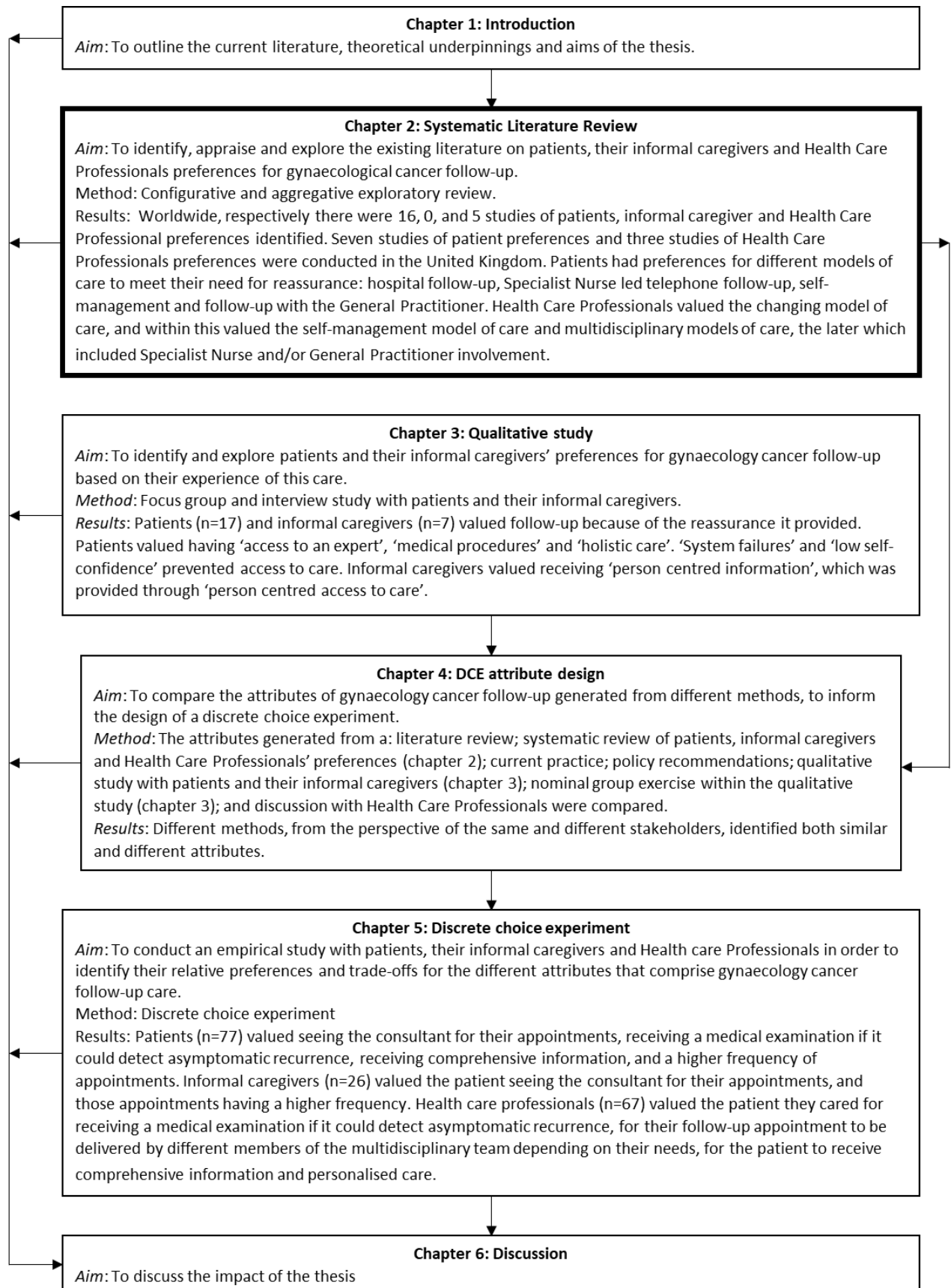
Timmis, L., Morrison, V., Yeo, S. T., Stuart, N., Leeson, S., & Edwards, R. T. (2015, September). *Gynaecological cancer follow-up preferences in BCUHB: a discrete choice experiment*. Paper presented at the Bangor University and Betsi Cadwaladr University Health Board Research showcase, Bangor, UK. (Poster presentation)

1.9 Chapter conclusion

There is a need to identify patients, their informal caregivers and Health Care Professional's preferences for gynaecology cancer follow-up, given the need for a prudent changing model of care. To understand this, it is important to first conduct a systematic literature review to identify, appraise and explore the existing literature on patients, their informal caregivers and Health Care Professionals preferences for gynaecological cancer follow-up.

Chapter 2:

**Patients, their Informal Caregivers
and Health Care Professionals’
preferences for gynaecology cancer
follow-up: a systematic literature
review**



2.1 Chapter Summary

Chapter 1 identified a need to improve outcomes of gynaecology cancer follow-up care from the perspective of both consumers and providers of care. Within this, Chapter 1 identified a need to systematically identify, appraise and explore existing research into patients, their informal caregivers and Health Care Professionals preferences for gynaecology cancer follow-up. Therefore this chapter identified all the available evidence generated between 1994 and 2014, that explored patients, their informal caregivers and Health Care Professionals preferences for gynaecology cancer follow-up (and presents this in the form of a descriptive map), and then appraises and synthesises the research conducted in the United Kingdom (and presents this in the form of a synthesis).

A systematic literature search was conducted using the following electronic databases MEDLINE via EBSCOhost; The Cochrane Collaboration Register and library; PsycINFO; Web of Science; ASSIA Applied Social Sciences Index and Abstracts, and a grey and unpublished literature search. Nineteen studies met the eligibility criteria, and eight studies were conducted in the United Kingdom and so explored within the synthesis using the framework approach. Patients believed that the aim of follow-up care was to detect recurrence and provide reassurance in the absence of disease. Patients had differing preferences for the model of care to meet their follow-up needs, valuing: hospital follow-up; Specialist Nurse led telephone follow-up, self-management, and follow-up delivered by the General Practitioner. No studies of informal caregivers' preferences were identified. Health Care Professionals felt that the aim of follow-up was to detect recurrence and monitor and treat the effects of treatment. Given the need for a changing model of care, Health Care Professionals valued the multidisciplinary and self-management models of care. The number of, content of, and methodological quality of identified studies, demonstrated a need for further high quality research (Chapter 3, 5).

2.2 Introduction

There is a need for a prudent changing model of gynaecological cancer follow-up care, to improve its effectiveness and cost-effectiveness from the perspective of the consumers (patients and their informal caregivers) and providers of the service (policy makers, health care commissioners and Health Care Professionals). See Chapter 1 for full discussion (Leeson et al., 2017; Lydon et al., 2009; Morrison et al., 2012; Mulley et al., 2012; Revenson et al., 2016; National Cancer Survivorship Initiative, 2013; van Hezewijk et al., 2011; Wales Cancer Network, 2016; Welsh Government and NHS Wales, 2016).

The literature searches conducted to frame the thesis (Chapter 1) highlighted a need to systematically identify, extract, appraise and explore the available evidence regarding patients, their informal caregivers and Health Care Professionals preferences for gynaecological cancer follow-up care. Therefore, the aim of this chapter was to systematically identify, extract, appraise and explore the existing literature on patients, their informal caregiver's and Health Care Professionals preferences for gynaecological cancer follow-up care. The specific objectives were to:

1. Identify existing literature on patients, their informal caregivers and Health Care Professionals preferences for gynaecology cancer follow-up care.
2. Critically appraise existing research conducted in the context of the United Kingdom.
3. Explore whether patients, their informal caregivers and Health Care Professionals had similar or differing preferences for gynaecological cancer follow-up.
4. Explore how patients, their informal caregivers and Health Care Professionals preferences for the model of gynaecology cancer follow-up compare to the standard model of care.
5. Explore how patients, their informal caregivers and Health Care Professionals preferences for the model of gynaecology cancer follow-up, compare to research regarding the effectiveness and cost-effectiveness of gynaecological cancer follow-up.
6. Identify whether further research is needed.
7. If required, use the attributes generated by patients, their informal caregivers and Health Care Professionals to aid the design of a subsequent discrete choice experiment (Chapter 4-5).

2.3 Methods

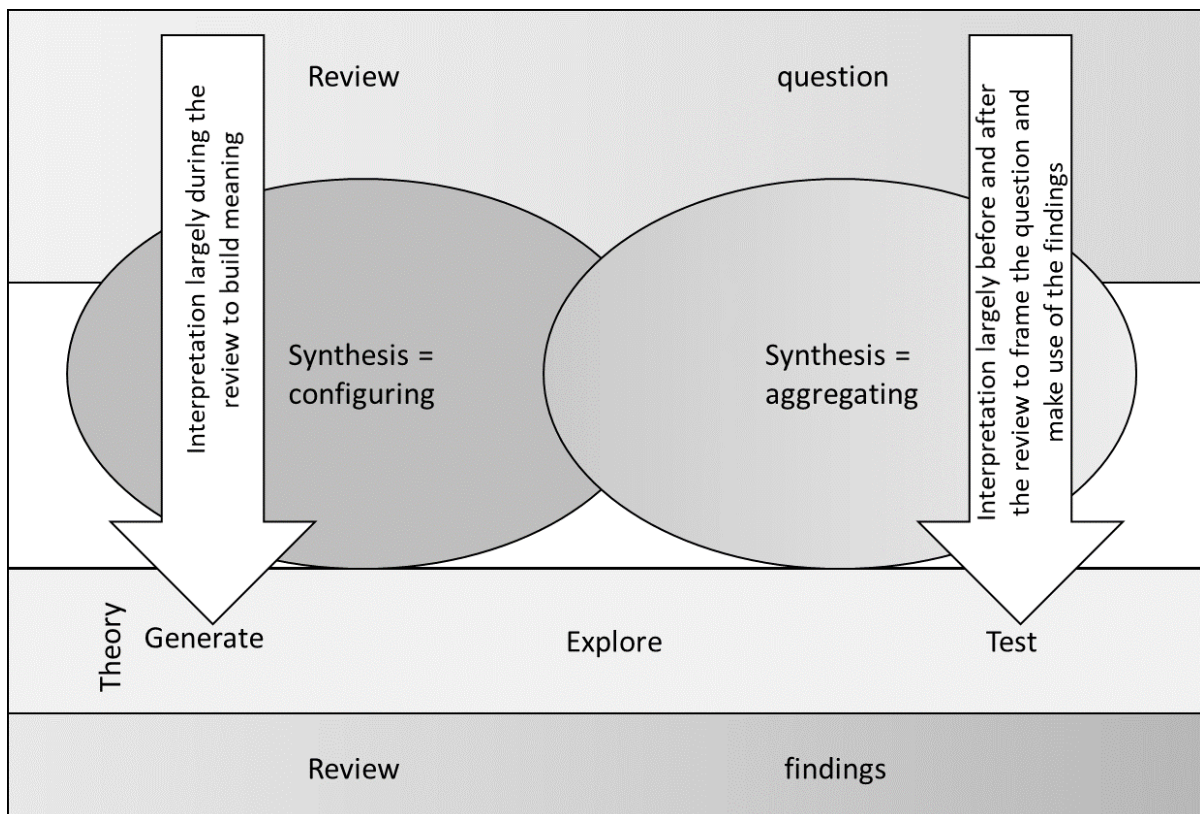
2.3.1 Protocol and registration

A systematic review protocol was published by the Centre for Reviews and Dissemination, at the University of York (Budd, Morrison, Seale & Edwards, 2015: see appendices).

2.3.2 Design

Given the overarching aim of the thesis and this systematic literature review, a mixed method review using both a configurative and aggregative perspective was conducted, following the Evidence for Policy and Practice Information (EPPI) centre guidance (EPPI-centre., 2010; Gough, Oliver & Thomas, 2012; Harden, 2010; Thomas et al., 2004: see Figure 1). This allowed for exploration both within and between each empirical study, to add to the existing knowledge base rather than merely summing the data.

Figure 1: Diagram of the configurative and aggregative continuum (Gough et al., 2012)



2.3.3 Eligibility criteria

2.3.3.1 Population

Gynaecology cancer follow-up patients, their informal caregivers and Health Care Professionals. Studies of other types of cancer follow-up patients, their informal caregivers or Health Care Professionals were excluded, unless the results were analysed separately.

2.3.3.2 Interventions

Follow-up care was defined in collaboration with SL (Consultant Gynaecologist and Oncologist) and NS (Consultant Oncologist). The definition of follow-up care was:

Care delivered from three months post-treatment, provided by the NHS and offered to all patients treated with curative intent (dependant or not dependant on risk of recurrence).

All models of follow-up care were of interest, for example, Specialist Nurse led telephone follow-up (see Chapter 1 for further discussion of models of follow-up care). Models of follow-up care (intervention or comparator) with co-interventions were excluded unless the co-intervention was part of the model. Studies which explored the experiences and preferences for the whole clinical pathway were excluded, unless the results were analysed separately in relation to patients in follow-up care, or if key themes relating to follow-up care were identified.

2.3.3.3 Outcome measures

Studies exploring process evaluation were included: preferences, perspectives, experiences, satisfaction, views, opinions, and utility, as considered by patients and/or informal caregivers and/or Health Care Professionals.

2.3.3.4 Study type

Empirical quantitative and qualitative research was included. Opinion papers, literature reviews and systematic literature reviews were excluded to reduce bias, and given the nature of the review, the need to identify and explore primary empirical data as per systematic review guidance (Gough et al., 2012).

2.3.4 Searches

2.3.4.1 Search Strategy

The search strategy followed the PICOS format (Population; intervention; comparison; outcome; study design: see table 1). The ‘population’ was gynaecological cancer patients, their informal caregivers and Health Care Professionals, the ‘intervention’ follow-up care and the ‘outcome’ preferences. The ‘comparison’ search was not used as studies comparing any model of follow-up care to any other model of follow-up care were of interest, so this criteria was not applicable. The ‘study’ search criteria was not used in accordance with systematic literature reviewing guidance (EPPI-centre, 2010).

Table 1. PICOS table

Population	AND	Intervention	AND	Outcomes
Gynaecological cancer patients, their informal caregivers and Health Care Professionals		Follow-up after treatment care		Preferences

Identifying search terms for each of these search categories was an iterative process. Numerous scoping and test searches were conducted to ensure that the results received were those that were expected. Discussion around the search terms to be used took place within the supervisory team and with support from information scientists (BH; YN). A preliminary search was undertaken using the thesaurus terms and free text terms used by previous systematic reviews of gynaecological cancer follow-up care (Aslam et al., 2016; Elit et al., 2009; Fung Kee Fung et al., 2006), follow-up care generally (Lewis et al., 2009a; Lewis et al., 2009b; Lewis et al., 2009c) and preferences (Dahl, Wittrup, Petersen, Blaakaer & Væggemose, 2015; de Bekker Grob, Ryan & Gerard, 2012; Linley, 2013; Ryan & Gerard, 2003). A pearl growing technique was then employed, whereby the thesaurus terms and free text terms (words used in the title and abstract), of research studies that met the inclusion criteria, were used in the search criteria.

Scoping searches identified that the terms used in the original search to describe the ‘intervention’, for example ‘aftercare’ and ‘posttreatment follow-up’, did not have enough sensitivity (defined as identification of as much evidence as possible: Gough et al., 2012), to

capture all studies that related to follow-up care with research studies wrongly excluded because of their poor indexing. These papers had indexed terms such as 'nurse' amongst others, which is a word used to describe an alternative model of follow-up care not follow-up care itself. Given the need to include all relevant papers, terms relating to the 'setting' of follow-up care were used to increase sensitivity of the intervention search even though this reduced specificity (defined as precision of the search: Gough et al., 2012).

The original scoping searches for the 'outcome' used search strings from previous systematic reviews of health economic preferences (De Bekker-Grob et al., 2012; Linley, 2013; Ryan & Gerard., 2003) and more psychological terms for preferences. This yielded a high number of hits with low specificity. In accordance with guidance from the information scientist (BH), the application of search term filters was used to improve the balance between sensitivity and specificity (see appendices for search terms used).

English and American spellings of words were included, and the truncation (*) was used when appropriate. The Boolean search terms AND, OR were used to connect search terms. Research papers were restricted to the English language given the resources available and the need for semantic and conceptual equivalence in translation.

2.3.4.2 Information Sources

The following electronic databases were searched in December 2014 for studies published since 1994: MEDLINE via EBSCOhost; The Cochrane Collaboration Register and library (including but not limited to: CENTRAL, NHS EED, DARE, Health Technology Assessment, The Cochrane Gynecological Cancer Review Group Trial Register, and The Cochrane Breast Cancer Review Group); PsycINFO; Web of Science; and ASSIA Applied Social Sciences Index and Abstracts. ScienceDirect would not allow for the long search string (see Appendices). The information scientist (BH) advised that searching this database was not essential given the large overlap with Web of Science, therefore it was subsequently excluded.

The search strategy was designed in PsycINFO and then translated to the appropriate MESH/thesaurus terms and formats for the other databases searched. This was an iterative process between and within databases, to ensure a balance between specificity and sensitivity.

To ensure that the search was comprehensive, any relevant research was not missed or wrongly excluded, to reduce the effect of publication bias, and to ensure that the review

included up to date but yet unpublished research, a search was also conducted of unpublished and grey literature, general search engines, the reference section of included papers, and contact was made with active researchers in the field (EPPI-centre, 2010: Table 2). The specific sources were chosen based on guidance from supervisors and information scientists.

Table 2. Grey and unpublished literature search

Type of source	Data sources
Unpublished and Grey literature	The Metaregister; The National Cancer Institute record of current clinical trials; www.greylit.org ; www.opengrey.eu ; www.base-search.net
Contacting experts	Macmillan Cancer Support; Cancer Research UK; Tenovus Cancer Care; key authors (Dahl)
Hand Search	Conference proceedings: The National Cancer Research Institute Conference; the European Society of Gynaecological Oncology Conference; the National Forum of Gynaecological Oncology Nurses. Reference list checking: of studies that met the eligibility criteria; relevant systematic reviews retrieved by the search (retrospective reference list checking).
General searches	'Google'; Google scholar'

2.3.5 Study Records

2.3.5.1 Data Management

References and abstracts identified from the final search of each database were downloaded into separate 'Refworks' files, and then collapsed into one file (EPPI-centre, 2010). Exact duplicates were removed before research studies were reviewed against the inclusion criteria. The hits that could not be excluded from each screening phase were copied to a separate file for the next screening phase. A PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) diagram was used to report the selection process in accordance with the guidance (Moher, Liberati, Tetzlaff & Altman, 2009).

2.3.5.2 Selection Process

The study selection process was piloted in a sample of retrieved papers to ensure that the inclusion/exclusion criteria could be interpreted reliably and consistently, and that it appropriately classified the studies. There were three phases of screening 1) title screening 2) abstract screening 3) full paper screening. At each phase, studies that did not meet the

inclusion criteria were excluded based on the first criteria they did not meet (EPPI-centre, 2010). The primary reviewer (LT) and secondary reviewer (TS) independently examined each study against the inclusion criteria. Any disagreements were discussed in accordance with the protocol, and when no decision could be reached were discussed with the review team (RTE, VM), and a majority decision made.

2.3.5.3 Data Extraction

A standardised data extraction form designed for the purpose of the descriptive map was completed for each study that met the inclusion criteria. This data extraction form collected information on the country of study; study aims that were relevant to this review; participants; research methods; model of follow-up care explored (for qualitative studies this was the content of the interview guide; for questionnaire studies this was the content of the questions; for intervention studies this was the content of the intervention).

A more comprehensive standardised data extraction form was completed for eligible studies conducted in the United Kingdom for the purpose of the synthesis. Synthesis of studies conducted in the United Kingdom ensured the thesis had contextualisation to the population of interest and its specific health care system, with models of care delivered across the world being very heterogeneous (Vistad et al., 2012). Different data extraction forms were devised for different study types. Data extraction forms were based on the guidance published by Cochrane, the Joanna Briggs Institute, the EPPI Centre (Gough et al., 2012; Noyes & Lewin, 2011; The Joanna Briggs Institute, 2014) and previous mixed method systematic reviews (Bray, 2015). Full papers of multiple reports of a single study were extracted and collated to ensure that each study, as opposed to each paper, was the unit of interest in the review. The data extraction forms were piloted to ensure that it collected all required information and could be interpreted reliably and consistently. The data was extracted by LT, and independently checked for accuracy by the secondary reviewer (TS). Any disagreements were resolved through discussion.

2.3.5.4 Risk of bias in individual studies

Critical appraisal allows for systematic assessment of the risk of bias in included studies. This enables the trustworthiness and relevance of individual studies to shape the conclusions of the systematic literature review (EPPI centre, 2010). The Weight of Evidence Framework was

used to critically appraise studies that met the inclusion criteria (Gough, 2007). For dimension one (soundness of studies) the appropriate CASP (critical appraisal skills programme) tool was used (Gough et al, 2012; <https://casp-uk.net/>). At the time of the review there was no CASP tool to evaluate questionnaire studies. Therefore, a critical appraisal tool developed for a previous health service systematic literature review was used to critically appraise questionnaire studies (Greenhalgh, Robert, MacFarlane, Bate & Kyriakidou, 2005). The second dimension (the appropriateness of the methodology to answer the systematic review research questions), and third dimension (relevance of the individual study to the review), were assessed in accordance with the aims and objectives of the systematic review. Each dimension was assessed as being of high, medium or low quality before an overall judgement of the whole study was made (see Table 3: Torgeson et al., 2008). Studies were not excluded from the synthesis based on the overall judgement as it was important to create a picture of the current field of research (Gough et al., 2012). The data was assessed for risk of bias by LT, and verified by TS. Any disagreements were resolved through discussion. Studies met the criteria of high or medium if they met all the criteria for that valuation.

2.3.6 Data Analysis

There were two outcomes of the systematic review in accordance with the aim of the review, 1) a descriptive map 2) a synthesis of the literature conducted in the United Kingdom.

The purpose of the descriptive map was to describe the literature on patients, informal caregivers and Health Care Professionals' preferences for gynaecological cancer follow-up care. Descriptive maps provide a methodical summary of the research in the specific field; aid in the identification of gaps in the research field which need future research attention; and help to provide a context for interpreting the results from the synthesis (EPPI-centre, 2010). The descriptive map was used to aid the design of the discrete choice experiment (see chapters 4-5).

A synthesis of the literature conducted in the United Kingdom was then explored. The original aim was to conduct a synthesis of all studies identified and discussed in the descriptive map. However, data extraction identified large variability in the model of standard care delivered between countries. It is known that experiences of past care influence preferences for future care (Ajzen, 1985; Ajzen, 1991; Morrison et al., 2012; Salani 2013; Salkeld, Ryan &

Table 3. Weight of evidence framework

	Qualitative research	Cross-sectional surveys	Intervention evaluation
WoE A: Soundness of studies	High: Methods and results are transparent and detailed for data collection and analysis of the data, and the interpretation of the findings is justified by the results. Choice of method of analysis justified. Medium: Methods and results are reported satisfactorily for data collection and analysis of the data, and the interpretation of findings is partly justified by the results. Low: Methods and results are reported poorly for data collection and analysis of the data, and the interpretation of findings is not justified by the results.	High: Methods and results are transparent and detailed for data collection and analysis of the data, and the interpretation of the findings is justified by the results. Sample size calculation, and met. Medium: Methods and results are reported satisfactorily for data collection and analysis of the data, and the interpretation of findings is partly justified by the results. Low: Methods and results are reported poorly for data collection and analysis of the data, and the interpretation of findings is not justified by the results.	High: Methods and results are transparent and detailed for data collection and analysis of the data, and the interpretation of the findings is justified by the results. Sample size calculation, and met. Medium: Methods and results are reported satisfactorily for data collection and analysis of the data, and the interpretation of findings is partly justified by the results. Low: Methods and results are reported poorly for data collection and analysis of the data, and the interpretation of findings is not justified by the results.
WoE B: Appropriateness of study design and analysis for answering the review question	High: Focus group or interviews with participants of varied demographics, reporting that data saturation had been reached. Exploration of why preferences for care differed by different demographics. Medium: Focus groups or interviews with participants of varied demographics, reporting that data saturation had been reached. Low: Focus groups or interviews. No reporting of key demographics and/or data saturation.	High: Surveys using validated questionnaires. Exploration of why preferences for care differed by different demographics. Medium: Surveys analysed using validated questionnaires, or questionnaires based on previous research and piloted. Reporting on key demographics. Low: Surveys using non validated questionnaires. No reporting of key demographics.	High: Randomised control trial with clearly reported intervention, using validated measures at multiple time points to evaluate the intervention. Exploration of why preferences for care differed by different demographics. Medium: Non-randomised trial or repeated measures design using measures at multiple time points to evaluate the interventions. Reporting of key demographics. Low: Non-randomised trial or evaluation.
WoE C: Relevance of the study focus to the review (topic, sample, population, measures, overall evidence focus)	High: Exploration of preferences for gynaecology cancer follow-up, for the standard or changing model of care using an inductive approach. The sample has varied demographics, and these are explored in	High: Exploration of preferences for gynaecology cancer follow-up, for the standard or changing model of care using relevant and thorough measures. The sample has varied demographics, and these	High: Exploration of preferences for gynaecology cancer follow-up, for the standard or changing model of care, with a detailed description of the intervention and relevant and through measures used to evaluate this. The sample has varied

	<p>relation to preferences for care.</p> <p>Medium: Exploration for sub-group of gynaecology cancer follow-up, for example, preferences for ovarian cancer follow-up, using an inductive approach. Sample has varied demographics.</p> <p>Low: Exploration for preferences for gynaecology cancer follow-up generally or as a sub-group.</p>	<p>are explored in relation to preferences for care.</p> <p>Medium: Exploration for sub-group of gynaecology cancer follow-up, for example, preferences for ovarian cancer follow-up, using relevant and thorough measures. Sample has varied demographics.</p> <p>Low: Exploration for preferences for gynaecology cancer follow-up generally or as a sub-group.</p>	<p>demographics, and these are explored in relation to preferences for care.</p> <p>Medium: Exploration for sub-group of gynaecology cancer follow-up, for example, preferences for ovarian cancer follow-up with a description of the intervention and relevant measures used to evaluate this. Sample has varied demographics.</p> <p>Low: Exploration for preferences for gynaecology cancer follow-up generally or as a sub-group.</p>
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Short, 2000). Therefore it was decided to limit the synthesis to studies conducted in the United Kingdom, to inform the wider discussion of the thesis.

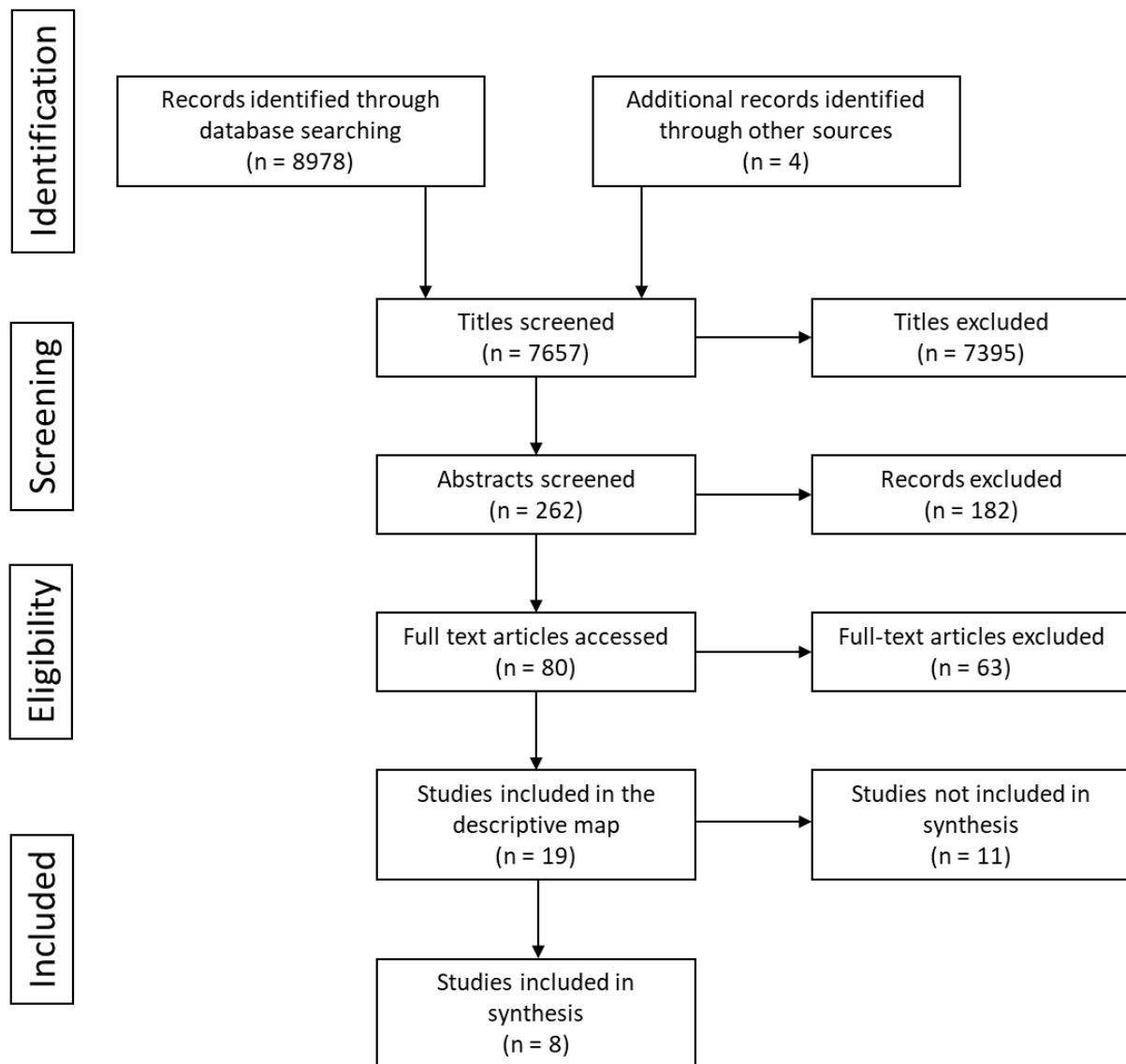
The synthesis was conducted in accordance with the aims and objectives of the systematic review. Data were analysed by stakeholder (patient, Health Care Professional) using framework analysis as it allowed for an inductive and deductive approach to synthesis, and to explore the data using a configurative and aggregative approach (Gough et al., 2012; Richie & Spencer, 1994). The primary research papers were read and re-read, and data that was relevant to the aims and objectives of the systematic review were extracted. The extraction of qualitative data was more of an iterative process, with the content and wording of the theme list amended with these iterations.

A comparative synthesis compared 1) the results by stakeholder, 2) the results to research into the effectiveness and cost-effectiveness of gynaecology cancer follow-up.

2.4 Results

The results from the searches and screening are reported in Figure 2 using the PRISMA guidance (Moher et al., 2009). Given the poor indexing of the intervention terms (as described in the methodology), there were a large number of records identified.

Figure 2: PRISMA diagram



Fourteen studies identified the preferences of patients (Bradley et al., 1999; Bradley et al., 2000; Brothers et al., 2013; Cox et al., 2008; Greimel et al., 2011; Howells, Bradley, Pitts, Calvert & Redman, 1999; Miller et al., 2003; Nicolaije, et al., 2012; Nicolaije, et al, 2013; Oshima et al., 2011, Oshima, Kisa, Terashita, Kawabata & Maezawa, 2013; Palmer, Pratt, Basu & Earl, 2006; Sekse et al., 2010; Stewart, Duff, Wong, Melancon & Cheung, 2000; Suprasat & Manopunya, 2011). No studies identified the preferences of informal caregivers. Three studies identified the preferences of Health Care Professionals (Dahl, et al, 2015; Penney, Kitchener & Templeton, 1995; Vistad et al., 2012). Two explored the preferences of patients and Health Care Professionals (Kew, Galaal, Manderville & Verleye, 2007/Kew et al., 2009; Lydon et al., 2009).

2.4.1 Descriptive map of patients preferences (n=16/19)

Of the sixteen studies that explored the preferences of patients, eight studies aimed to explore the preferences for gynaecology cancer follow-up generally (Brothers et al., 2013; Greimel et al., 2011; Howells et al., 1999; Kew et al., 2007/Kew et al., 2009; Miller et al., 2003; Oshima et al., 2011/2013; Sekse et al., 2010; Suprasert & Manopunya, 2011), two explored the preferences for endometrial cancer follow-up generally (Nicolaije et al., 2013; Nicolaije et al., 2012), four explored the preferences for ovarian cancer follow-up generally (Cox et al., 2008; Lydon et al., 2009; Palmer et al., 2006; Stewart et al., 2000), two explored the preferences for low stage endometrial cancer follow-up (Bradley et al., 2000), and one explored the preferences for low stage gynaecology cancer follow-up (Bradley et al., 1999: see Table 4).

Of these, two were intervention studies (Cox et al., 2008; Brothers et al., 2013), one an audit and evaluation study (Palmer et al., 2006), nine were cross-sectional studies (Bradley et al., 2000; Greimel et al., 2011; Howells et al., 1999; Kew et al., 2007/Kew et al., 2009; Miller et al., 2003; Nicolaije et al., 2013; Nicolaije et al., 2012; Stewart et al., 2000; Suprasert & Manopunya, 2011), two were focus group studies (Lydon et al., 2009; Oshima et al., 2011/Oshima et al., 2013), and two were interview studies (Bradley et al., 1999; Sekse et al., 2010).

Seven studies were conducted in the United Kingdom (Bradley et al., 1999; Bradley et al., 2000; Cox et al., 2008; Howells et al., 1999; Kew et al., 2007/Kew et al., 2009; Lydon et al., 2009; Palmer et al., 2006), three in the America (Brothers et al., 2013; Miller et al., 2003; Stewart et al., 2000), one in Japan (Oshima et al., 2011/Oshima et al., 2013), one in Thailand (Suprasert & Manopunya, 2011), one in Austria (Greimel et al., 2011), one in Norway (Sekse et al., 2010) and two in the Netherlands (Nicolaije et al., 2012; Nicolaije et al., 2013).

Studies explored heterogeneous models of standard and alternative models of care, using inductive and deductive approaches. The attributes of care explored are detailed below in Table 5 (taken from Table 4 and summed).

Table 4. Descriptive map of patients preferences for gynaecology cancer follow-up (n=16/19)

Author, year of publication, Country	Study aims relevant to this review	Participants	Research methods	Model of follow-up care explored (for qualitative studies, content of interview guide; for quantitative questionnaire study content of measures/questions; for intervention study content of intervention)
Bradley et al (1999), UK	Investigate why some patients need follow-up care when there is no obvious clinical benefit, and to identify the advantages and disadvantages of follow-up care.	N=12 Treatment completed in previous 6-72 months Type of cancer: not reported Stage of disease: diagnosed with stage I-II disease (inclusion criteria) Treatment received: not reported Any co-morbidities: not reported.	Interview study	Preferences for follow-up care and discharge from care, feelings before clinic visit, coping with recovery, sexual recovery.
Bradley et al (2000), UK	Identify whether patients diagnosed with early stage gynaecological cancer want to continue to receive follow-up or be discharged from care	*N=54 Type of cancer: n=14/53 endometrial cancer patients, n=17/53 cervical cancer patients, n=17/53 ovarian cancer patients, n=5/53 vulvar cancer patients Stage of disease: not reported Treatment received: primary treatment not adjuvant therapy (inclusion criteria) Follow-up: n=30/53 <18 months, n=23/53 >18 months	Cross sectional study	Standard care, discharge from hospital follow-up care with re-entry if health concern

		Any co-morbidities: n=8/48 other cancer (current or treated), n=40/48 no other cancer (missing data not reported)		
Brothers et al (2013), USA	To explore whether a survivorship care plan improves perception of quality of care	<p>N=121</p> <p>Type of cancer: n=65 (54%) endometrial cancer patients, n=35 (29%) ovarian cancer patients, n=16 (13%) cervical cancer patients, n=5 (4%) vaginal cancer diagnosis.</p> <p>Stage of disease: not reported</p> <p>Treatment received: not reported</p> <p>Any co-morbidities: not reported.</p> <p>Duration in follow-up: All participants in first year of follow-up (part of inclusion criteria).</p>	Randomised cross-sectional control trial (randomised by Health Care Professional) comparing standard care and a survivorship care plan with standard care	<p>Survivorship care plan: Treatment summary, care plan based on treatment summary, and copy of test results.</p> <p>Standard care: physician led secondary care appointment, medical examination, monitor signs of recurrence and manage treatment related morbidities, referrals to other Health Care Professionals if required, nurses and/or patient resource centre provide patient education information.</p>
Cox et al (2008), UK	Evaluate holistic nurse led telephone follow-up for ovarian cancer patients, to identify 1) satisfaction with telephone follow-up, 2) reported benefits to telephone follow-up.	<p>N=46 completed measures</p> <p>Type of cancer: 49% diagnosed with stage 1 or 2 disease, 51% diagnosed with stage 3 or 4 disease.</p> <p>Stage of disease: not reported</p> <p>Treatment received: not reported</p> <p>Any co-morbidities: not reported.</p> <p>Duration in follow-up: not reported.</p>	Repeated measures intervention study	Intervention: 3 monthly telephone appointment with the Specialist Nurse to give blood test results (taken 2 weeks previously at a local clinic) and discussion of result implications (if any), discussion of any symptoms (with referral to the clinic if required); tailored biopsychosocial support and information. Onward referrals if

				<p>necessary and information about local support networks</p> <p>Preference measures: experience, satisfaction and advantages to this model of follow-up</p>
Greimel et al (2011), Austria	To identify patients' views of follow-up care, and anxiety and distress experienced before follow-up appointment.	<p>N=210 (but data reported for n=200)</p> <p>Cancer type: n= 64 (32%) ovarian cancer patients; n= 56 (28%) endometrial cancer patients; n= 47 (24%) cervical cancer patients; n= 23 (12%) vulva cancer patients; n= 10 (5%) other cancer.</p> <p>Stage of disease: n= 103 (52%) diagnosed with stage 1 disease; n= 34 (17%) diagnosed with stage 2 disease; n= 52(26%) diagnosed with stage 3 disease; n= 11 (6%) diagnosed with stage 4 disease.</p> <p>Treatment received†: n= 186 (93%) had surgery; n=83 (42%) had adjuvant chemotherapy; n=28 (14%) had adjuvant radiotherapy; n=17(8.5%) had concomitant radio-chemotherapy.</p> <p>Length of time since initial diagnosis: n=11 (6%) 6months – 2years since diagnosed; n= 48 (24%) 2 – 5 years since diagnosed; n=114(57%) 5 – 10 years since diagnosed, n= 27 (14%) 10> years since diagnosed.</p>	Cross-sectional study.	<p>Frequency of appointments</p> <p>Content of appointment (consultation itself, physical examination, blood test/tumour marker, radiology tests, ultrasound, chest x-ray), content of discussions with Health Care Professional during follow-up period (causes of cancer; cancer prevention; immune system function; nutrition; physical weakness/fatigue; sexual issues; rehabilitation and social service support; psychosocial support; complimentary therapies)</p> <p>Satisfaction with model of follow-up care</p>

		Comorbidities: not reported		
Howells et al (1999), UK	Identify patients' opinions for continued follow-up or discharge from hospital follow-up	N=90 Type of cancer: n=23 ovarian cancer, n=37 cervical cancer, n=19 uterus cancer, n=11 vulvar cancer Stage of disease: not reported Treatment received: not reported Duration in follow-up care: mean 30 months (range 1-5 years)	Cross-sectional study	Views and preferences for hospital follow-up, follow-up with General Practitioner, patient led free access (self-management)
Kew et al (2009) Kew et al (2007), UK	To identify understanding and views on the current model of care and perception of the specialist nurses role in follow-up	N=96 patients Cancer type: n= 31 (32%) ovarian cancer, n=14 (14%) endometrial cancer, n=23 (24%) cervical cancer, n= 9 (9%) vulva cancer, n= 1 (1%) vaginal cancer, n=18 (19%) missing data. Stage of disease: not reported Treatment received†: n=90 (94%) surgery and adjuvant therapy, n=22 (23%) chemotherapy, n=7 (7%) radiotherapy, n=1 (1%) missing data. Any comorbidities: not reported Duration in follow-up: not reported	Cross-sectional study	Frequency of care Health care professional seen for appointments (hospital doctor, specialist nurse, General Practitioner, other) Content of care (consultation, examination, blood tests, Health Care Professional listen to concerns and answer questions)
Lydon et al (2009), UK	Identify ovarian cancer patients and health care professionals preferences for care	Patients (n=6, one focus group) Stage of disease: not reported Treatment received: not reported Any comorbidities: not reported Duration in follow-up care: not reported	Focus group	Perspective of current and alternative (Specialist Nurse led, General Practitioner led, telephone delivered) models of follow-up care, clinic waiting time
Miller et al (2003), USA	Views of the physicians' role in	N=95	Cross sectional study	Preferences for physicians to meet their psychosocial needs

	meeting follow-up needs identified	<p>Type of cancer: n=47 (50%) cervical cancer; n=28 (30%) endometrial cancer; n=11 (12%) ovarian cancer; n=6 (6%) vulval cancer; n=3 (3%) other</p> <p>Stage of disease: not reported</p> <p>Type of treatment†: n=70 (74%) surgery; n=44 (46%) radiotherapy; n=18 (19%) chemotherapy</p> <p>Any comorbidities: not reported</p> <p>Duration in follow-up care: not reported (had to be at least 6 months post treatment to take part)</p>		
Nicolaije et al (2013), The Netherlands	Explore patients preferences for the model of care received	<p>N=582 endometrial cancer patients</p> <p>Stage of disease: n=88 (15%) stage 1A; n=300 (52%) stage 1B; n=155 (27%) stage 1c; n=20 (3%) stage 2A; n=19 (3%) stage 2B.</p> <p>Type of treatment: n=582 surgery; 22% then had radiotherapy, <1% then had chemotherapy</p> <p>Any comorbidities: n=124 (21%) none; n=149 (26%) one; n=309 (53%) two or more.</p> <p>Duration in follow-up care: n=80 in 1st year of follow-up; n=103 in 2nd year of follow-up; n=210 in 3rd to 5th year of follow-up; n=189 in the 6th to 10th year of follow-up.</p>	Cross-sectional study	Preferences for frequency of appointments

Nicolaije et al (2012), The Netherlands	Identify endometrial cancer follow-up patients' level of information received and satisfaction with this, and identify any association between these preferences and patients' socio demographic and clinical characteristics.	N=742 Stage of disease: 92% stage 1; 8% stage 2 Treatment received: 76% surgery only; 23% surgery and radiotherapy; 1% surgery and chemotherapy. Any comorbidities: 20% none; 26% one; 54% two or more. Years since diagnosis: mean 4.9 years (SD 2.5 years), 13% < 2 years; 41% 2-5 years; 45% > 5 years	Cross-sectional study	Information received and satisfaction with this
Oshima et al (2011) Oshima et al (2013), Japan	To explore patients' perceptions and preferences for follow-up care, and their care seeking behaviour during the follow-up period	N=28 Type of cancer: n=9 cervical cancer; n=11 endometrial cancer; n=7 ovarian cancer; n=1 vulva cancer Stage of cancer: not reported Treatment received: n=12 surgery alone; n=15 surgery and chemotherapy; n=1 surgery, chemotherapy and radiotherapy. Time since completed treatment: n=5 1-2 years; n=3 2-5 years; n=3 3-4 years; n=6 4-5 years; n=1 5-6 years; n=4 6-7 years; n=3 7-8 years; n=0 8-9 years; n=3 9-10 years Any co-morbidities: not reported	Focus group study	The purpose, experience of and perspectives and expectations of follow-up care. Current model of care was comprised of frequent appointments in secondary care over a given duration, comprised of pelvic examinations, multiple diagnostic tests and possible onward referrals.
Palmer et al (2006), UK	To evaluate the use of CA125 testing in ovarian cancer	Questionnaire 1: N=90 Stage of cancer: not reported	Audit	Patients received CA125 testing at clinic appointment.

	<p>follow-up and dependant on patients' preferences instigate a change in practice at this cancer centre</p>	<p>Treatment received: not reported Any co-morbidities: not reported Time since diagnosis: n=31 (35%) <1 year; n=21 (23%) 12-18 months; n=12 (13%) 19-24 months; n=4 (5%) 25-36 months; n=20 (22%) >3 years; n=2 (2%) missing data Questionnaire 2: N=26 Stage of cancer: not reported Treatment received: not reported Any co-morbidities: not reported Time since diagnosis: n=4 (15%) 25-36 months; n=22 (85%) >3 years</p>	<ul style="list-style-type: none"> • If results elevated and patient symptomatic, patients scheduled earlier follow-up appointment • If CA125 elevated and patient asymptomatic or CA125 not elevated, patients received their results at their next follow-up appointment or over the telephone <i>if</i> the patient telephoned. <p>Questionnaire 1: Understanding of CA125 testing, satisfaction with current CA125 testing location; preference for result to be available at appointment; acceptability of hypothetical attendance at GP surgery 2 weeks prior to follow-up appointment for CA125 testing; any further comments.</p> <p>Change in practice so CA125 testing pre scheduled appointment, and results given in follow-up appointment.</p>
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				Questionnaire 2: understanding of CA125 testing; were the CA125 results were available at clinic follow-up appointment; has follow-up been improved by this result being available at follow-up appointment; how could follow-up be improved further?
Suprasert & Manopunya (2011), Thailand	To identify the burden of attending gynaecology cancer follow-up at a tertiary hospital	N=200 Type of cancer: n=126 (63%) cervical cancer; n=29 (15%) ovarian cancer; n=32 (16%) endometrial cancer; n=3 vulval cancer; n=10 (% not given) other Stage of cancer: not reported Treatment received: no reported Duration in follow-up care: not reported Any comorbidities: not reported	Cross sectional study	Cost of attending the follow appointment; travel time; clinic waiting time; preferences to attend follow-up at local provincial hospital
Sekse et al (2010), Norway	To understand gynaecology cancer survivors experiences of care five years post treatment	N=16 Type of cancer: n=11 endometrial cancer; n=3 ovarian cancer; n=2 cervical cancer. Stage of cancer: n=13 stage 1; n=1 stage 2; n=2 stage 3 Treatment received: n=11 surgery only; n=2 surgery and radiotherapy; n=3 surgery and radiotherapy Duration in follow-up care: post discharge (care delivered for 5 years post	Repeated measures interview study (12 months apart)	Experience of 5 year follow-up care at the hospital outpatient clinic

		treatment and to be eligible had to be 5 years post treatment)		
Stewart et al (2000), USA	To identify the information and decision making preferences of ovarian cancer patients	N=61 in follow-up period Demographics reported by total sample not split by patients in treatment and follow-up phases	Cross-sectional study	Focus of, amount and type of information received
† more than one response per participant, so % > 100% Where % is not reported, % has not been reported in the primary paper N.B: only information relevant to the aims of the systematic review were extracted. For example, aims and questionnaire items not relating to follow-up care were not extracted.				

Table 5. Attributes identified from the descriptive map (patients, n=16/19)

Health Care Professionals that delivers care	<ul style="list-style-type: none"> • Standard Consultant/hospital Doctor led care (Bradley et al., 1999; Bradley et al., 2000; Brothers et al., 2013; Howells et al., 1999; Lydon et al., 2009; Sekse et al., 2010; Kew et al., 2007/Kew et al., 2009) • Specialist Nurse led care (Cox et al., 2008; Lydon et al., 2009; Kew et al., 2007/Kew et al., 2009) • Telephone follow-up (Cox et al., 2008; Lydon et al., 2009) • General Practitioner led care (Howells et al., 1999; Kew et al., 2007/Kew et al., 2009; Lydon et al., 2009;) • Self-management (Bradley et al., 1999; Bradley et al., 2000; Howells et al., 1999)
Content of Care	<ul style="list-style-type: none"> • Consultation itself (Greimel et al., 2011; Kew et al., 2007/Kew et al., 2009) • Health Care Professional listen to concerns (Kew et al., 2007/Kew et al., 2009) • Health Care Professional answers questions (Kew et al., 2007/Kew et al., 2009) • Holistic discussion with Health Care Professional (Greimel et al., 2011) • Survivorship care plan (Brothers et al., 2013) • Medical examination (Brothers et al., 2013; Greimel et al., 2011; Kew et al., 2007/Kew et al., 2009; Oshima et al., 2011/2013) • Diagnostic tests (Cox et al., 2008; Greimel et al., 2011; Kew et al., 2007/Kew et al., 2009; Oshima et al., 2011; Oshima et al., 2013; Palmer et al., 2006) • Location of CA125 testing (Cox et al., 2008; Palmer et al., 2006). • Delivery of results from diagnostic tests (Cox et al., 2008; Palmer et al., 2006) • Monitor signs of recurrence (Brothers et al., 2013; Cox et al., 2008) • Manage treatment related morbidities (Brothers et al., 2013) • Onward referrals (Brothers et al., 2013; Cox et al., 2008; Oshima et al., 2011; Oshima et al., 2013) • Information received (Brothers et al., 2013; Cox et al., 2008; Nicolaije et al., 2012; Stewart et al., 2000) • Holistic care (Cox et al., 2008; Miller et al., 2003)
Frequency of appointments	Cox et al., 2008; Greimel et al., 2011; Kew et al., 2007/Kew et al., 2009; Nicolaije et al., 2013; Oshima et al., 2011; Oshima et al., 2013
Financial cost of attending appointment	Suprasert & Manopunya., 2011
Travel time	Suprasert & Manopunya., 2011

Preferences for care at provincial hospital	Suprasert & Manopunya., 2011
Clinic waiting time	Suprasert & Manopunya., 2011; Lydon et al., 2009

2.4.2 Descriptive map of Health Care Professionals' preferences (n=5/19)

Of the five studies that explored the preferences of Health Care Professionals, all studies explored the preferences of Doctors (including Consultants and Registrars: Dahl et al., 2015; Kew et al., 2007; Lydon et al., 2009; Penney et al., 1995; Vistad et al., 2012), four studies explored the preferences of Nurses (Dahl et al., 2015; Kew et al., 2007; Lydon et al., 2009; Vistad et al., 2012), and one study explored the preferences of primary Health Care Professionals (Vistad et al., 2012: see Table 6).

Two studies aimed to explore the preferences for gynaecology cancer follow-up generally (Kew et al., 2007; Vistad et al., 2012), one explored the preferences for ovarian cancer follow-up (Lydon et al., 2009), one explored the preferences for vulvar cancer follow-up (Penney et al., 1995), and one explored the preferences for low stage gynaecology cancer follow-up (Dahl et al., 2015).

Three studies were conducted in the United Kingdom (Kew et al., 2007; Lydon et al., 2009; Penney et al., 1995), one in a sample of Danish participants (Dahl et al., 2015), and one in a sample of participants from across Europe (Vistad et al., 2012).

A range of attributes were explored in the descriptive map, using inductive and deductive approaches. Attributes explored in these studies are described in Table 7 (taken from Table 6 and summed). It must be noted that the studies explored the provision of care by different Health Care Professionals (for example, consultant, specialist nurse, GP), but this was not regarding these different Health Care Professionals working together to provide care (multidisciplinary care).

Attributes identified from studies included in the descriptive map of patients and their Health Care Professionals preferences for care, were used to aid design of the subsequent discrete choice experiment (see Chapter 4).

Table 6. Descriptive map of Health Care Professionals preferences for gynaecology cancer follow-up (n=5/19)

Study: author, year of publication, country	Study aims that are relevant to this review	Participants	Research methods	Model of follow-up care explored (for qualitative studies, content of interview guide; for quantitative questionnaire study content of questions; for intervention study content of intervention)
Kew et al (2007), UK	To identify understanding and views on the current model of care and perception of the specialist nurses role in follow-up	N=32 Health Care Professional's (results reported for n=34) Health Care Professional speciality: n=29 gynaecological oncologists, n=3 medical oncologists, n=1 clinical oncologists, n=1 gynaecologist Health Care Professionals profession: n=22 consultants, n=4 sub-speciality trainees, n=1 associate specialist, n=5 specialist registrars, n=2 cancer nurse specialist	Cross sectional quantitative questionnaire designed for the purpose of this study	Frequency of care Health care professional seen for appointments (hospital Doctor, Specialist Nurse, General Practitioner, other) Content of care (consultation, examination, blood tests, Health Care Professional listen to concerns and answer questions)
Lydon et al (2009), UK	Identify ovarian cancer patients and health care professionals preferences for care	Health care professionals (n=7 working in medical oncology, one focus group) Professional status: n=1 consultant oncologist, n=2 senior registrars, n=1 junior registrar, n=2 research registrars, n=1 nurse clinician.	Cross sectional focus group study	Current approach to the delivery of follow-up care (consultation, examination, diagnostic tests), and perspective on alternative models of follow-up (specialist nurse/ General Practitioner led/ involvement, telephone appointments)
Penney et al (1995), UK	To identify Health Care Professional's preferences for managing vulva cancer	N=78 consultant gynaecologists involved in the management of vulva cancer.	Cross sectional questionnaire designed for the purpose of this study	Frequent follow-up over a long duration; location of follow-up appointments

Dahl et al (2015), Denmark	To explore the experiences and views of health care professionals delivering follow-up care to low stage gynaecology cancer patients	N=6 Health Care Professional's Profession: n=3 Doctors, n=3 nurses	Cross sectional focus group	Existing model of follow-up care (frequent checks in the first year, and reducing frequency for a total duration of 3-5 years diagnosis dependant; content – frequent objective physical examination which dependant on diagnosis may include an ultrasound, patients being asked about their wellbeing and given their next follow-up appointment; Life after cancer; the future of follow-up care (nurse involvement, holistic care)
Vistad et al (2012), Europe (conducted in Norway)	Explore European experts views of gynaecology cancer follow-up care	N=375 Profession: n=348 (93%) Doctor in hospital setting; n=72 (19%) Gynaecologist working in private practice; n=1(0%) Specialist Nurse; n=18 (5%) General Practitioner	Cross sectional questionnaire designed for the purpose of this study	Views of the evidence available for the model of care delivered (the model of care delivered had large variance in frequency, duration and content of care), and views for the delivery of follow-up in primary care for all or subgroup(s) of patients
Where % is not reported, % has not been reported in the primary paper N.B: only information relevant to the aims of the systematic review were extracted. For example, aims and questionnaire items not relating to follow-up care were not extracted.				

Table 7. Attributes identified from the descriptive map (Health Care Professionals, n=5/19)

Health Care Professional seen	<ul style="list-style-type: none"> • Hospital Doctor (Kew et al., 2007; Vistad et al., 2012) • Specialist Nurse (Kew et al., 2007; Lydon et al., 2009; Vistad et al., 2012) • Nurses (Lydon et al., 2009) • General Practitioner (Kew et al., 2007; Lydon et al., 2009; Vistad et al., 2012) • Self-management (Lydon et al., 2009) • Telephone appointments (Lydon et al., 2009)
Content of care	<ul style="list-style-type: none"> • Consultation (Kew et al., 2007; Lydon et al., 2009) • Examination (Dahl et al., 2015; Kew et al., 2007; Lydon et al., 2009; Vistad et al., 2012) • Diagnostic tests (Dahl et al., 2015; Kew et al., 2007; Lydon et al., 2009; Vistad et al., 2012) • Health Care Professional listens to concerns (Kew et al., 2007) • Health Care Professional answers questions (Kew et al., 2007) • Holistic care (Dahl et al., 2015)
Telephone appointments	Lydon et al., 2009
Frequency of appointments	Dahl et al., 2015; Kew et al., 2007; Penney et al., 1995; Vistad et al., 2012;
Duration of care	Dahl et al., 2015; Vistad et al., 2012;
Location of appointments	Penney et al., 1995

2.4.3 A synthesis of the research conducted in the United Kingdom

The research studies identified in the descriptive map and conducted in the United Kingdom (n=8) were further explored using framework analysis (Richie & Spencer, 1994). Five studies were conducted in a population of patients (Bradley et al., 1999; Bradley et al., 2000; Cox et al., 2008; Howells et al., 1999; Palmer et al., 2006), one in a population of Health Care Professionals (Penney et al., 1995), and two in a population of patients and Health Care Professionals (Kew et al., 2007/Kew et al., 2009; Lydon et al., 2009).

Four of the studies of patients' preferences were quantitative in nature (Bradley et al., 2000; Howells et al., 1999; Kew et al., 2007/Kew et al., 2009; Palmer et al., 2006), one an intervention study (Cox et al., 2008) and two qualitative in nature (Lydon et al., 2009; Bradley et al., 1999). Two of the studies of Health Care Professionals preferences were quantitative in

nature (Penney et al., 1995; Kew et al., 2007), and one qualitative in nature (Lydon et al., 2009).

2.4.3.1 Synthesis of patients preferences (n=7/8)

Four studies explored preferences for follow-up across different types of gynaecological cancer (Bradley et al., 1999; Bradley et al., 2000; Howells et al., 1999; Kew et al., 2007/Kew et al., 2009), and three studies explored preferences for ovarian cancer follow-up (Cox et al., 2008; Lydon et al., 2009; Palmer et al., 2006: Table 8).

Two studies were limited to exploring the preferences of patients diagnosed with an early stage of disease (Bradley et al., 1999; Bradley et al., 2000: see Table 8 for how this was defined). One study was limited to exploring the preferences of patients diagnosed with stage II to stage IV disease, however, in the results section reported having recruited patients diagnosed with stage I to IV disease (Cox et al., 2008). Four studies did not report having restricted recruitment by stage of disease (Howells et al., 1999; Kew et al., 2007/Kew et al., 2009; Lydon et al., 2009; Palmer et al., 2006). However, five studies did not report the stage of disease of participants recruited (Bradley et al., 1999; Howells et al., 1999; Kew et al., 2007/Kew et al., 2009; Lydon et al., 2009; Palmer et al., 2006). Six studies did not report the treatment received by participants (Bradley et al., 1999; Bradley et al., 2000; Cox et al., 2008; Howells et al., 1999; Lydon et al., 2009; Palmer et al., 2006).

There was one focus group study (Lydon et al., 1999), one interview study (Bradley et al., 1999), three cross sectional studies (Bradley et al., 2000; Howells et al., 1999; Kew et al., 2007/Kew et al., 2009), one audit (Palmer et al., 2006) and one repeated measures intervention study (Cox et al., 2008). Using the weight of evidence framework as described above, two studies were assessed as being of an overall medium quality (Howells et al., 1999; Kew et al., 2007/Kew et al., 2009) and five as overall low quality (Bradley et al., 1999; Bradley et al., 2000; Cox et al., 2008; Lydon et al., 2009; Palmer et al., 2006: see Table 8). Whilst studies were not excluded based on their assessment, it should be considered when interpreting and applying the results of the synthesis.

Table 8. Study design and reported effects in patient sample (n=7/8)

	Study characteristics				Study Results		Weight of Evidence
Author, year	Study type	Sample size	Demographics	Inclusion/exclusion criteria	Themes	Results relating to theme	A/B/C/D*
Bradley et al., 1999	Interview study	N=12	Treatment completed in previous 6-72 months Type of cancer: not reported Stage of disease: diagnosed with stage I-II disease (inclusion criteria) Treatment received: not reported Any co-morbidities: not reported.	Women diagnosed with early stage (defined as stage I or II) endometrial, ovarian, cervical or vulvar cancer, have been in follow-up care for at least 6 months.	Hospital follow-up GP follow-up	Aim of follow-up: provide reassurance. Post-treatment patients experienced high levels of anxiety. This anxiety built up before their appointment, in regard to fear of a detection of recurrence, and was reduced by attending the follow-up appointment when reassured that there was no sign of recurrence. “I rely on them telling me nothing’s wrong...” Patients purposefully sampled to include patients with a preference for discharge. Those with a preference to be discharged from care (n=5) and patients with a preference for continued hospital follow-up (n=5) and those not sure	A: Low B: Low C: Low D: Low

						<p>(n=2) if they wanted continued follow-up or to be discharged, all experienced this anxiety.</p> <p>Hospital follow-up: patients valued hospital follow-up because of the informational reassurance provided through effective communication in the consultation, having the opportunity to express symptoms concerns, and being informed about what symptoms of recurrence were. Prior to diagnosis, participants reported not having symptoms but on further investigation symptoms were present. Being informed of possible symptoms of recurrence enabled monitoring behaviour which reduced anxiety. Patients also received reassurance from the physical examination detecting no signs of recurrence. One patient paid for a scan to receive technical reassurance.</p>	
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						GP follow-up: Patients did not value the idea of GP follow-up, as felt that General Practitioners were not experts, so therefore could not provide the reassurance needed.	
Bradley et al., 2000	Cross-sectional study	*N=54	*Type of cancer: n=14/53 endometrial cancer patients, n=17/53 cervical cancer patients, n=17/53 ovarian cancer patients, n=5/53 vulvar cancer patients Stage of disease: not reported Treatment received: primary treatment not adjuvant therapy (inclusion criteria) Follow-up: n=30/53 <18 months, n=23/53 >18 months	Diagnosed with early stage gynaecological cancer (defined as not requiring adjuvant therapy) and attending routine follow-up	Hospital follow-up Self-management	Preference to continue with standard care or be discharged (defined as no routine follow-up appointment, a contract to say re-entry to clinic if change mind or symptomatic): 39% (n=21/54) had a preference for standard hospital follow-up (exact model of care not defined). 61% (n=33/54) had a preference to be discharged. Explored whether demographics and scores on demographic measures differed by those who had a preference for discharge or continued follow-up. Participants with a preference for discharge were more likely to, and was significantly predicted by owning a car, be unemployed, and have lower levels of anxiety (as measured on the mental adjustment to cancer scale).	A: Low B: Medium C: High D: Low

			Any co-morbidities: n=8/48 other cancer (current or treated), n=40/48 no other cancer (missing data not reported)				
Cox et al., 2008	Repeated measures intervention study (no control group).	N=46 completed measures	Type of cancer: Ovarian cancer. Stage of disease: 49% diagnosed with stage 1 or 2 disease, 51% diagnosed with stage 3 or 4 disease (n not reported). Stage of disease: not reported Treatment received: not reported Any co-morbidities: not reported.	Patients diagnosed with stage II-IV stable ovarian cancer and had completed initial cancer treatment. Women who did not own a telephone, were unable to speak English or were confused were excluded.	Hospital follow-up Specialist nurse telephone follow-up	Participants were given a specialist nurse led telephone follow-up intervention. Participants received three phone calls over a 10 month period. Patients received CA125 testing in the GP clinic before appointment. The phone call lasted 20 minutes (with a call back if longer needed) and focused on the detection of recurrence and personalised psychosocial support. 73% (n=32/44) participants had a preference for Specialist nurse led telephone follow-up, 18% (n=8/44) a preference for standard hospital follow-up, and 9% (n=4/44) were not	A: Low B: Low C: Low D: Low

			Duration in follow-up: not reported.			<p>sure which model of care they preferred.</p> <p>On a scale of 1 (dreadful) to 10 (excellent) the mean score for telephone support received was 8.24 (SD 2.4)</p> <p>Regarding aspects of specialist nurse led telephone follow-up valued (n=35 respondents but 43 reasons given):</p> <ul style="list-style-type: none"> • 39% valued the relationship with the nurse and areas of discussion “knowing the nurse and feeling that she really understands” (P13) “I chose to talk openly ... the oncology nurse is not embarrassed” (P19) • 33% valued the convenience (time/travel) “not having to go to the clinic and wait a long time” (P26) “Not have to take time off work” (P48) 	
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Howells et al., 1999	Cross- sectional study	N=90	Type of cancer: n=23/90 ovarian cancer, n=37/90 cervical cancer, n=19/90 uterus cancer, n=11/90 vulvar cancer Stage of disease: not reported Treatment received: not reported Duration in follow-up care: mean 30 months (range 1-5 years)	Patients receiving follow-up care for gynaecological cancer.	Hospital follow-up GP follow-up Self- management	<p>60% (n=54) wanted continued follow-up appointments. 40% (n=36) had a preference to be discharged from follow-up care and instead have a self-management model of care (defined as no planned follow-up appointments in secondary or primary care, given contact number if any concerns with rapid re-access to the clinic by self-referral if required).</p> <p>Of the n=54 that had a preference for continued follow-up appointments, 32% (n=29/54) wanted these to be held in secondary care and 28% (n=25/54) wanted these to be held in primary care.</p> <p>By cancer type there was no statistically significant differences in the preference for continued follow-up or discharge.</p>	A: Medium B: Low C: High D: Medium
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Kew et al., 2007/2009	Cross-sectional study	N=96	<p>Cancer type: n= 31/96 (32%) ovarian cancer, n=14/96 (14%) endometrial cancer, n=23/96 (24%) cervical cancer, n= 9/96 (9%) vulva cancer, n= 1/96 (1%) vaginal cancer, n=18/96 (19%) missing data.</p> <p>Stage of disease: not reported</p> <p>Treatment received†: n=90/96 (94%) surgery and adjuvant therapy, n=22/96 (23%) chemotherapy, n=7/96 (7%) radiotherapy, n=1/96 (1%) missing data.</p>	Patients receiving follow-up care for gynaecological cancer.	<p>Hospital follow-up</p> <p>GP follow-up</p> <p>Self-management</p>	<p>Aim of follow-up: detect recurrence (1.40 MR: mean rank), monitor treatment recovery (3.01 MR), treat symptoms (3.39 MR), discuss concerns (3.82 MR), treat side effects of treatment (4.16), data collect (5.22 MR)</p> <p>Preference for which Health Care Professional to provide care: 89% (n=52/92) hospital Doctor, 24% (n=22/92) Specialist Nurse, 7% (n=6/92) General Practitioner.</p> <p>Most important content of care: consultation itself is the most important 43% (n=41/96), 73% (n=70/96) examination is the most important, 20% (n=19/96) blood test is the most important. Patients with prior experience of a blood test were more likely to identify it as being the most important content of care 55% (n=16/29) compared with no experience of a blood test 5% (n=3/61). Women with ovarian</p>	<p>A: Medium</p> <p>B: Medium</p> <p>C: High</p> <p>D: Medium</p>
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			Any comorbidities: not reported Duration in follow-up: not reported.			cancer more likely to report a blood test as being the most important content of care (unknown whether patients with each type of gynaecological cancer experienced a blood test)	
Lydon et al., 2009	Focus group	Patients (n=6, one focus group)	Cancer type: Ovarian cancer. Stage of disease: not reported Treatment received: not reported Any comorbidities: not reported Duration in follow-up care: not reported	Diagnosed with epithelial ovarian cancer (most common form of ovarian cancer) and a stable, partial or complete response to hormonal treatment/chemotherapy but not currently receiving treatment for disease progression, and	Hospital follow-up Self-management Specialist nurse telephone follow-up GP follow-up	Aim of follow-up: check for cancer recurrence Need for hospital follow-up or discharge from care was mediated by duration in follow-up, with patients at the beginning of their journey having a need to see the consultant for follow-up appointments, with patients further from diagnosis feeling confident in managing their own symptoms and feeling that they were wasting the Health Care Professionals' time by having appointments.	A: Medium B: Low C: Low D: Low

				<p>willing and able to take part in a focus group.</p> <p>Excluded if any adverse circumstances that would require exclusion</p>		<p>Hospital follow-up: patients in the earlier stage of their follow-up journey felt the need to see the hospital Doctor for appointments in order to be monitored for symptoms and so potential recurrence, and didn't feel able to manage this themselves. Patients felt they needed an examination, to receive information and support, re access to the clinic in between appointments if needed and support in between their last treatment and first scheduled three month follow-up appointment, in order to receive the reassurance they felt they needed. The presence of the Specialist Nurse at the follow-up clinic added to the feeling of reassurance. However, some patients waited for their next follow-up appointment to discuss concerns regarding symptoms of recurrence rather than re-accessing care in between these scheduled appointments. Patients valued the</p>	
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						<p>long waiting times in the clinic, as valued the opportunity to support others.</p> <p>Specialist nurse delivered telephone follow-up: There were mixed views regarding this hypothetical provision. Some patients valued the idea of nurse led telephone follow-up due to it being less strain logistically, but others did not value the idea as it would mean having no examination which was assumed to be essential, and that over the telephone there are no visual cues so symptoms might be missed.</p> <p>GP delivered follow-up: Patients did not value the idea of GP led follow-up, as 1) felt that General Practitioner's did not have specialist knowledge and expertise, 2) felt that they did not have the same level of rapport with their General Practitioner as they did with hospital Doctors, 3) there was not continuity</p>	
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						of Doctor seen in primary care but there was in secondary care and 4) they felt more rushed in GP appointments than they did when they attended hospital follow-up.	
Palmer et al., 2006	Audit	<p>Questionnaire 1: N=90</p> <p>Questionnaire 2: N=26</p>	<p>Type of cancer: Ovarian cancer</p> <p>Questionnaire 1: Stage of cancer: not reported Treatment received: not reported Any co-morbidities: not reported Time since diagnosis: n=31/90 (35%) <1 year; n=21/90 (23%) 12-18 months; n=12/90 (13%) 19-24 months; n=4/90</p>	Ovarian cancer patients receiving follow-up care	Hospital follow-up	<p>Current practice was that CA125 was undertaken at the clinic. If CA125 level was normal or elevated and patient asymptomatic patients were not informed of their CA125 level until they attended their next follow-up visit unless they telephoned the clinic. If CA125 raised and patient symptomatic next appointment brought forward. This was logistically problematic for the clinic, patients misunderstood a slight raise in CA125 levels over the phone and patients had increased anxiety.</p> <p>Questionnaire to identify if patients wanted CA125 result available in clinic, and if patients were informed</p>	<p>A: Medium B: Low C: Low D: Low</p>

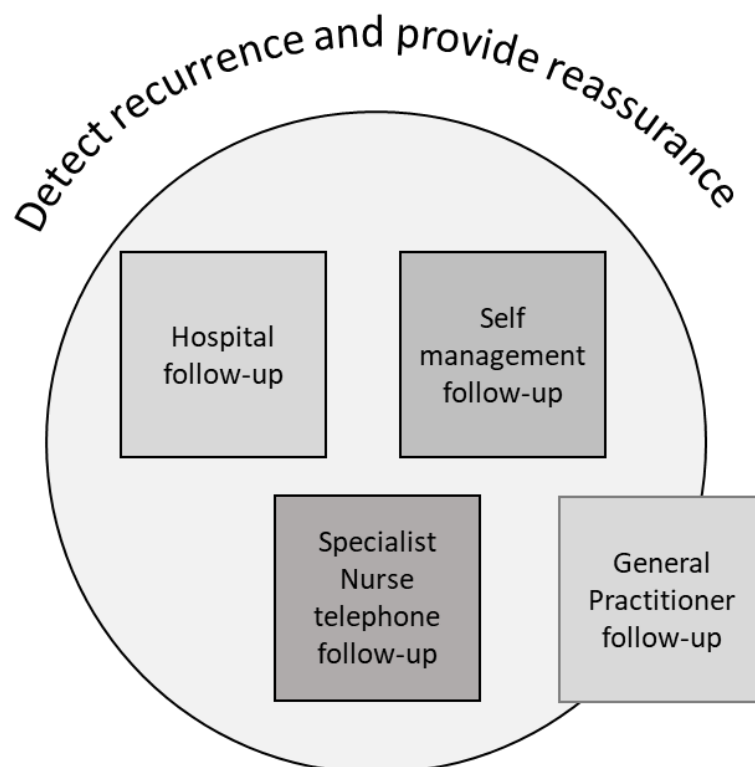
			<p>(5%) 25-36 months; n=20/90 (22%) >3 years; n=2/90 (2%) missing data</p> <p>Questionnaire 2</p> <p>Stage of cancer: not reported</p> <p>Treatment received: not reported</p> <p>Any co-morbidities: not reported</p> <p>Time since diagnosis: n=4/26 (15%) 25-36 months; n=22/26 (85%) >3 years</p>		<p>about the importance of CA125 testing.</p> <p>Under old model: 50% (n=45/90) satisfied with care, 34% (n=31/90) not satisfied. 81% (n=73/90) wanted the result available at the clinic appointment, 2% (n=2/90) did not want the result available at the clinic appointment, 16% (n=14/90) had no preference. 82% (n=74/90) were happy to attend the local GP surgery for CA125 testing prior to their clinic appointment, 16% (n=14/90) were not happy to attend the local GP surgery for CA125 testing prior to their clinic appointment. 81% had a basic understanding of CA125 testing.</p> <p>There was a change in practice to CA125 testing in GP clinic prior to secondary care appointment, with results available at this follow-up appointment. CA125 patient education information produced.</p>	
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						<p>Under new model: 92% (n=24/26) felt the model of care had been improved, 4% (n=1/26) did not think it had been improved, 4% (n=1/26) provided no response. For 92% (n=24/26) of patients, their CA125 result was available at the clinic, for 4% (n=1/26) it usually was and for 4% (n=1/26) it was not. 89% felt that they had a basic understanding of CA125 testing.</p>	
<p>*Weight of evidence A as assessed by relevant CASP form/ Weight of evidence B as assessed by the suitability of the design and analysis in answering the research question of the review/ Weight of evidence C as assessed by the suitability of the topic, sample, population, context, measures and overall evidence focus in answering the research question of the review/ Weight of evidence D overall judgement taking into consideration all of the above (Gough et al, 2013)</p>							

Aim of follow-up

As identified in a cross-sectional study of patients diagnosed with early stage gynaecology cancer (Bradley et al., 1999), a cross-sectional study of patients diagnosed with gynaecology cancer (stage of disease not reported: Kew et al., 2007/Kew et al., 2009) and a single focus group of patients diagnosed with ovarian cancer (stage of disease not reported: Lydon et al., 2009), patients felt that the primary aim of follow-up care was to detect any recurrence and provide reassurance in the absence of disease detection (Bradley et al., 1999; Kew et al., 2007/Kew et al., 2009; Lydon et al., 2009). A cross-sectional study of patients diagnosed with early stage gynaecological cancer, identified that follow-up appointments reduced patients' post-treatment anxiety which peaked before their appointment, but was felt to be necessary in reducing their anxiety regarding perceived risk of recurrence (Bradley et al., 1999). Patients had differing preferences for the model of care to meet these needs: 'hospital follow-up', 'Specialist Nurse led telephone follow-up', and 'self-management'. Patients were generally sceptical of the idea of 'General Practitioner led follow-up', questioning the General Practitioner's ability to detect recurrence and so provide reassurance. These iteratively identified themes and sub-themes are illustrated by the emerging framework in Figure 3.

Figure 3: Framework of patients' preferences for follow-up care



Hospital follow-up

The theme 'hospital follow-up' was identified by all studies. Three studies (a study of gynaecology cancer generally, Kew et al., 2007/Kew et al., 2009; ovarian cancer, Lydon et al., 2009; early stage gynaecology cancer, Bradley et al., 1999) illustrated that this was because patients placed a strong value on hospital follow-up because of its perceived value in detecting recurrence, and so provision of reassurance when there were no signs of recurrence (Bradley et al., 1999; Kew et al., 2007/Kew et al., 2009., Lydon et al., 2009).

Lydon et al., (2009): 'I don't think that is pleasant (internal examination) but I feel that it is necessary and relevant and that may save your life'

Results from a study of early stage gynaecology cancer follow-up demonstrated that 39% (n=21/54) of participants stated a preference for hospital follow-up (compared to self-management: Bradley et al., 2000). Similarly, in a study of preferences for gynaecology cancer follow-up (not limited by stage) 60% (n=54/90) of participants stated a preference for continued follow-up as opposed to self-management, and of these 54% (n=29/54) wanted these appointments to be held in secondary care compared to primary care (Howells et al., 1999). Additionally, in a cross-sectional study of gynaecology cancer follow-up not limited by stage of disease, 89% (n=52/92) of the participants that responded to this question had stated a preference for care to be delivered by a hospital Doctor (Kew et al., 2007/Kew et al., 2009).

Findings from an interview study of early stage gynaecology cancer follow-up (Bradley et al., 1999), and a focus group study of preferences for ovarian cancer follow-up that did not report stage of disease (Lydon et al., 2009), identified that reassurance was provided by the consultation itself through 1) effective communication between the Health Care Professional and patient (Bradley et al., 1999), 2) being provided with the opportunity to express concerns (Bradley et al., 1999), 3) receiving blood tests (Bradley et al., 1999), 4) an internal examination (Bradley et al., 1999; Lydon et al., 2009), 5) receiving information regarding symptoms of recurrence (Bradley et al., 1999; Lydon et al., 2009), 6) support, and 7) re-access to the clinic in between appointments if concerns (Lydon et al., 2009). Ovarian cancer follow-up patients in a focus group comprised of six participants (with unknown demographics), had a preference to receive care in between their last treatment and first follow-up appointment in order to have the support they felt they needed (Lydon et al., 2009). A cross-sectional study of 96

gynaecology cancer patients with an unknown stage of disease but differing treatment pathways, verified these findings (Kew et al., 2007/Kew et al., 2009). Patients rated the 'most important' aspects of care as the consultation (43%, n=41/96), the examination (73%, n=70/96) and the blood test (20%, n=19/96). However, participants provided multiple answers, limiting the findings that can be taken from these results. Interestingly, participants with prior experience of a blood test were more likely to respond that it was the most important aspect of care (Kew et al., 2007/Kew et al., 2009). Similarly, an interview study of 12 participants diagnosed with early stage gynaecological cancer, identified that one patient paid to have a scan to receive technical reassurance, as she felt she needed further reassurance than that was provided under standard care (Bradley et al., 1999). A focus group of patients diagnosed with ovarian cancer (with unknown stage, treatment received and duration in follow-up), identified that hospital follow-up was particularly valued by participants earlier on in their follow-up pathway, where patients relied more on the Consultant to manage their disease. Conversely, a patient in follow-up for 10 years felt like she was able to manage her disease and that she was wasting resources by still attending follow-up appointments (Lydon et al., 2009). However, the stage of disease at diagnosis, treatment received and duration in follow-up was not reported (Lydon et al., 2009). Nevertheless, provision of scheduled follow-up appointments resulted in some patients waiting till their next scheduled appointment to discuss symptoms of recurrence, rather than re-accessing the clinic in between appointments (Lydon et al., 2009). Despite sometimes a long wait in the clinic for a scheduled hospital follow-up appointment, some patients valued this wait, as they wanted to be able to support others in the earlier stages of their cancer pathway, who were also waiting (Lydon et al., 2009). Whilst the results from this study are interesting and add to the knowledge base they should be interpreted with caution, as the researchers were known to patients, only one focus group with 6 ovarian cancer patients were recruited from one hospital, and the sample was reported to be not reflective of the cancer population, although exact demographics were not reported (Lydon et al., 2009).

An audit with ovarian cancer follow-up patients identified that, rather than being tested for CA125 levels at their follow-up appointment, and if asymptomatic or having no rise in CA125 levels not receiving the results till their next follow-up appointment, 81% (n=73/90) had a preference to receive the result at the follow-up appointment, and 82% (n=74/90) were happy to receive the test in primary care so that this was possible. Following a change in

practice so that testing took place at the patients' primary care surgery and results available at the hospital clinic, with also the design of patient education information, 92% (n=24/26) felt the model of care had been improved (Palmer et al., 2006). It should be noted that testing of CA125 levels in the follow-up of ovarian cancer is no longer recommended and so no longer generally part of routine practice in the United Kingdom (Leeson et al., 2017).

Specialist Nurse led telephone follow-up

Preferences for Specialist Nurse led telephone follow-up were explored in a focus group of 6 ovarian cancer patients where the stage of disease and treatment received was unknown (Lydon et al., 2009); a cross sectional study of 96 patients diagnosed with gynaecology cancer (where the stage of disease was not reported, but treatment received included surgery, chemotherapy and radiotherapy: Kew et al., 2007/Kew et al., 2009); and in a repeated measures intervention study of ovarian cancer patients (49% diagnosed with stage I-II disease, 51% diagnosed with stage III-IV disease: Cox et al., 2008). In the focus group of ovarian cancer patients, there were mixed views regarding this hypothetical provision. Some patients valued the idea of Specialist Nurse led telephone follow-up, as they felt that not physically attending appointments would place less strain on them. However, others did not value the idea given 1) the assumed importance of the physical examination in detecting recurrence, and so concomitant perception of the physical examination as a necessity, and 2) that over the telephone there are no visual cues so symptoms might be missed (Lydon et al., 2009).

Supporting these findings were results from a repeated measures intervention study (Cox et al, 2008). Forty-six patients diagnosed with stage I-IV ovarian cancer received three Specialist Nurse telephone delivered follow-up appointments over a period of 10 months, with an aim of detecting any recurrence and providing personalised psychosocial support (with CA125 testing conducted at the primary care surgery and results given in the follow-up appointment). Post intervention 73% (n=32/44) of these patients had a preference for this model of care, 18% (n=8/44) had a preference for standard hospital follow-up, and 9% (n=4/44) were unsure which model of care they had a preference for. In particular patients valued the relationship with the Specialist Nurse and areas of discussion, and the convenience of telephone appointments in terms of time and travel.

Cox et al., (2008: P13): "knowing the nurse and feeling that she really understands"

Cox et al., (2008: P26): "not having to go to the clinic and wait a long time"

The authors report that other aspects of Specialist Nurse led telephone follow-up were valued, but did not report themes or data to support this. Additionally, the reason for patients valuing the standard model of hospital follow-up, or reasons for participants not being sure which model of care they preferred were not explored, and experience and satisfaction with care prior to the intervention was collected but not reported (Cox et al., 2008).

A questionnaire study explored the perception of the role of the Specialist Nurse in follow-up. However, the question used the term 'nurse' not 'Specialist Nurse' so the results lack validity, and as such are not reported here (Kew et al., 2007/Kew et al., 2009).

General Practitioner led follow-up

The theme 'General Practitioner led follow-up' was identified by a focus group comprised of patients diagnosed with ovarian cancer with their stage of disease unreported (Lydon et al., 2009); an interview study comprised of 12 patients diagnosed with early stage gynaecology cancer (Bradley et al., 1999); a cross-sectional study of 90 patients diagnosed with gynaecology cancer with stage of disease unreported (Howells et al., 1999); and a cross-sectional study of 96 gynaecology cancer patients, also with stage of disease unreported (Kew et al., 2007/Kew et al., 2009). The focus group (Lydon et al., 2009) and interview study (Bradley et al., 1999) identified that the majority of patients did not value the idea of General Practitioner led follow-up, as:

- 1) They felt that General Practitioners did not have specialist knowledge and expertise (Bradley et al., 1999; Lydon et al., 2009).
- 2) They felt they did not have the same level of rapport with their General Practitioner as they did with hospital Doctors (Lydon et al., 2009).
- 3) There was not continuity of Doctor seen in primary care but there was in secondary care (Lydon et al., 2009).
- 4) They felt more rushed in appointments with their General Practitioner than they did when they attended hospital follow-up appointments in secondary care (Lydon et al., 2009).

It was therefore felt that General Practitioners could not provide patients with the reassurance they felt they needed (Bradley et al., 1999). In support of the findings, in a cross-sectional study of gynaecology cancer follow-up, 7% (n=6/92) of participants who responded to this question, had a preference for General Practitioner led follow-up over follow-up delivered by a hospital Doctor or Specialist Nurse. The reasons for this are unknown (Kew et al., 2007/Kew et al., 2009). Nevertheless, in a cross-sectional study, Howells et al (1999) identified that 28% (n=25/90) of ovarian cancer patients had a preference for follow-up to delivered by the General Practitioner, but again the reason for this was not explored (Howells et al., 1999).

Self-management

Three studies explored the self-management model of care: a cross-sectional study of 54 patients diagnosed with early stage gynaecology cancer (defined as not requiring adjuvant therapy, but exact staging not reported: Bradley et al., 2000); a cross-sectional study of 90 gynaecology cancer patients (stage of disease not reported: Howells et al., 1999); a focus group study comprised of one focus group with six ovarian cancer follow-up patients (stage of disease not reported: Lydon et al., 2009). Results from the cross-sectional study of early stage gynaecology cancer follow-up, demonstrated that 61% (33/54) stated a preference for the self-management model of care compared to standard care. Standard care was not defined but self-management was defined as no routine follow-up appointment, and a contract to say re-entry to the clinic if the patient changed their mind and wanted hospital follow-up, or if the patient had symptoms of recurrence (Bradley et al., 2000). The authors explored differences in demographics and psychometric scores between those who had a preference for discharge (n=33) and standard care (n=21). The authors reported that participants with a preference for the self-management model of care were more likely to have lower levels of anxiety, be unemployed or own a car. However, there was a large risk of error due to the small sample size and number of statistical tests conducted (independent t-tests, chi squared tests, and multiple regressions), therefore these findings lack validity (Bradley et al., 2000). Similarly, the cross-sectional study of gynaecology cancer follow-up patients (not limited by stage), demonstrated that 40% (n=36/90) had a preference for this model of care compared to scheduled appointments. In this study, in addition to the above, self-management was also defined as the patient being given the contact details for the clinic in case any concerns, with

rapid re-access by self-referral if required (Howells et al., 1999). Howells et al (1999) reported that there was no statistically significant difference between preferences for self-management care by cancer type (Howells et al., 1999). Neither of these cross-sectional studies explored why these patients had a preference for the self-management model of care (Bradley et al., 2000; Howells et al., 1999). In the focus group study of ovarian cancer follow-up, one participant 10 years post-treatment explained that as the duration from treatment increased, her confidence in her ability to self-manage her disease increased, and she felt that continued hospital follow-up was a waste of the clinicians time (Lydon et al., 2009). Whilst an interview study recruited participants in part to include those with a preference for discharge and included a preference for continued care or discharge in the semi-structured interview guide, preferences for discharge were not explored or presented in the results of the study (Bradley et al., 1999).

Conclusion

In conclusion, from the studies identified in the systematic search and explored above, it can be seen that patients place a large value on scheduled follow-up appointments because of the reassurance that these appointments provide in absence of detection of recurrence of disease. Because of this, patient's valued hospital led follow-up provision. However patients, particularly those with experience of Specialist Nurse led telephone follow-up, value Specialist Nurse led telephone follow-up. Even without experience of alternative models of care some patients have a preference for Specialist Nurse led, General Practitioner led or the self-management model of care. However, reasons for these preferences for alternative models of care are largely unexplored or not reported.

2.4.3.2 Synthesis of Health Care Professionals preferences (n=3/8)

Three studies exploring the preferences of Health Care Professionals were conducted in the United Kingdom (Kew et al., 2007; Lydon et al., 2009; Penney et al., 1995). A cross-sectional study explored preferences for gynaecology cancer follow-up generally, in a sample of 32 Health Care Professionals with varied occupations (but when displaying the results reported that the total sample size was 34: Kew et al., 2007). Another study used 2 items in a cross-sectional questionnaire to explore the preferences for vulvar cancer follow-up in a sample of 78 consultant gynaecologists in Scotland (Penney et al., 1995). A focus group study comprised

of seven Health Care Professionals of mixed occupations working in one hospital, was used to explore preferences for ovarian cancer follow-up (Lydon et al., 2009). Using the weight of evidence framework as described above, two studies were assessed as being of a medium quality (Kew et al., 2007; Penney et al., 1995) and one of low quality (Lydon et al., 2009: see Table 9).

A cross-sectional study identified that in order of importance, Health Care Professionals felt the aim of gynaecology cancer follow-up was to: discuss concerns, detect recurrence, treat symptoms and side effects of treatment, monitor recovery from treatment and collect data (Kew et al., 2007). This finding is in part supported by Lydon et al (2009), who qualitatively identified that Health Care Professionals from one treating hospital within one focus group, felt that the aim of ovarian cancer follow-up was to detect recurrence (Lydon et al., 2009).

To meet these aims, Health Care Professionals had a strong preference for a changing model of care given:

- 1) The lack of evidence regarding the effectiveness of standard care in detecting asymptomatic recurrence.
- 2) That if the standard model of care did detect recurrence patients would not be treated with curative intent.
- 3) The burden these unnecessary appointments created on the clinic. This need for a changing model of care was highlighted implicitly by the aim of the studies, questionnaire items and semi-structured interview guide (Kew et al., 2007; Lydon et al., 2009), and explicitly by the data generated from Lydon et al's (2009) focus group study:

Lydon et al (2009) 'Is there any justification for continuing this intensive form of follow-up in the absence of any benefit?' [Consultant]

Within the core theme of a 'changing model of care', two further themes were identified 'self-management' and 'multidisciplinary care', the latter with sub themes 'Specialist Nurse involvement' and 'General Practitioner involvement'. These iteratively identified themes and sub themes are illustrated by the framework in Figure 4.

Table 9. Study design and reported effects in Health Care Professional sample (n=3/8)

	Study characteristics				Study Results		Weight of Evidence
Author, year	Study type	Sample size	Demographics	Inclusion/exclusion criteria	Themes	Results relating to theme	A/B/C/D*
Kew et al (2009)	Cross-sectional questionnaire	N=32 (results reported for N=34)	<p>Health Care Professional speciality: n=29/34 gynaecological oncologists, n=3/34 medical oncologists, n=1/34 clinical oncologists, n=1/34 gynaecologist</p> <p>Health Care Professional profession: n=22/34 consultants, n=4/34 sub-speciality trainees, n=1/34 associate specialist, n=5/34 specialist registrars, n=2/34 cancer nurse specialist</p>	Involved in follow-up and working in the department at the recruiting hospital or attending a national gynaecology oncology meeting	<p>Changing model of care</p> <p>Multidisciplinary care</p>	<p>Aim of follow-up to talk about concerns (1.9), detect recurrence (3.2), treat symptoms (3.4), treat side effects of treatment (3.5), check treatment recovery (3.6) and collect data (5.4: mean ranking)</p> <p>Health care professional seen for appointments: 73% (n=25) felt hospital Doctor should deliver appointments; 56% (n=19) felt the specialist nurse should be involved in the delivery of follow-up care; 24% (n=8) felt that General Practitioner's should be involved in providing follow-up appointments.</p> <p>Content of care: 73% (n=27) of health care professionals rated the consultation as the most important aspect of follow-up care, 15% (n=5) rated the examination as the most</p>	<p>A: Medium</p> <p>B: Medium</p> <p>C: High</p> <p>D: Medium</p>

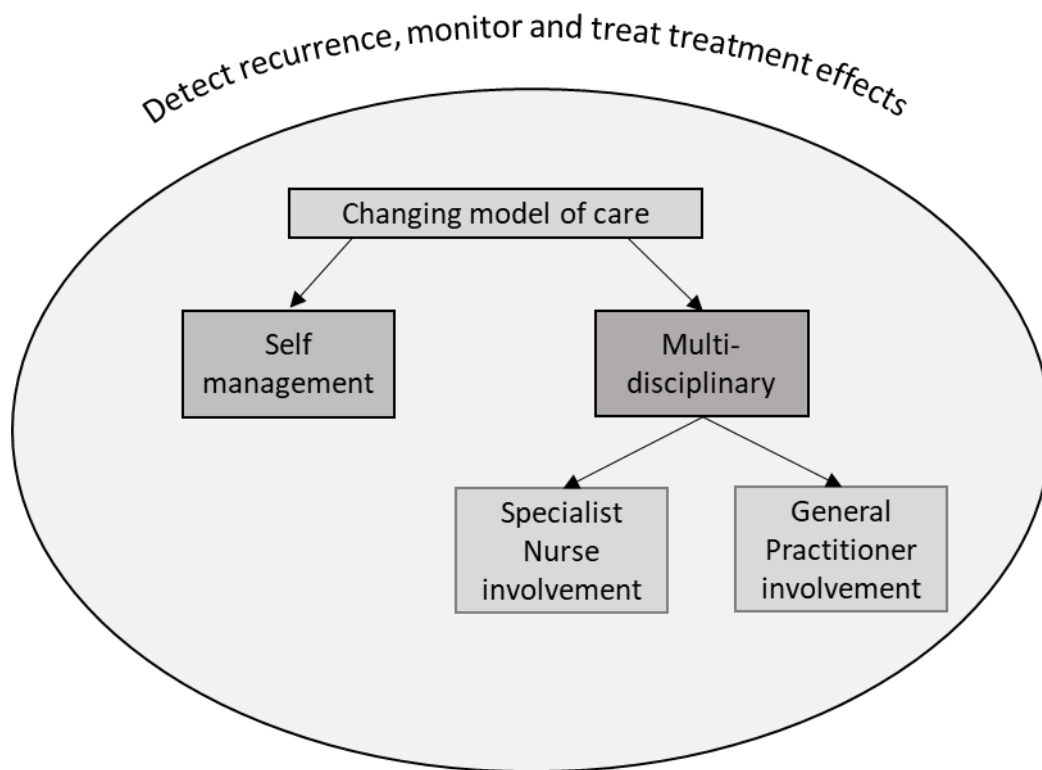
						important aspect of follow-up, 6% (n=2) rated the blood test as the most important aspect of follow-up.	
Lydon et al (2009)	Cross-sectional focus group	N=7	Health Care Professional profession: n=1 consultant oncologist, n=2 senior registrars, n=1 junior registrar, n=2 research registrars, n=1 nurse clinician.	A member of the oncology team (all with experiences of delivering ovarian cancer follow-up), or until recently a part of the oncology team at the recruiting hospital	Changing model of care Self-management Multidisciplinary care	Health care professionals were asked about the current model of care and their view on alternative models of follow-up care. Health Care Professional's felt that the aim of follow-up was to monitor symptoms of recurrence but that there was a need for a changing model of follow-up care as 1) no evidence to show that the current model was effective in detecting recurrence in the absence of symptoms, with recurrence usually symptomatic or detected by increased CA125 levels 2) if recurrence was detected the patient would not receive curative treatment 3) the current model of care is based on tradition and not questioned but creates unnecessary workload. Participants felt that follow-up care could better meet patients needs and reduce the burden on the clinic if an alternative model of care was delivered:	A: Medium B: Low C: Low D: Low

						<p>Content of care: Health care professionals valued the consultation for patients at risk of disease progression but questioned the importance of consultations for asymptomatic patients. Instead they valued: alternating nurse led telephone follow-up clinic with face to face secondary appointments to increase patients' confidence in managing their own disease, and felt that this model of care would be more acceptable to patients than just receiving nurse led telephone follow-up or a self-management model of care given patients' reluctance to let go of consultant led care. Health care professionals felt that GP's sharing the delivery of follow-up care would also improve follow-up care for patients and reduce the demand and so increase the quality of follow-up delivered in secondary care. However, participants had the strongest preference for a self-management model of care to place the responsibility of disease monitoring back on the patient improving patient wellbeing, reducing patient burden of clinic attendance and reducing the</p>	
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						<p>burden on the clinic. Participants felt they needed to encourage patients more in taking ownership of their disease to inform and empower them, and that oral and written information with symptoms of recurrence and contact details should be given towards the end of treatment. CA125 testing was assumed by participants to be a core component of follow-up care, with nurse led telephone follow-up care only possible if this test was conducted prior to the appointment in primary or secondary care. Health Care Professionals were reluctant to ask patients lots of questions during follow-up appointment, as didn't want patients to discuss things not relevant to the cancer and that could be addressed in primary care.</p> <p>Health Care Professionals did not think frequent visits improved survival, and had a strong preference for their frequency to be reduced in line with the above alternative models of care</p>	
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Penney et al (1995)	Cross sectional questionnaire	N=78	N=78 consultant gynaecologists	Consultant gynaecologists involved in the management of vulvar cancer practising in Scotland	Changing model of care Multidisciplinary care	97% of participants strongly agreed or agreed that patients should receive frequent follow-up appointments over a long duration 45% of participants strongly agreed or agreed that follow-up should take place at a specialised multidisciplinary follow-up clinic	A: Medium B: Medium C: Low D: Medium
<p>*Weight of evidence A as assessed by relevant CASP form/ Weight of evidence B as assessed by the suitability of the design and analysis in answering the research question of the review/ Weight of evidence C as assessed by the suitability of the topic, sample, population, context, measures and overall evidence focus in answering the research question of the review/ Weight of evidence D overall judgement taking into consideration all of the above (Gough et al, 2013)</p> <p>†report that N=32 but report 34 was the total possible number of responses to questionnaire items</p>							

Figure 4: Framework of Health Care Professionals preferences for follow-up care



2.4.3.3 Self-management

The theme ‘self-management’ was identified by a focus group study exploring preferences for ovarian cancer follow-up (Lydon et al., 2009). In the one focus group conducted, comprised of Health Care Professionals of different occupations at one treating hospital, where the researchers were known to participants, it was reported that the self-management model was the best option in terms of a changing model of care, with patients encouraged to take ownership of their disease and given verbal and written information about symptoms of recurrence, and who to contact should they experience any of these symptoms. However, it was acknowledged that this model of care is unlikely to be acceptable to patients given that:

*Lydon et al (2009): “Patients do not want to be ‘cut loose’ from the clinic scenario”
[Specialist Nurse]*

Further, a cross-sectional study of Health Care Professionals preferences for gynaecology cancer follow-up generally, identified that 73% (27/34) of participants felt the most important attribute of follow-up care was the consultation itself, compared to the examination (15%,

n=5/34), and blood tests (6%, n=2/34: Kew et al., 2007). By its nature, a self-management model of care would take away the consultation, and so examination and blood tests. However, blood tests are not currently delivered as part of standard care in the United Kingdom (Leeson et al., 2017).

2.4.3.4 Multi-disciplinary care

The theme 'multi-disciplinary care' was identified by all three identified studies (Kew et al., 2007; Lydon et al., 2009; Penney et al., 1995). Two items in a questionnaire to identify preferences for vulvar cancer follow-up identified that of 78 consultant gynaecologists recruited in Scotland, 45% either 'strongly agreed' or 'agreed' it should take place at a special multi-disciplinary clinic, and 97% either 'strongly agreed' or 'agreed' that there should be "regular and long term follow-up" (Penney et al., 1995). However, these attributes of care were not defined in the journal article and it is unknown whether it was defined to participants. Comparatively, in the focus group study, Health Care Professionals felt that multidisciplinary care was the way forward in regard to the changing model of care for ovarian cancer follow-up (Lydon et al., 2009). However, preferences identified in the cross-sectional study for vulvar cancer follow-up were concurrent with a preference for regular follow-up over a long duration (Penney et al., 1995). Whereas preferences identified in the focus group study for ovarian cancer follow-up were driven by the need to reduce the number of appointments, and so the burden on secondary care (Lydon et al., 2009). These differences could be attributed to differences in the rate of and presentation of recurrence (Guerts et al., 2012; Leeson et al., 2017; Oonk et al., 2003; Rustin et al., 2010).

2.4.3.5 Specialist Nurse involvement

A focus group study comprised of different Health Care Professionals working in medical oncology within one treating hospital, identified a preference for Specialist Nurse led telephone appointments. However, it was felt that alternate appointments with standard care were likely to be more acceptable to patients, given patients assumed need for frequent Consultant led appointments (Lydon et al., 2009). In support of these findings, a cross-sectional study with Gynaecologists and Oncologists and two Specialist Nurses, identified that 56% (n=19/34) of Health Care Professionals felt that the Specialist Nurse should be involved with the delivery of care (Kew et al., 2007). However, only 15% (n=5/34) of these Health Care

Professionals strongly agreed and 33% (n=11/34) agreed, that it was the role of the Specialist Nurse to try and detect recurrence. Health Care Professionals were in more agreement that it was part of the Specialist Nurses role to listen to concerns (76%, n=25/34 strongly agreeing, 24%, n=8/34 agreeing) and answer questions (56%, n=19/34 strongly agreeing and 38%, n=13/34 agreeing: Kew et al., 2007).

2.4.3.6 General Practitioner involvement

Care led by the General Practitioner was also identified as being a viable option in the changing model of care. The focus group study identified that care led by the General Practitioner would 1) reduce the burden on secondary care clinics, and so 2) increase the quality of care for patients that needed regular appointments in secondary care, and 3) help to solve the problem of Consultants not wanting to ask patients lots of questions in case it unearthed non-cancer issues that should be addressed in primary not secondary care. Health Care Professionals explained that all of these options would be possible if patients received CA125 testing prior to their telephone or General Practitioner appointment (Lydon et al., 2009). It must be acknowledged that this was the general consensus of seven secondary Health Care Professionals in one treating hospital. In a quantitative cross-sectional questionnaire to identify preferences for gynaecology cancer follow-up generally, only 24% (n=8/34) of Health Care Professionals felt that the patients General Practitioner should be involved with the delivery of follow-up care, with 73% (n=25/34) of Health Care Professionals having a preference for care to be delivered by a Doctor in secondary care (Kew et al., 2007).

2.4.3.7 Conclusion

From the empirical studies of Health Care Professionals preferences identified in the systematic search and explored above, it is demonstrated that Health Care Professionals felt that the aim of follow-up care was to detect any recurrence, monitor effects of treatment and treat any side effects of treatment. Given this, and due to the lack of efficacy of traditional follow-up appointments and the resulting unnecessary burden on hospital clinics, Health Care Professionals valued the changing model of care. Within the changing model of care, Health Care Professionals generally valued the 'self-management' model and 'multidisciplinary' models of care, the later with involvement from the Specialist Nurse and/or General Practitioner. However, consultant gynaecologists in Scotland valued regular follow-up

appointments over a long duration for patients diagnosed with vulvar cancer. This is unsurprising given the higher incidence of asymptomatic recurrence in vulvar cancer (Oonk et al., 2003) and the high rate of cure if the recurrence is localised (Salom & Penalver, 2002).

2.4.4 Comparative synthesis between patients and Health Care Professionals preferences

Analysis of studies conducted in the United Kingdom using the framework approach (Richie & Spencer, 1994) identified similarities and differences between patients and Health Care Professionals preferences for gynaecology cancer follow-up, in terms of theme structure and theme content. Patient and Health Care Professional themes related to overall models of care, rather than individual attributes and attribute levels that comprised these. This may be attributed to the deductive nature of studies identified, allowing for generation of data which matched the researchers and policy makers agenda, rather than patients'. Themes identified from the exploration of data conducted from the perspective of patients were: 'hospital follow-up', 'Specialist Nurse led telephone follow-up', 'General Practitioner led follow-up' and 'self-management'. Themes identified from the exploration of data conducted from the perspective of Health Care Professionals were: 'changing model of care' with themes 'self-management' and 'multidisciplinary care', the latter which had sub themes 'Specialist Nurse involvement' and 'General Practitioner involvement'. Differences in themes between exploration of patient and Health Care Professionals preferences are reflected in the policy need for a changing model of care as identified from the synthesis of Health Care Professionals data. Interestingly, whilst themes relating to the Specialist Nurse and General Practitioner were identified from the patient and Health Care Professionals perspective, from the patient perspective this was to be led by these respective professionals, whereas from the Health Care Professionals perspective this was involvement from these professionals so using a more multi-disciplinary approach. Given the importance of preference misdiagnosis in health care, the finding that some patients and Health Care Professionals stated a preference for the self-management model of care, but that Health Care Professionals assumed that this model of care would be unacceptable to patients, is of particular interest.

2.4.5 Comparative synthesis between preferences for care and standard care

Patients had differing preferences for the model of care they valued most in detecting recurrence and providing reassurance in the absence of signs of recurrence. Some patients

had a preference for the standard model of follow-up care, whilst others had a preference for alternative models of care. However, Health Care Professionals had a preference for a changing model of care as opposed to standard care.

2.4.6 Comparative synthesis between preferences for care and systematic reviews of effectiveness and cost effectiveness

There are three current Cochrane reviews of gynaecology cancer follow-up. One of these reviews is of ovarian cancer follow-up (Clarke et al., 2014), one of cervical cancer follow-up (Lanceley et al., 2013) and one of patient reported outcome measures for gynaecology cancer follow-up. Two of these reviews identified no primary papers (Lanceley et al., 2013; Nama et al., 2013), and one identified that routine CA125 testing as part of hospital follow-up resulted in earlier treatment with no survival benefit, compared to commencement of treatment when relapse was detected based on symptoms (Clarke et al., 2014). Studies identified in this chapter, assumed the importance of CA125 testing (Cox et al., 2008; Lydon et al., 2009; Palmer et al., 2006), and in one study patients were educated on this (Palmer et al., 2006). However, findings from this Cochrane review question the need for CA125 testing in terms of the effectiveness and so cost effectiveness of ovarian cancer follow-up (Clarke et al., 2014). CA125 testing is no longer part of routine follow-up appointments in the United Kingdom (Leeson et al., 2017).

2.5 Discussion

The aim of this chapter was to systematically identify, extract, appraise and explore the existing literature on patients, their informal caregiver's and Health Care Professionals preferences for gynaecological cancer follow-up care. Within this, the specific objectives were to:

1. Identify existing literature on patients, their informal caregivers and Health Care Professionals preferences for gynaecology cancer follow-up care.
2. Critically appraise and explore existing research conducted in the context of the United Kingdom.

3. Explore whether patients, their informal caregivers and Health Care Professionals had similar or differing preferences for gynaecological cancer follow-up.
4. Explore how patients, their informal caregivers and Health Care Professionals preferences for the model of gynaecology cancer follow-up compare to the standard model of care.
5. Explore how patients, their informal caregivers and Health Care Professionals preferences for the model of gynaecology cancer follow-up, compare to research regarding the effectiveness and cost effectiveness of gynaecological cancer follow-up.
6. Identify whether further research is needed.
7. If required, use the attributes and attribute levels valued by patients, their informal caregivers and Health Care Professionals to aid the design of a subsequent discrete choice experiment (chapter 4-5).

The systematic literature review identified 19 studies of stakeholders' preferences for care. Fourteen studies explored patients' preferences, no studies explored informal caregivers' preferences, three studies explored Health Care Professionals' preferences, and two studies explored patients and Health Care Professionals' preferences. These studies were used to form the descriptive map (objective 1). Of these, five studies of patients' preferences were conducted in the United Kingdom, one study of Health Care Professionals preferences was conducted in the United Kingdom, and two studies of patients and Health Care Professionals preferences were conducted in the United Kingdom. These studies were used in the thematic synthesis (objective 2).

This review identified that patients and Health Care Professionals preferences for gynaecology cancer follow-up, have both similarities and differences (objective 3). Patients felt the aim of follow-up care was to detect recurrence and then provide reassurance that there was no sign of disease. Patients had preferences for differing models of care to meet this need, with preferences for hospital follow-up, Specialist Nurse led telephone follow-up, self-management and follow-up with the General Practitioner. Whereas Health Care Professionals felt that the aim of follow-up was to detect recurrence, and monitor and treat any effects of treatment. Given the need for a changing model of care, Health Care Professionals had a preference for follow-up to be delivered via self-management or within

the multi-disciplinary team via involvement from the Specialist Nurse and/or General Practitioner (objective 2-3). The low number of studies identified, the general deductive nature of studies and lack of exploration of why preferences differed, and the general poor level of reporting of participant demographics, meant that whilst the synthesis aimed to explore why preferences differed between demographics, this exploration was not possible (objective 2).

The systematic literature review identified that in relation to standard care, some patients valued the current model of care, whilst others had a preference for alternative models of care (Specialist Nurse led telephone follow-up care; General Practitioner led care; self-management). Whereas Health Care Professionals had a preference for a changing model of care (either self-management; multidisciplinary care with Specialist Nurse and/or General Practitioner involvement: objective 4).

A comparison of the preferences for care, to research regarding the effectiveness and cost-effectiveness of gynaecology cancer follow-up yielded little benefit, as two of the three current Cochrane reviews identified no empirical papers (Lanceley et al., 2013; Nama et al., 2013). Cochrane reviews were chosen as they are the gold standard for systematic reviews in health care (Smith, 2013). This review identified that patient and Health Care Professionals have assumed importance of CA125 testing in ovarian cancer follow-up. However, research regarding the effectiveness and cost effectiveness of routine CA125 testing identified no survival benefit and a negative impact on quality of life, compared to symptomatic detection of recurrence (Clarke et al., 2014: objective 5).

This systematic literature review identified that further research is needed, as:

- 1) Lack of exploration of why preferences differed within studies and poor reporting of demographics, hindered exploration of differences in preferences between studies and participant demographics.
- 2) The quality of studies was not high.
- 3) The deductive approach generally taken meant that attributes valued by the researcher and policy makers, not participants were generally explored in the primary papers.
- 4) Data saturation was not reported and there was a small sample size in the qualitative studies.

- 5) Two patient studies focused on CA125 testing which is no longer undertaken as part of standard care (Leeson et al., 2017).

Given the above, the conclusions that can be taken from this systematic literature review are limited. Therefore further research is required (objective 6).

The systematic literature review identified that no discrete choice experiments to explore preferences for gynaecology cancer follow-up had been conducted. The descriptive map was used to aid design of the discrete choice experiment (objective 7: chapter 4-5).

2.5.1 Previous findings

Prior to this review, a review of quality of life, needs and preferences for gynaecology cancer follow-up had been published. However, the search for preferences for care was not systematic and studies referenced in this section of the literature review include the systematic reviews of preferences for follow-up in cancer care generally, as well as empirical studies of preferences for breast cancer follow-up care (Dahl et al., 2013). There is one known published systematic review of preferences for follow-up in a general cancer population (Lewis et al., 2009b). The findings of this chapter build on the findings from this systematic literature review, which identified that fear of recurrence governed preferences for care with patients reassured at the follow-up appointment, but this anxiety returned at the next appointment (Lewis et al., 2009b). A preference for psychosocial support and alternative models of care was also identified (Lewis et al., 2009b). Interestingly, Lewis and colleagues (2009b) report that Health Care Professionals acknowledged that patients were not informed of the aims and limitations of the current model of care, and if they were to be informed, they would likely have different preferences for care. However, Lewis and colleagues (2009b) did not synthesise the studies of patients and Health Care Professionals preferences separately, limiting the comparisons that could be made (Lewis et al., 2009b). This chapter synthesises the findings by stakeholder separately, as well as being the first systematic literature review of preferences for gynaecology cancer follow-up care.

Unsurprisingly, a recent literature review and opinion paper exploring the future of gynaecology cancer follow-up care echoes the findings of this chapter regarding Health Care Professionals preferences for care. However, states that given the lack of efficacy of

gynaecology cancer follow-up care on survival, the aims of gynaecology cancer follow-up care should be modified prioritising the outcomes psychological morbidity and quality of life, given the effects of the diagnosis, disease and treatment reported in the follow-up period (Leeson et al., 2017).

The finding that patients and Health Care Professionals have both similar and differential preferences for care is supported by findings from a systematic review of patients and Health Care Professional's preferences for care (Harrison et al., 2017).

2.5.2 Strengths and limitations

This was the first systematic review of patients, their informal caregivers and Health Care Professionals preferences for gynaecology cancer follow-up. Importantly, this review identifies and presents all the available research in the form of a descriptive map, mapping the existing research. A synthesis of studies conducted in the United Kingdom adds to the knowledge base of preferences for care within the NHS.

A strength of this systematic review is the rigorous and evidenced based methods used at each stage of the reviewing process (Gough et al., 2012). This ensured for a robust review in accordance with the aims, objectives and epistemological viewpoint. This review was exploratory, using a configurative and aggregative approach. This is novel for systematic reviews conducted for the purpose of health economic research, which are traditionally aggregative in nature. The transparent application of methods used and the clear benefit of these methods, encourages and enables methodological replicability for future health economic systematic reviews exploring preferences for care.

A further strength of this systematic literature review is the use of the Weight of Evidence Framework to critically appraise studies which met the inclusion criteria (Gough, 2007; Torgeson et al., 2008). Use of an explicit multi-stage process to assess risk of bias using established critical appraisal tools, and suitability of the empirical papers using criteria stated apriori, aided transparency in this process.

A limitation of this systematic review is the dearth of evidence and quality of evidence identified. The studies generally had poor reporting of participant demographics restricting the comparison that could be made between studies to explore potential variables of preferences for care. No studies were assessed overall as being of high methodological quality,

and five studies were assessed overall as being of low quality (Bradley et al., 1999; Bradley et al., 2000; Cox et al., 2008; Lydon et al., 2009; Palmer et al., 2006). Therefore, the findings from this systematic literature review are limited and have to be taken and applied with caution.

An objective of this review was to inform the subsequent phases of this PhD thesis. Methodological challenges arose at the stage of the systematic review protocol, and due to available resources there was a need to design the subsequent studies whilst these methodological issues were addressed. As a result the review was designed alongside the design of the qualitative study, and the searches conducted, data extracted, appraised and descriptive map formed after the qualitative research study but before the discrete choice experiment, and the synthesis conducted after the discrete choice experiment. The systematic literature review informed the design of the discrete choice experiment (chapters 4-5), therefore it was important that the searches were not updated.

2.5.3 Future Research

This systematic review highlights that little is known about patients and Health Care Professionals preferences for gynaecology cancer follow-up in the context of the United Kingdom, and that nothing is known about informal caregivers preferences for gynaecology cancer follow-up.

Given the limited research conducted into patients, their informal caregivers and Health Care Professionals preferences for gynaecology cancer follow-up within the United Kingdom, further research is needed. This is of particular importance given the policy emphasis on a changing model of care due to the lack of evidence for the current model of care and its cost to the National Health Service, and strategic focus on prudent health care in Wales (National Cancer Survivorship Initiative, 2013; Welsh Government & NHS Wales, 2016). Such research needs to be of a high methodological design to reduce the effect of bias and increase the validity and reliability of the results. Both an inductive and deductive approach needs to be taken to ensure that plausible and policy relevant attributes of care are identified whilst valuing patients and their informal caregivers' experiences and preferences. Such research should be conducted in a population of gynaecology cancer survivors generally, given the similarities in the presentation of recurrence, similarities in the model of follow-up care delivered and focus on a changing model of care for gynaecology cancer generally (Fung-Kee-Fung et al., 2006; Guerts et al., 2012; Leeson et al., 2017; Leeson et al., 2013; Lim et al., 2004).

2.6 Conclusion

This systematic review aimed to identify, appraise and explore the existing literature on patients, their informal caregivers and Health Care Professionals preferences for gynaecological cancer follow-up. This systematic review identified that patients felt the aim of follow-up care was to detect recurrence and provide reassurance in the absence of disease detection, whilst Health Care Professionals felt the aim of follow-up was to detect recurrence and discuss concerns. Patients had preferences for different models of care to meet this need: hospital follow-up, Specialist Nurse led telephone follow-up, self-management and follow-up with the General Practitioner. Given the need for a changing model of care, Health Care Professionals valued the self-management model of care and multidisciplinary models which included Specialist Nurse and/or General Practitioner involvement. No studies of informal caregivers' preferences were identified. Future research should explore patients, their informal caregivers and Health Care Professionals preferences for gynaecology cancer follow-up within the United Kingdom, using both an inductive and deductive approach, and be conducted and reported to a high standard.

2.7 Novel Contributions

2.7.1 Methodological contributions

- This systematic literature review aims to explore using a configurative and aggregative approach in accordance with the guidance produced by the EPPI centre. In accordance with this guidance there is not a 'one size fits all' method of conducting a systematic review, instead the methodologies used should be dependent on the aims and objectives of the review. The conduct of an exploratory review is novel in health economics, where traditionally systematic reviews aim to aggregate not configure.
- The transparent reporting of the methodology of this review aids the design of future systematic reviews of preferences for care within health economics, when there is a need to configure not merely aggregate.
- Deciding upon search terms in this systematic literature review was a particular challenge due to the indexing of relevant papers. For example, some research studies exploring follow-up care did not include any terms that relate to follow-up care in its heading, title or abstract, but were found by searching terms such as 'nurse' (see methodology). Previous reviews of follow-up care have restricted their search by terms related to the intervention not setting, and so may not have identified all relevant studies (as indicated by the review being conducted by Aslam et al., 2016). This systematic review highlights the need for more extensive scoping searches in the development of the search criteria.

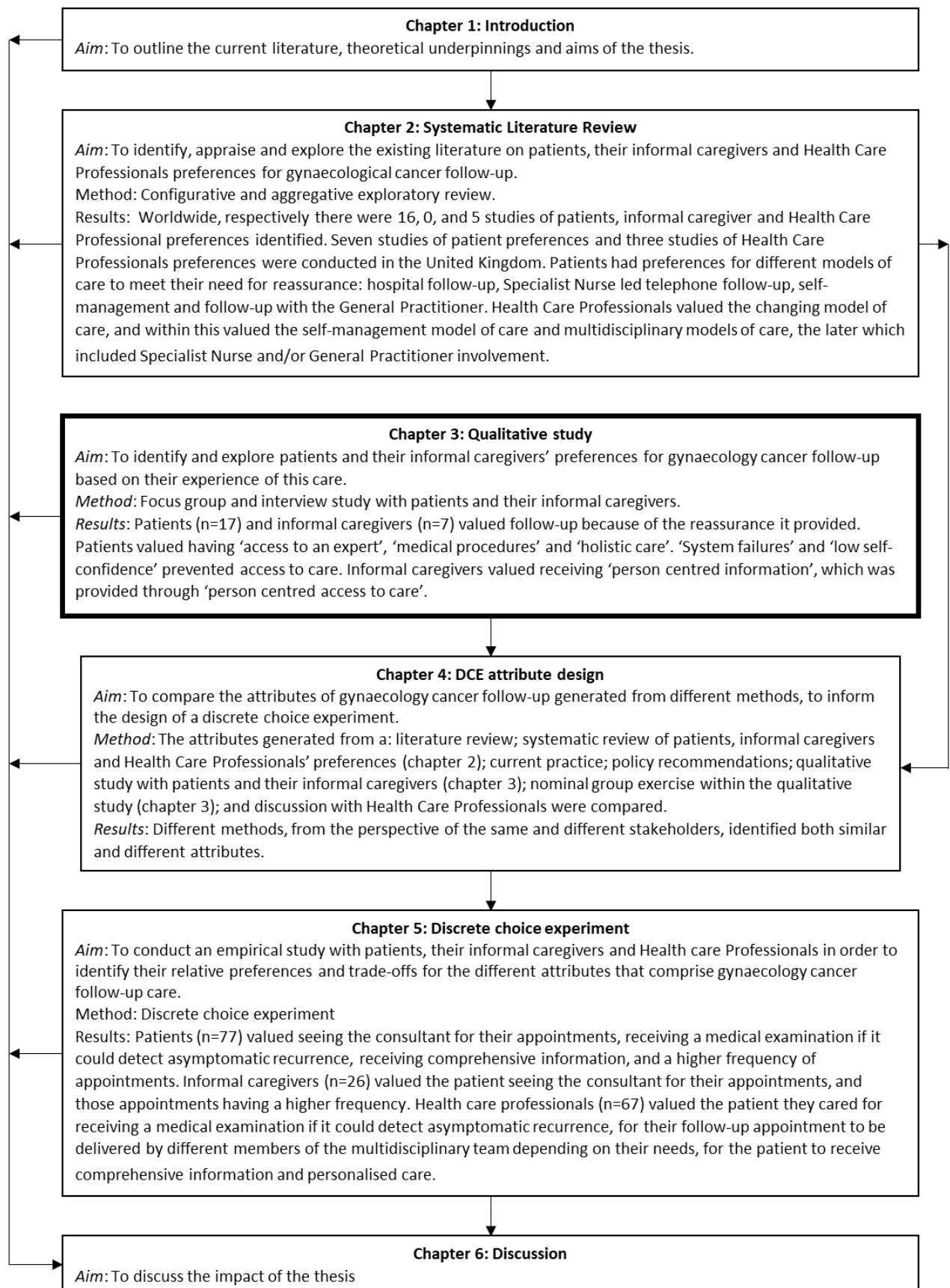
2.7.2 Policy contributions

- There are no previous systematic reviews of patients, their informal caregivers and Health Care Professionals preferences for gynaecological cancer follow-up care.
- Systematic literature reviews are used by policy makers to aid decision making. The systematic identification, extraction, appraisal and exploration of the existing literature can be used by policy makers to aid in their decision making for the prudent changing model of care.
- It is particularly important to address this research question given the increasing population of gynaecological cancer survivors, finite resources of the health care system, and efficacy of the current model of care in detecting asymptomatic

recurrence, increasing survival and meeting patients and their informal caregivers' needs.

Chapter 3:

**“To know that there is a light at the
end of the tunnel”: Patients and
their informal caregivers
preferences for gynaecology cancer
follow-up, a focus group and
interview study**



3.1 Chapter Summary

Chapter 1 identified a dearth of high quality evidence into patients and their informal caregiver's preferences for gynaecology cancer follow-up, despite the importance of this from a policy perspective. The study described in this chapter sought to identify and explore patients and their informal caregivers preferences for gynaecology cancer follow-up, based on their experience of this care, and use the attributes generated to inform the design of a subsequent discrete choice experiment (Chapters 4-5). A cross-sectional focus group or interview was held with 17 patients (n=4 groups with 12 participants, range of participants per group=2-5, and n=5 interviews), and 7 informal caregivers (n=1 group with 3 participants, n=4 interviews). Data generated was analysed using the Framework approach. Patients' preferences were governed by the need for reassurance that there was no sign of recurrence. Patients wanted their needs to be met by having access to an expert and the provision of holistic care, because of the reassurance this provided. Patients experienced barriers to accessing care and so the reassurance that this provided, through system failures and low self-confidence. Informal caregivers preferences for care were also governed by a need for reassurance that the patient had no sign of recurrence. This need was met through the provision of person centred information, which in turn was met by the provision of person centred access to care. The emotional impact of caregiving had an effect on informal caregivers' preferences for care. These results can be used to help improve patients and their informal caregivers' outcomes, and be used to aid the design of a subsequent discrete choice experiment.

3.2 Introduction

An aim of the thesis was to identify patients and their informal caregivers preferences for gynaecological cancer follow-up care. Only two studies conducted in the United Kingdom have explored patients' preferences for gynaecological cancer follow-up using qualitative research methodology: one of these held one focus group with ovarian cancer follow-up patients (Lydon et al, 2009), and the other used a grounded theory approach so was purely inductive, not exploring all attributes and levels that were policy relevant (Bradley et al., 1999). No known research has explored informal caregivers' preferences for gynaecological cancer follow-up care despite the role informal caregivers play in assisting patients navigate the health care system and attend health care appointments (Williams et al., 2013; Wolff & Roter, 2011). The support provided by informal caregivers has shown to effect the patients quality of life (Courstens, et al., 1996; Leung, et al, 2014; Luszczynska, et al., 2013).

There is a policy focus on understanding patients and their informal caregivers' preferences for care (Department of Health, 2012; Mulley et al., 2012). To understand preferences for care, it is important that underlying illness representations, experiences, attitudes and beliefs are understood. These factors explain elements of decision making which are not described by utility models. Illness representations, experiences, attitudes and beliefs are explained by psychological models of health behaviour. The most commonly applied social cognitive models to understanding and predicting behaviour are the Health Belief Model (Becker, 1974; Becker & Rosenstock, 1984; Rosenstock, 1974: Figure 5) and Theory of Planned Behaviour (Ajzen, 1985; Ajzen, 1991: Figure 6). The most commonly applied model of illness appraisal is the Self-Regulation Model (Leventhal et al., 1992: Figure 7).

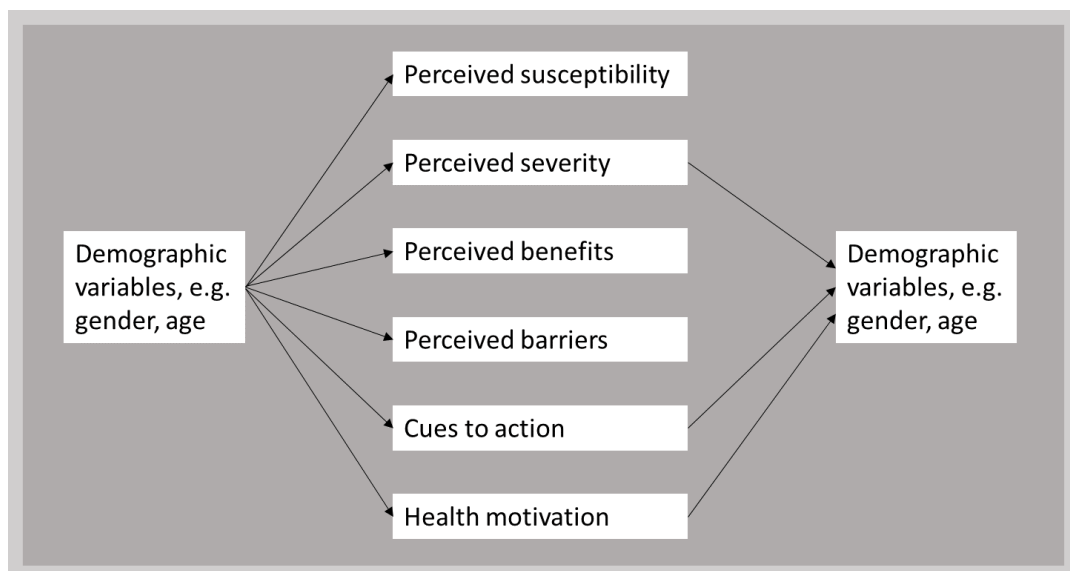
3.2.1 *Health Belief Model*

The Health Belief Model (Becker, 1974; Becker & Rosenstock, 1984; Rosenstock, 1974: Figure 5) is derived from the theory of subjective expected utility (Edwards, 1954), that individuals are rational decision makers who made decisions based on the option that has the highest level of perceived utility. In accordance with the Health Belief Model, demographic variables, for example, the individuals' age, directly predict behaviour, and indirectly predict behaviour through beliefs regarding the perception of the threat (perceived susceptibility; perceived

severity), evaluation of the behaviour (perceived benefits; perceived barriers), cues to action (added by Becker & Maiman, 1975) and motivation for health (added by Becker et al., 1977).

The Health Belief Model has been applied to understanding screening for breast and cervical cancer (for example, Tanner-Smith & Brown, 2010) and engagement in cancer follow-up (Cardella et al., 2007). Interventions developed on the model of Health Belief to increase screening behaviour have shown to increase perceptions of threat of the disease, behavioural evaluations of screening, cues to action for engagement in screening, motivation as well as behaviour (Kolutek, Avci & Sevig, 2016).

Figure 5: Health Belief Model (Becker, 1974; Becker & Rosenstock, 1984; Rosenstock, 1974)



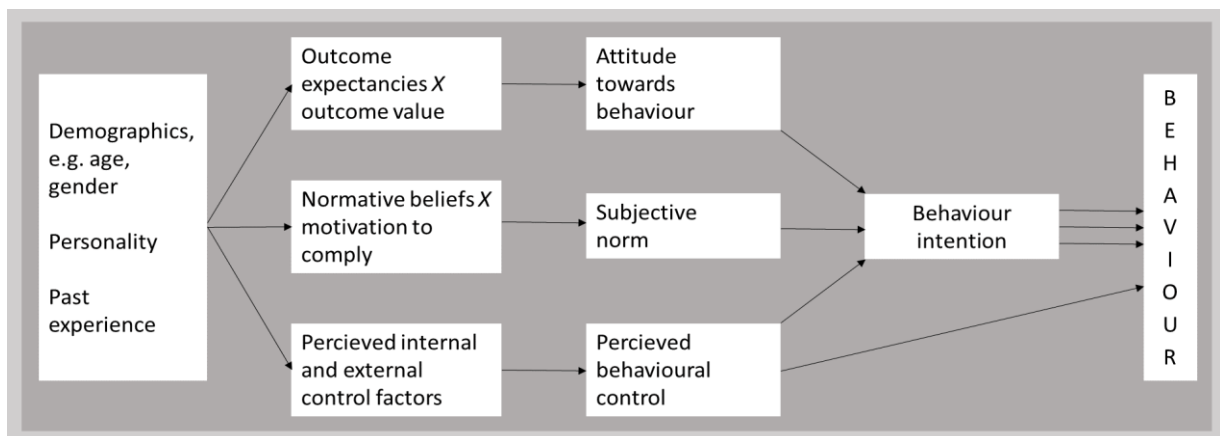
Cardella et al (2007) applied the Health Belief Model to explore the attitudes, barriers and compliance with hospital follow-up following surgery for colorectal cancer, from the perspective of patients and Health Care Professionals. Similar to gynaecological cancer follow-up, colorectal cancer follow-up aims to detect recurrence but unlike in gynaecological cancer, follow-up has shown to increase survival (Figueredo et al., 2003; Jeffery Hickey & Hider, 2002; Renehan, Egger, Saunders & O'Dwyer, 2002).

3.2.2 Theory of Planned Behaviour

Whilst the Health Belief Model assumes that behaviour is a result of cognitions, the Theory of Planned Behaviour (Ajzen, 1985; Ajzen, 1991: and previously the Theory of Reasoned Action, Ajzen & Fishbein, 1970) more explicitly acknowledges the effect of social context on cognitions

and the behaviour itself. The Theory of Planned Behaviour proposes that behaviour is a result of a behavioural intention and perceived behavioural control (an individuals' belief that they can control their behaviour). Behavioural intention reflects attitude toward the behaviour (which is a reflection of beliefs about the outcome, for example, attending follow-up will detect any recurrence, and the value of that outcome, for example, it is important to detect any recurrence); subjective norm, defined as perceived social norms (which may elicit social pressure, and is a reflection of the perception of the beliefs that others have, for example, my husband thinks I should attend follow-up, and the individuals motivation to comply with this, for example, I would like to do what my husband wants me to do); and perceived behavioural control (a reflection of perceived internal and external control factors, for example, I have had a telephone follow-up appointment before and it assessed me for any recurrence, and I believe it will at the next telephone appointment). In accordance with this model, these determinants of behavioural intention are influenced by an individual's demographics, personality and past experience (see Figure 6).

Figure 6: Theory of Planned Behaviour (Ajzen, 1991; Ajzen, 1985)



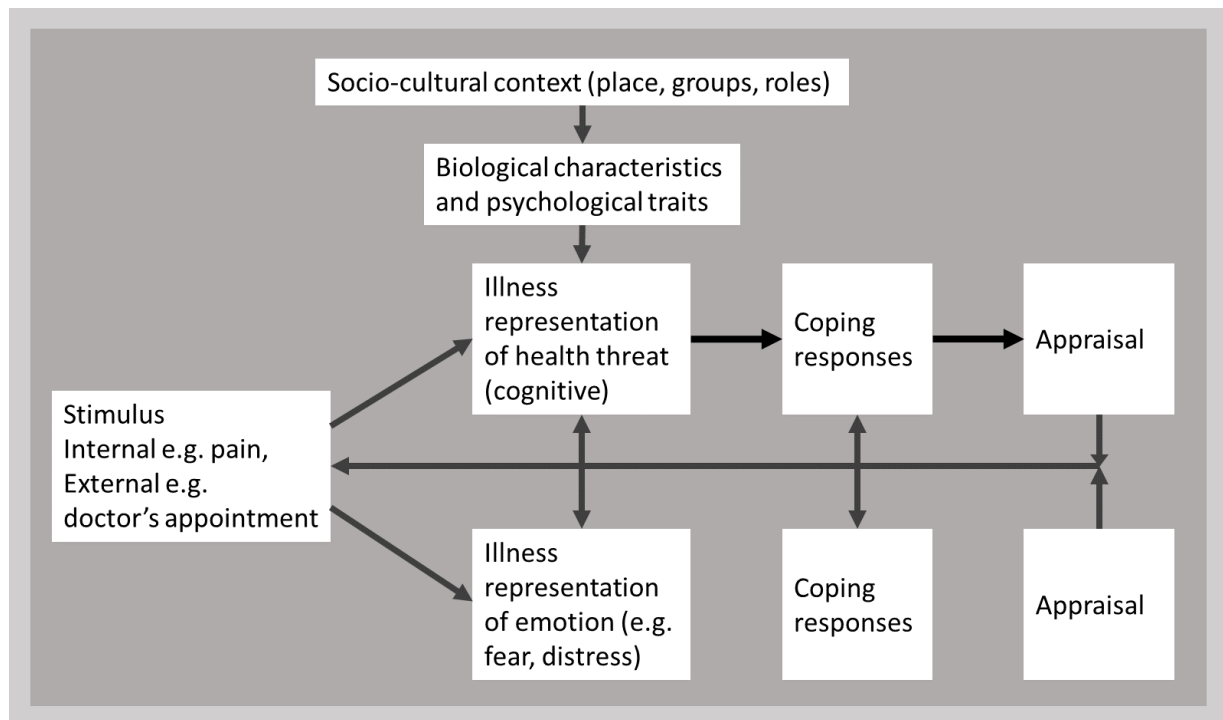
The Theory of Planned Behaviour has been applied to predict screening behaviour (Cook & French, 2008), and used to design interventions to increase intentions for, and the uptake of, cervical cancer screening (Sheeran & Orbell, 2000), as well as to increase attendance at General Practitioner led follow-up for colorectal cancer (Ngune, McManus & Parsons, 2015).

3.2.3 *The self-regulation model of illness*

Within health psychology the self-regulation model, also known as the 'common sense model', is the model most commonly employed within studies seeking understanding of the role illness beliefs play in illness self-management, or in other words coping with illness (Leventhal, Diefenbach & Leventhal, 1992: Figure 7). As demonstrated by Figure 7, the model theorises that in response to an internal (for example, bodily sign or change) or external stimulus (for example, cancer screening invitation), the individual forms or brings to memory a cognitive representation of the relevant symptoms, illness or disease, for example, "I am bleeding, it must be cancer" and the emotional response to this, for example, fear. This objective and subjective representation is referred to as illness representations. It has been identified through studies across many health conditions that there are five broad components to an individuals' cognitive representation:

- 1) Identity: defined as the symptoms that a person considers are attached to a labelled health threat/illness, for example fatigue, weight loss and cancer.
- 2) Cause: belief about the cause of the health threat/illness, for example, being overweight.
- 3) Timeline: perceived time frame that the health threat/illness will last for (chronic, acute or cyclical).
- 4) Consequences: beliefs about the possible effect of the health threat/illness on an individuals' life, for example, pain and disability, death/loss of life.
- 5) Curability/controllability: the belief of the extent that the threat to health can be cured or controlled, for example, cancer treatment will cure the cancer. This dimension has been amended to:
 - a. Personal control: the extent to which someone believes they have personal control over the illness, its symptoms, or its treatment for example, I can attend treatment/take my medication.
 - b. Treatment control: the extent to which someone believes that treatment can take control over the illness and/or its symptoms, for example, surgery might remove the cancer, or medication may manage my pain.

Figure 7: The self-regulation model of illness (Leventhal et al., 1992)



This model highlights that illness representations and coping responses are not static, but dynamic, and that illness regulations and coping can be amended, for example, when new symptoms arise or when the coping response does not have the desired effect. Sociocultural factors, for example, culture and cultural norms around illness, educational status, as well as biological characteristics (for example, genetics, family history, age) and psychological traits (for example, mood, neuroticism, optimism), as well as past experience with illness and healthcare, all combine to shape an individual's model of illness/ illness representation. This model provides a framework for understanding individual differences in illness representations, coping responses and appraisal to a stimuli (Leventhal et al., 1992) and has received significant empirical support.

Those experiencing the stimulus, for example, a cancer diagnosis, as well as the individuals' family, friends and wider society, all hold their own illness representations and this affects how each cope and appraise the stressor event (for example, diagnosis), as well as how they may interact with the direct recipient of the diagnosis (Onwumere et al., 2016; Song, Rini, Ellis & Northouse, 2016; Wu, Mohammed, Winkel & Diefenbach, 2013).

Qualitative research allows researchers to explore, provide insight, map the range and understand phenomenon (Krueger & Casey, 2009).

The conduct of qualitative research to aid the design of a discrete choice experiment is recommended, as it improves the content validity (see Chapter 4 for further discussion: Bridges et al., 2011; Coast & Horrocks., 2007; Coast et al., 2012; Kløjgaard et al., 2012; Louviere, Hensher & Swait, 2000; Lancsar & Louviere, 2008). Therefore, the primary aim of this study was to identify and explore patients and their informal caregivers' preferences for gynaecology cancer follow-up, based on their experience of this care. The secondary aim was to use the attributes and attribute levels identified, to aid the design of a subsequent discrete choice experiment. Within this, the specific objectives were too:

1. Develop an understanding of the attributes of care valued by patients and their informal caregivers.
2. Develop an understanding of why these attributes are valued as they are.
3. Identify attribute levels for each of the identified attributes.
4. Identify and understand hypothetical willingness to pay thresholds for gynaecology cancer follow-up care.
5. Identify and understand how long patients and their informal caregivers would be willing to wait between follow-up care appointments.
6. Develop an understanding of how patients and informal caregivers feel that follow-up care could be improved.

3.3 Methods

3.3.1 Design

The study was approved by Bangor University and NHS ethics and R&D committees. Patient and informal caregiver representatives aided with the design of the recruitment documentation to confirm readability and ease of understanding (INVOLVE, 2012).

A cross-sectional semi structured focus group or interview was undertaken with 17 patients (n=4 groups with 12 participants, range of participants per group=2-5, and n=5 interviews), and 7 informal caregivers (n=1 group with 3 participants, n=4 interviews). The aim of the focus groups and interviews were to explore the aspects of NHS delivered

gynaecological cancer follow-up care that patients and their informal caregivers valued based on their experience of this care.

Focus groups were chosen as 1) focus groups encourage participants to be open and honest about things that they do not normally talk about, particularly as group participants shared an experience of gynaecological cancer and follow-up, or experience of providing care for someone with gynaecological cancer, 2) group discussion encourages individual reflection and discussion of things previously not thought about, 3) through such discussion preferences and the rationale for this can be seen as they unfold, given the need to negotiate and defend within the social group context (Barbour, 2008; Crabtree et al., 1993; Kitzinger & Barbour, 1999; Krueger & Casey, 2009). Separate focus groups were held for patients and their informal caregivers for two reasons. Firstly, patients and informal caregivers have different illness representations and experiences and may have different attitudes and beliefs and so preferences for care (Janda et al., 2008; Leventhal et al., 1992; Reamy et al., 2011; Sheldon, Davis, & Parsons, 2008; Shin et al., 2013; Walker et al., 2015). Secondly, the dynamics of interpersonal relationships can mean that discussion is dominated by one person in the relationship whilst the other remains silent, even if the participant who is silent disagrees with what is said (Krueger & Casey, 2009).

Interviews were held with participants who were unable to make any focus group because of other commitments, comorbidities, or geographical constraints of gathering groups across wide rural areas. Two participants elected to have a telephone interview for these reasons. The same protocol was delivered to participants who received a focus group, interview or telephone interview. The use of interviews where necessary ensured that the views, experiences and beliefs of participants often otherwise excluded from focus group research, were understood (Barbour, 2008; Thompson et al., 2003a; Thompson et al., 2003b). One informal caregiver chose for the patient they provided care for to be present in the interview.

3.3.2 Participants

Gynaecological cancer patients (and their respective informal caregivers) who were attending follow-up after treatment for endometrial, ovarian or cervical cancer at the recruiting hospitals, who had attended at least one follow-up appointment, were free from disease at their last follow-up appointment, were over 18 years of age and could speak English were

eligible to take part in the study. Participants were excluded if they did not have the capacity to consent.

Patients were purposefully sampled (Kuzel, 1992) to ensure that the sample was reflective of the patient population in terms of cancer site prevalence and five year survival rate, given that the overarching aim of this PhD is to influence policy.

3.3.3 Recruitment

One hundred and sixty-four patients were screened for eligibility at their gynaecology, oncology or radiology follow-up appointments by their Health Care Professional (Consultant/Registrar/Specialist Nurse), within the recruiting hospitals between June and October 2014. Sixty-five patients were eligible to take part, of these, 57 patients were given the recruitment packs (the reason for the recruitment pack not being given was not provided by the screening Health Care Professional). Informal caregivers were opportunistically recruited to the study: they were identified and given the informal caregiver recruitment pack by the patient they provided care for. Patients were not excluded if their informal caregiver did not consent to take part in the study.

Twenty-four patient consent forms were returned to the researcher, and 17 patient participants took part in either a focus group or interview. Seven patient participants withdrew (n=1 had a recurrence, n=2 co-morbidities, n=1 last minute childcare responsibilities, n=1 accessibility, n=2 no response). It is unknown how many informal caregivers were invited to take part in the study. Ten informal caregiver consent forms were returned to the researcher, and 7 informal caregivers took part in a focus group or interview. Three informal caregivers withdrew (n= 1 patient had recurrence, n=1 informal caregiver was unwell, n=1 did not show). In the population of endometrial, cervical and ovarian cancer patients receiving gynaecology cancer follow-up, 57% are diagnosed with endometrial cancer, 27% are diagnosed with ovarian cancer, and 17% are diagnosed with cervical cancer (Office for National Statistics, 2013). Comparatively, of the sample recruited to the study, 47% were diagnosed with endometrial cancer, 24% were diagnosed with ovarian cancer and 29% were diagnosed with cervical cancer (see Table 10). Therefore the sample was under representative of endometrial cancer patients and over representative of cervical cancer patients compared to the population.

3.3.4 Materials

3.3.4.1 Materials for participants

The recruitment booklet contained a research invitation letter; information sheet; request for more information; two copies of the consent form; demographic questionnaire and two freepost envelopes (see appendices). On the participant information sheet, (potential) participants were informed that the researcher was undertaking the research as part of her PhD studies. Each material was printed on a different coloured piece of paper to reduce participant burden (Bray, 2015). The debrief form was given to participants following the focus group or interview. The materials followed the guidance outlined in the Good Clinical Practice Guidelines (National Institute for Health Research, 2013), and were printed on study letter headed paper. The research invite letter was printed on the health board's letter headed paper and, with an aim to increase recruitment rates, signed by the prospective participant's Consultant (Krueger & Casey, 2009).

An audio recording device was used to record the focus groups and interviews. Following each focus group and interview, the audio recording was uploaded to the researcher's encrypted university laptop and deleted from the recording device.

3.3.4.2 Materials for Health Care Professionals

An eligibility screening form (using a tick box format for ease and to reduce error), was used to screen each patient on the clinic list against the eligibility criteria, which, following

Table 10. Incidence, 5 year survival and recruitment prevalence

Cancer type	Population incidence rate	Incidence rate x 5*	Population 5 year survival	Number of patients alive 5 years post diagnosis	Percentage of patients in follow-up	Sample recruited n (%)
Endometrial	8,984 per year	44920	79%	35487 alive at 5 years post diagnosis	57%	8 (47%)
Ovarian	7,270 per year	36350	46.2%	16794 alive at 5 years post diagnosis	27%	4 (24%)
Cervical	3,126 per year	15630	67.4%	10535 alive 5 years post diagnosis	17%	5 (29%)
Total	19,380 per year	96900		62816 alive 5 years post diagnosis	101%	17 (100%)
*Incidence rate x 5 as patients are traditionally followed up for 5 years within the local health board						

completion, was stored in the patients' file. Recruiting Health Care Professional's transferred the number of patients screened, given recruitment documents to and consented, to the 'Screening, eligibility and recruitment pack record' following each clinic. This was given to the researcher for monitoring and recording recruitment.

3.3.4.3 Materials for Chief Investigator

Each contact between the researcher and potential participant/participant was recorded on a contact form, then transferred to the potential participants/participants case report form as per good clinical practice (National Institute for Health Research, 2013).

3.3.5 Measures

3.3.5.1 Demographic questionnaire

The demographic questionnaire was split into three sections. The first section contained questions to ascertain contact details and information regarding informal caregiver presence and caregiving duration. The second section contained four questions to identify key socio-demographic variables. The third section (patient questionnaire only) contained 12 questions with an aim of identifying key variables relating to the patient's diagnosis, treatment and follow-up care received. Tick boxes were provided where possible. Free text lines were provided where applicable (Boynton & Greenhalgh, 2004).

3.3.5.2 Semi structured interview guide

A semi structured focus group/interview guide was followed to ensure that the aims and research questions were addressed in each focus group and interview. In accordance with health economic and health psychology theory a top down (deductive) and bottom up (inductive) approach was used to explore the what (patients and their informal caregivers' preferences) and the why (why patients and their informal caregivers had these preferences: see Chapter 1 for general discussion, and Chapter 4 for discrete choice experiment attribute requirements: Abihiro et al., 2014; Ajzen, 1985; Ajzen, 1991; Rosenstock, 1974; Becker, 1974; Becker & Rosenstock, 1984; Coast & Horrocks, 2007; Coast et al., 2012; Hall, Vinney, Hass & Louviere, 2004; Hilgsmann et al., 2013; Leventhal et al., 1992; Mangham, Hanson & McPake, 2008; Powell et al., 2015). Participants were encouraged to reflect on their experiences of

care. Brief hypothetical case studies were used to encourage self-reflection and discussion (see below). Prompts, probes and follow-up questions were used to seek response clarification, encourage thought processes, elaboration of responses and discussion. There were three sections, which began after discussion of ground rules and conduct of an ice breaker (name and something interesting about yourself: Barbour, 2008; Krueger & Casey, 2009):

Section 1:

- 1) Participants were asked '*What is the purpose/aim of follow-up care?*' This aim of this question was to contextualise subsequent responses. Participant's responses were written on flipchart paper and stuck to the wall so that 1) the researcher could refer back to them and 2) participants had them as a point of referral when reflecting and responding to the following questions.

Health Economics questions (nominal group technique):

- 2) As a group, participants brainstormed what they thought the different aspects to follow-up care were. If the group struggled to identify different aspects the researcher gave prompts such as:

"Other groups/people thought that the person who delivered their care/how often their appointment was, was important to them".

In focus groups, responses were brainstormed as a group and displayed on the wall for referral back to if required. In interviews, this was written down by the researcher for referral back to by the researcher or participant if required.

- 3) In focus groups, participants were then asked to rank on their response sheet, what attributes were important to them in order of importance. For example, if they felt four attributes were important to them they listed these and ranked them in order of importance. This ensured that the voices of participants who did not feel confident discussing their preferences in the group situation were still 'heard'. This task was conducted verbally in the interviews.

- 4) In focus groups, participants were asked as a group to identify their 'Top 5' attributes in order of importance, for the purpose of designing the subsequent discrete choice experiment. Responses were listed and displayed on the wall for referral back to if required. This task was omitted in interviews as by its nature duplicated question 3.

Case studies:

- 5) Participants were presented with two case studies with the aim of encouraging patients and their informal caregivers to think about their own experiences of care and follow-up needs, and their attitudes, beliefs and preferences for care based on these experiences and needs. The case studies were designed to be 'opposite' to each other to encourage reflection and discussion (Finch, 1984; Barbour, 2008)

Case study 1: Maggie is fifty years old and was diagnosed with early stage endometrial cancer last year. She goes to see the Consultant every 3 months to have an internal examination to check that the cancer hasn't returned. Maggie found the first couple of weeks after her hysterectomy hard, but has since returned to work and is living the life she lived before.

Case study 2: Ann is seventy years old and was diagnosed with an advanced form of ovarian cancer four years ago. Ann goes to see the Specialist Nurse once a year for an internal examination to check that the cancer hasn't returned. Ann has struggled with daily living since she was treated for the cancer. Because of her treatment she has had problems with her bowel functioning, which she still suffers with.

After each case study was read out by the researcher, participants were asked: "*What do you think her needs are, and how do you think follow-up care can meet those needs?*"

- 6) Participants were then asked, thinking about these case studies,
 - a. *What is good about the follow-up care that you/the person you care for receives?*

- b. Are any of your needs/the person you care for's needs not met by their follow-up care? Which ones?*
- c. What aspects of follow-up care do you think could be improved (explain why)?*
- d. How long would you like between follow-up appointments?*
- e. Hypothetically, if you had to pay for follow-up after treatment care, how much would you be willing to pay per follow-up visit?*

It was hoped that the case study questions and discussion around these case studies would encourage reflection, and break down the stoic 'everything is fine' perspective. This was particularly important at the time of the conduct of the study given the political climate regarding the Welsh NHS not meeting care standards and receiving further financial cuts, and discussion around privatisation of the NHS in England (BBC News, 2014a: BBC News 2014b).

Prompts for further explanation and discussion were used, with specific prompting by eye contact and name to quieter members of the group to encourage elicitation of their preferences based on their experience of care. To ensure that participants' voices were valued, off topic discussion was not 'cut off' in accordance with guidance for the conduct of qualitative research. Instead the researcher used questions to bring the conversation back to the aims of the study (Krueger & Casey, 2009). For example, if participants discussed delayed diagnosis they were asked about their thoughts on their General Practitioner delivering follow-up care. This brought the discussion back to the topic of follow-up care, and helped participants think about their experiences in relation to their needs, attitudes, beliefs and preferences.

3.3.6 Procedure

On receipt of the returned recruitment documents the researcher telephoned the participant to acknowledge their receipt, to build up a rapport with the participant, and to answer any questions they had. Once sufficient consent forms were returned a letter was sent to participants with the time, date and location of the focus group. Participants were telephoned a couple of days before the focus group as a reminder of the group (Krueger & Casey, 2009).

Focus groups were held on the Tenovus Cancer Care mobile unit (a large and 'airy' mobile van with comfortable seating parked in an accessible location), with an aim to create a relaxed, open and supportive environment (see Figure 8 and Figure 9). Refreshments were

provided, and the option of travel expenses reimbursed (Krueger & Casey, 2009). Interviews were held in the participants' home or on the telephone to fit around these participants' co-morbidities and other commitments. Focus groups and interviews were audio recorded.

Before the focus group or interview, the researcher went through the information sheet and participants signed another copy of the consent form. Ground rules were made, an ice breaker conducted (Barbour, 2008; Krueger & Casey, 2009), and the semi structured interview guide followed. An observational researcher (TS/STY) was present at each focus group, to assist in welcoming participants, monitor the timing of the group, and make field notes (Barbour, 2008; Krueger & Casey, 2009).

Figure 8: Tenovus Cancer Care mobile unit

Copyright Tenovus Cancer Care



3.3.7 Ethical considerations

Given the possible sensitive nature of the discussion, the author was very mindful of potential distress arising from participation in the research study. In the ground rules, the right for each individual to be treated with respect, confidentiality, withdrawal and having a break from the discussion was conversed. An observational researcher and Tenovus cancer care employee was on hand in the focus groups to assist in managing the risk of distress if required. The researcher directed any requests for advice to the Tenovus Cancer Care staff member (to be discussed after the focus group), or to their Specialist Nurse (Krueger & Casey, 2009). The

Tenovus Cancer Care member of staff was not part of the group discussion and remained in the reception area, and did not have access to the data.

Figure 9: Tenovus Cancer Care Mobile unit (set up of 'room' for focus groups)



3.3.8 Data Analysis

Data was collected between August and October 2014. Focus groups ranged in duration from 58 minutes to 79 minutes (mean 70 minutes). Interviews ranged in duration from 23 minutes to 63 minutes (mean 48 minutes). Demographic data was analysed to explore the characteristics of the sample. Audio recordings were transcribed verbatim by LT/an independent transcriber, and checked for accuracy by TS or LT. No qualitative data management software was used. Patient data was analysed separately to informal caregiver data, then the dyadic data explored (where both a patient and their informal caregiver participated).

Following data collection the primary researcher immersed herself in the data and analysed the results iteratively using the Framework approach, allowing for an inductive and deductive analysis in accordance with the aims of the research study (Barbour, 2008; Krueger & Casey, 2009; Rabiee, 2004; Richie & Spencer, 1994: Table 11). Iterative discussion took place with VM throughout this process. Data saturation was reached.

The researcher was very aware of her responsibility to make each voice heard in the analysis and to do the data justice, whilst making sure that the research questions were

Table 11. Stages of data analysis using the Framework approach

<i>Familiarisation</i>	The aim of this phase is to become familiarised with the data (Richie & Spencer, 1994). The primary researcher immersed herself in all audio recordings, respective transcripts, the group brain storms and ranking exercises. The researcher brainstormed emerging themes from each unit of analysis whilst listening and re-listening to the respective audio recording and reading and re-reading the respective transcript. It is acknowledged that it is not always possible to include all data sources in the analysis due to the volume of qualitative data. A decision of what data to use should depend on 1) the methods used, 2) number of researchers, 3) the length of time available for analysis, and 4) heterogeneity of participant demographics (Srivastava & Thomson, 2009). It was apparent that several participants did not understand the individual ranking exercises, and more confident and vocal group members input was inevitably heard more with their reasons for rankings taking dominance in the group task. Additionally, the process of condensing the views of multiple participants into 'one ranking preference' by a group of people who have their own experiences and respective needs, attitudes, beliefs and preferences, is logistically impossible without de-validating individuals' perspectives. This task was used as it is recommended and used in health economic research (see Chapter 4), however as is illustrated, lacks validity (see Chapter 4).
<i>Identifying a thematic framework</i>	The aim is to identify emerging themes in the data (Richie & Spencer, 1994). The brainstorms from each focus group and interview were displayed around the researcher's office and read and re-read multiple times. A provisional thematic framework was deductively identified based on these themes. Following discussion with VM, a more deductive thematic framework emerged. This involved further iterative familiarisation, reflection and discussion. A small subset of transcripts were independently coded by TS to check for accuracy (Carroll, Owen-Smith, Shaw & Montgomery, 2012). No new themes were identified confirming data saturation and providing rigour (Krueger & Casey, 2009).
<i>Indexing</i>	The aim of this phase was to code each section of the data that relates to each theme (Richie & Spencer, 1994). Using the thematic framework, the researcher indexed all sections of each transcript that corresponded to each theme, using comment boxes in Microsoft word for ease (Richie and Spencer, 1994). The full name of the themes opposed to numerical codes were used to prevent confusion and keep meaning. Some units of text had multiple themes. The majority of text was coded highlighting that the thematic framework was a good fit for the data. As part of this process it became apparent that the thematic framework should be adjusted to better represent the data, keeping the same themes but with a different structure.

<i>Charting</i>	The aim is to chart each indexed theme (Richie & Spencer, 1994). Quotes were lifted from the primary data and placed in the appropriate theme chart in chronological order with participants' pseudonyms (Richie & Spencer, 1994). Charts were made for each focus group and interview then transferred to charts for the whole data set.
<i>Mapping and interpretation</i>	The aim of this phase is to define the concepts, map the range of the data, find associations within the data and provide explanation and typologies of the data in accordance with the aims of the research question (Richie and Spencer, 1994). Each theme table was read, re-read and reflected upon to further understand each theme and sub themes, and the connections both between and within the themes. More complex themes involved more extensive and more iterative mapping and interpretation

adhered to (Krueger & Casey, 2009; Richie & Spencer, 1994). While the aim of qualitative research is not to have an accurate account of the 'truth' but to understand why people recount stories in the way they do, given the overarching aim of the research study to evaluate current care with a view to influencing policy, it was important that participant's accounts were not too detached from reality (Barbour, 2008). For this purpose, contact was made with clinicians (LH, SL) to clarify the current and historical format of follow-up care within the recruiting hospitals.

A summary of the attributes valued by each group/interview was sent to its participants for verification (Krueger & Casey, 2009). No participants provided comments or corrections. Participants were given pseudonyms for the purpose of data reporting.

Results

3.3.9 Patient demographic data

Seventeen patients and seven informal caregivers participated in the study. The mean age of patients was 63 (SD 11) with an age range of 40-76 years. Eight patients had been diagnosed with endometrial cancer, four with ovarian cancer and five with cervical cancer. Sixteen patients had been treated with surgery, four with chemotherapy and five with radiotherapy including brachytherapy. Further patient demographics are detailed in Table 12, with a breakdown of key dyad demographics in Table 13.

Table 12. Patient demographic data

Demographic characteristics	Patients N=17 (%)
Gender	
Female	17 (100)
Male	
Age	
40-50 years	3 (18)
51-60 years	2 (12)
61-70 years	8 (47)
71-80 years	4 (24)
Ethnicity	
British	1 (6)
English	7 (41)
Welsh	8 (47)
Scottish	1 (6)
Employment status	
Employed for wages	7 (41)
Retired	8 (47)
Semi-retired	1 (6)
Unable to work	1 (6)
Type of gynaecological cancer diagnosed with	
Endometrial cancer	8 (47)
Ovarian cancer	4 (24)
Cervical cancer	5 (29)
Stage of disease	
Stage 1	8 (47)
Stage 2	1 (6)
Stage 3+	1 (6)
Unknown	6 (35)
Cannot remember	1 (6)
Treatment received*	
Surgery	16 (94)
Chemotherapy	4 (24)
Radiotherapy (including brachytherapy)	5 (29)
Duration in Follow-up	
0-6 months	3 (18)
7-12 months	2 (12)
13-18 months	2 (12)
2-3 years	3 (18)
3-4 years	2 (12)
4-5 years	4 (24)

10 years	1 (6)
Location of follow-up appointment*	
Hospital treated at	17 (100)
On the telephone	4 (24)
Health Care Professional that delivers the appointment*	
Consultant	15 (88)
Specialist Nurse	9 (53)
Other	2 (12)
Oncologist	1 (6)
Not specified	1 (6)
Current frequency of appointments*	
Every 3 months	7 (41)
Every 6 months	3 (18)
Every 12 months	5 (29)
When I request one	1 (6)
Discharged at last appointment	2 (12)
Number of follow-up appointments received	
1-5 appointments	6 (35)
6-10 appointments	10 (59)
>10 appointments	1 (6)
Content of follow-up appointment*	
Medical test	16 (94)
Asked how I am feeling	13 (76)
Onward referrals	2 (12)
Relationship to informal caregiver nominated	
Spouse	10 (59)
Sister	2 (12)
Mum	1 (6)
Not nominated anyone/not cared for	4 (24)
Duration provided informal caregiver	
Pre diagnosis/diagnosis to post treatment	4 (24)
Pre diagnosis/diagnosis to current	8 (47)
A week post operatively	1 (6)
Not reported	4 (23)

*Participants provided more than one response so N > 17 and % > 100

3.3.10 Patient Thematic Results

Focus groups and interviews were analysed in accordance with the aims of this research study. The core emergent theme from the patient data was one of a need for reassurance, perceived by all patients as the aim of follow-up care as described by Louise and Betty. Reassurance that: 1) there was no sign of recurrence (bar Edna who explicitly said she did not think there was a risk of her cancer recurring), 2) they were healing as planned, 3) reassurance by providing

emotional support, and 4) reassurance by providing an opportunity to ask any questions and have these answered. It was perceived that follow-up could meet these outcome expectancies providing patients with value (theory of planned behaviour: Ajzen, 1985; Ajzen, 1991), and

Table 13. Key dyad demographics

Name	Cancer	Stage	Treatment			Duration in FU	Carer	Carer duration (as reported by patient)	Caregiver Name
			Surgery	Chemo	Radiotherapy/Brachytherapy				
Elizabeth	Ovarian	3+	Y	Y		3-4 y	Y	Since diagnosis	Oliver
June	Ovarian	1	Y			4-5 y	Y	Since diagnosis	Kate
Shirley	Ovarian	Unknown	Y		Y	0-6 m	Y	From 6 months pre diagnosis	Sian
Meinir	Ovarian	2	Y	Y		3-4 y	Y	From 6 months pre diagnosis	-
Florence	Endometrial	Unknown	Y		Y	7-12m	N	n/a	-
Edna	Endometrial	Unknown	Y			13-18 m	N	n/a	-
Marjorie	Endometrial	1	Y			0-6m	N	n/a	-
Linda	Endometrial	Earliest form	Y			0-6m	Y	Since diagnosis	Andrew
Leslie	Endometrial	1	Y			4-5y	Y	For 3 months	-
Lisa	Endometrial	1B	Y		Y	13 -18m	Y	Diagnosis to post treatment	Eamonn
Myfanwy	Endometrial	Unknown	Y		Y	2-3y	Y	Approximately 3 years	-
Pat	Endometrial	Unknown	Y	Y	Y	4 -5y	N	n/a	-
Jenny	Cervical	Forgotten	Y			10 y	Y	From pre diagnosis	-
Kirsty	Cervical	1A1	Y			4-5y	Y	Post diagnosis for a couple of months	-
Louise	Cervical	1B1	Y			2-3y	Y	For a week after post operation	-
Tania	Cervical	1	Y			2-3y	Y	Unknown	Jim
Betty	Cervical	Unknown	N	Y	Y	7-12m	Y	Since diagnosis	Alfred

these perceived benefits (health belief model: Rosenstock, 1974; Becker, 1974; Becker & Rosenstock, 1984).

Betty (I1): "... giving you confidence to to know that there is a light at the end of the tunnel ... you've always got to a you know somebody to go to ... it's amazing how important the follow-up is it is really important ... you know that you are going ahead you are not at a standstill you are going you are improving you know it's going alright it's been worth it all ..."

Louise (G5): "Check on you wellbeing I suppose to you know mental and physical ... you know physically you know the cancer can come back can't it so they have to make sure that that everything's ok and then obviously you've got healing afterwards so you need to check on that and then mentally it's a lot to go through isn't it for you and your family so I suppose they need to make sure that you are doing ok after that"

Patients assumed that attending follow-up appointments helped to maintain life, through detection of recurrence, reducing the perceived severity of a recurrence (Health belief model: Rosenstock, 1974; Becker, 1974; Becker & Rosenstock, 1984) as illustrated by Lisa.

Lisa (I4): "I'm thankful to be monitored because I want to be here... "

Within the core theme of reassurance, two key themes emerged, each with sub themes: (1) *Meet my needs please* (access to an expert; procedures; holistic care) which provided reassurance (2) *Barriers to Care* (system failure; low self-confidence) which prevented access to reassurance. These iteratively identified themes and sub themes are depicted in Figure 10.

Figure 10: Patient themes and sub themes

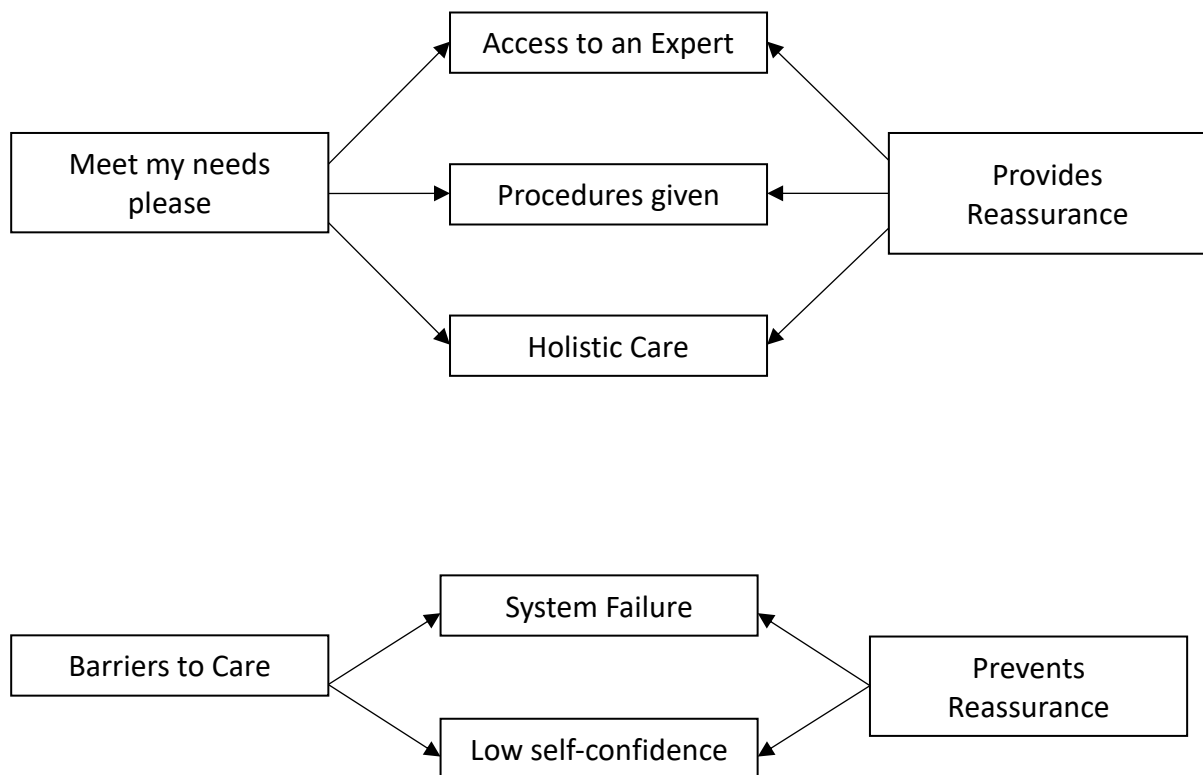
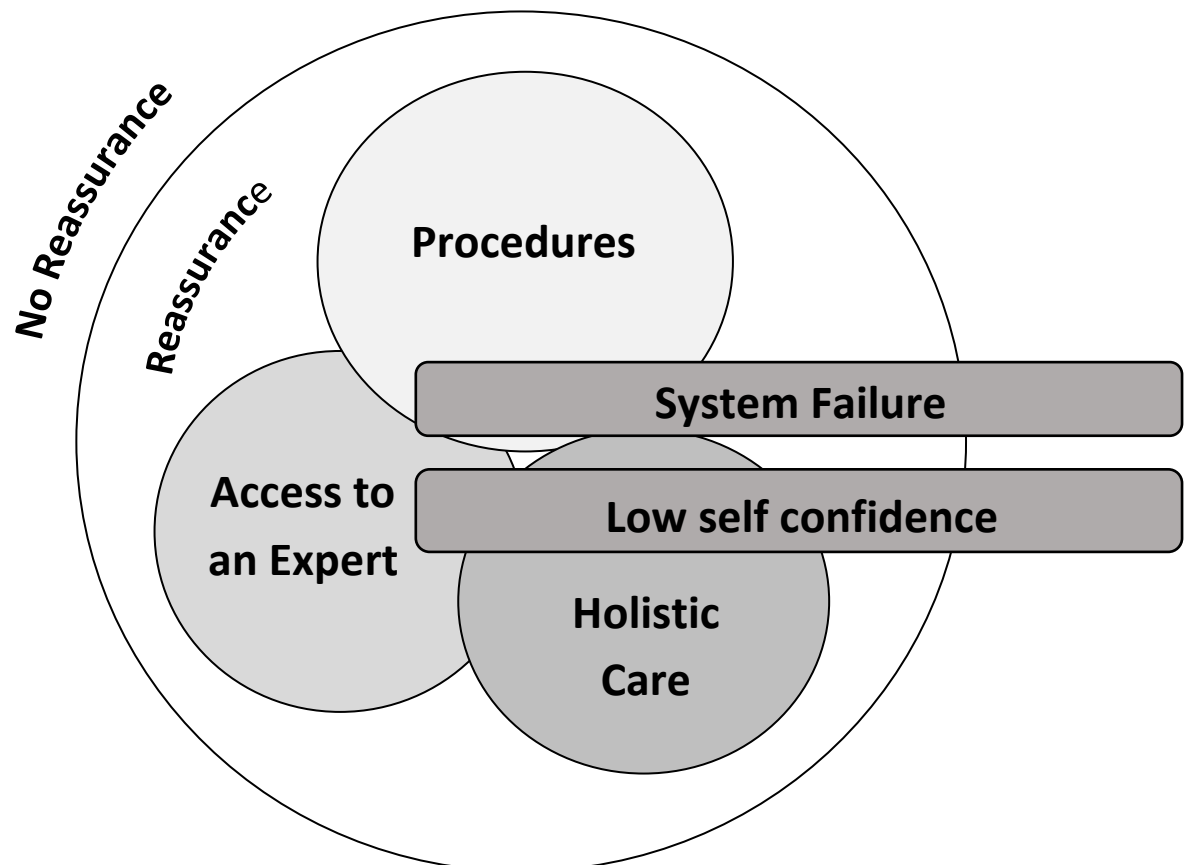


Figure 11: Thematic theme diagram (patients)



There was overlap between themes. The relationship between reassurance and the themes is illustrated in Figure 11.

3.3.10.1 Meet my needs please

Access to an Expert

Patients described how the act of care provision, and 'being in the system' provided them with reassurance. Participants explained how they valued the appointment itself; appointment letter; level of expertise of the Health Care Professional; Health Care Professional's attitude; continuity of the Health Care Professional seen; the mode of contact (face-to-face or telephone); being provided with the contact details for, and encouraged to call the Specialist Nurse in between appointments with any concerns, with re-access to the clinic between these scheduled appointments if needed; the frequency of appointments with these being more frequent at the beginning and having a reducing frequency over the follow-up period.

The act of having follow-up care appointments in itself provided reassurance, as patients knew that they would be having 'check ups' with a specialist, as illustrated by Kirsty. Tania summed how vulnerable patients felt post-diagnosis and treatment, and how because of this, follow-up care is perceived to be so crucial (particularly in the first couple of years post treatment).

Kirsty (I6): "Just to know that I had those appointments and you know whether they were 6 months or yearly is a peace of mind in itself"

Tania (I8): "...you feel as though you want people to keep checking up on you every 5 minutes to make sure you [laughs] you're really alright you know ... we have this urm the doctor knows best don't we [laughs] so you want to see the doctor"

Physically receiving the appointment letter provided reassurance to participants as it was a physical 'thing' demonstrating that they were going to be seen at a certain time. Each appointment was viewed as a goal post to get to as depicted by Tania, thus the provision of care in itself acting as a coping strategy (Leventhal et al., 1992).

Tania (I8): "...as soon as I'd seen I'd had my appointment I'd be thinking please let the next one [appointment letter] come ... I put it on the calendar you know ... I've been seen in January and now I need to be seen in March"

Seeing an expert in secondary care provided reassurance. Other factors being equal (attitude of Health Care Professional seen; continuity of Health Care Professional seen), patients with *experience* of seeing different Health Care Professionals for their appointments did not have a preference for the secondary Health Care Professional that delivered their appointments, as described by Edna and Marjorie.

Marjorie (G1): "I don't think [it matters] so

Edna (G1): no I don't mind as long as they know there subject I'm not bothered whether it is a doctor or a nurse or

Marjorie (G1): no

Edna (G1): what their sort of level"

Patients who did not have experience of seeing different secondary Health Care Professionals for their appointments either did not discuss this attribute or were strongly opposed to seeing someone other than the Consultant for their appointments, when the hypothetical possibility was discussed through presentation of the case study examples, as illustrated by Tania. The idea of this was alien to her and she did not have confidence in their ability.

Tania (I8): "... I mean personally again I would want to see ... my doc are you saying... Specialist Nurse instead of the ...Urm [pause] it just no it's just building up this trust I guess ... maybe I don't I'm not informed enough as to how how well trained the Specialist Nurses are ... I don't know I'd have to think about that one I think ... urm NO. no not for me not Specialist Nurse no"

Patients did not value the idea of having their follow-up care appointments delivered by a General Practitioner as they felt that General Practitioners did not have enough knowledge about gynaecological cancer (as a 'General' practitioner), and felt that there was a lack appointments and time in primary care, as summed by Group 2. However, some participants

did value the support of their General Practitioner for more non-specialist follow-up care needs (needs (June, Edna, Elizabeth, Lisa). Two participants explained that they made appointments with their General Practitioner's to discuss their follow-up care needs (Lisa regarding the effect on her bowels; Louise regarding an onward referral for counselling due to impeded quality of life from treatment).

Florence (G2): "The doctors ...They only do 2 weeks in advance so ...

Linda (G2): well they are not necessarily oncologists are they they are just GPs"

Participants placed a strong value on the Health Care Professional having a caring and empathic attitude. This went a long way to support and reassure patients, as illustrated by Betty. However, Lisa explained that in her experience a caring attitude was not always shown.

Betty (I1): "...It's this lovely, his attitude is lovely...the way he goes about reassuring you its nice...sort of casual but see [laughs] you know what I mean making you feel right. Yeah he's they've all been great actually yeah"

Lisa (I4): "You need to get into the gym, you need to do this!"... sometimes you just think I'm just... I'm getting my head round what's gone on do you know what I mean? ... I do think they need to watch how they speak to people in some ways, for the approach of a follow-up"

Participants valued having continuity in the Health Care Professional seen as this helped engender trust, as 1) participants felt they knew the Health Care Professional and the Health Care Professional knew them, and 2) participants knew what to expect (with the method of examination sometimes differing between Health Care Professional's). This was viewed as particularly important given the intimate nature of the examination and the effect this has on the patient, as summed by Kirsty. However, it was acknowledged by some participants that seeing the same Health Care Professional is not always possible. Two participants felt that whilst it was good to see the same person for each follow-up care appointment, if they differed they still felt confident in them as they assumed they had looked at their notes beforehand, so believed that they 'knew' them (as explained by Louise). It is worth noting that

participants recruited from one hospital, reported continuity in the Health Care Professional seen, whereas participants in the other recruiting hospital did not. This is due to the format of the clinic. This difference did not emerge from the data, as participants from different hospitals attended different focus groups due to geographical constraints. Patients who saw the same Health Care Professional for each of their follow-up care appointments (as was standard practice due to the format of the clinic in one of the two recruiting hospitals), did not raise the topic of continuity of Health Care Professional seen. However, Jenny explained that this was only something she valued in hindsight with originally her appointments being delivered by the same Health Care Professional, but five years post treatment being delivered by a different Health Care Professional on each appointment.

Kirsty (I6): "You're not going to sort of get your finger nails cut....it's quite nice to just have one person that you're having to, you know, examine you in that way"

Louise: "I tend to see the same Consultant each time but urm occasionally one of them will do a different clinic but that's fine cos you know when you go in that they've looked at your notes..."

Jenny: "...having the same specialist I felt was really important ... I appreciated afterwards when I was er given somebody different everytime ... I only sort of appreciated looking back ...you know that they can compare from ah the previous meetings"

Participants with experience of follow-up care appointments delivered by the Specialist Nurse over the telephone valued them strongly on the grounds of their accessibility. These participants valued being given the choice of whether they had telephone appointments or face-to-face appointments with the Consultant/Registrar, as illustrated by Marjorie and Edna (G1). However, June made it clear that she would not value telephone appointments (it should be noted that she was promised these appointments but did not receive them, see 'System Failure').

Marjorie (G1): "... my next follow-up and several are just going to be a telephone call I think is just fantastic it saves you going to the hospital and sitting in, and waiting...

Edna (G1): yeah, I thought that was really great idea... I was glad of that...

Marjorie (G1): ... don't like going to the hospitals ... it's so much simpler to be at home...

Edna (G1): and to be honest with me its, its sheer pressure of time...

Marjorie (G1): I mean some people, maybe don't really care for face to face and find it easier over the phone but then you get it the other way ...

Edna (G1): yeah. And to be fair they did, they asked me is that alright

Marjorie (G1): yes"

June (G2): "I wouldn't want to pay for a telephone call that's for sure"

For all patients just knowing that an expert was at the other end of the telephone for support and providing re-access to the clinic if needed was reassuring and highly valued, even if participants had not used this service. The simple knowledge of available telephone support helped patients to feel cared for and not feel like they were not on their own. Participants who did call the Specialist Nurse explained that she provided reassurance that how they were feeling physically and psychologically was normal, but that if the Specialist Nurse felt or they felt they needed a clinic appointment, a clinic appointment was made for them, as summed up by participants in Group 5.

Louise (G5): "I can phone her [the Specialist Nurse] up and leave a message and she always gets back to me ... I don't have to worry and think oh I've got 3 months to wait till the next appointment ... she'll get me in some times on a clinic within a week or so
Meinir (G5): yeah she does yes yeah"

Patients valued having regular appointments at the beginning of their follow-up care journey, given the level of reassurance they felt they needed at this stage. The gradual reducing frequency of clinic appointments was perceived as a positive sign that the Health Care Professional thought they needed to be seen less often, as illustrated by Tania.

Tania (I8): "I don't think until you've been through cancer you don't realise you just are so you feel so vulnerable you just want to know that you're going to be followed up and that they're watching you I mean that feeling obviously diminishes as time goes by but ... the first year 18 months it's just terribly important....[its] reassuring in itself to see ... the time between each appointment stretching out ... on my way to the the magic 5 years and then I don't have to see them at all"

This reduced frequency did not make patients feel less detached from secondary care experts and the reassurance this provided, as they knew they had the telephone number for the Specialist Nurse if needed demonstrated by Louise.

Louise (G5): "With me It it was every 3 months after my surgery's and now it's gone down to 6 months and I'm I'm happy with that cos I know if I've got any worries I can phone [Specialist Nurse's name removed]"

Leslie explained that even though she valued the appointments she dreaded them and they were a reminder of the disease.

Leslie (G4): "...follow-up appointments ... won't let me let it go because I am constantly going back and although they're good and I need to have them, they are a constant reminder ... I have 6 more months to see the surgeon and I'm ... off into the blue yonder... that is great for me, because I want to get on now. I have been getting on but... it is a reminder..."

The majority of participants had three monthly appointments at the onset of care. Interestingly, because of how well she felt Marjorie would have preferred to have yearly appointments at the onset of her follow-up.

Marjorie (G1): "Maybe wouldn't want them so frequently but yet I understand why they are. I wouldn't say oh, it's a waste of time ... because of the way I feel I think well why every 3 months... you know yearly would suit me but thats me

...

Edna (G1): "... have to leave that with the professionals ... I don't really think I can give an opinion on how often I think they ought to happen because you, you, you imagine that the people that are that are giving the care know how frequently you need to be seen"

Despite the value placed on all these elements of having 'access to an expert', four patients acknowledged that the question that they all want answering cannot be answered, as illustrated below by Betty and Pat.

Betty (I1): "And it's knowing now, like [pause] I have not got cancer, have I? Or have I because I am still sort of still on that recovery period, as it and nobody can definitely say whether it has definitely gone or whether it is going to occur anywhere else".

Pat (G4): "... the big question that we have is "Will it come back again?" A doctor can't give you that answer...it's like putting a car in for its MOT and it's fine. And then it, a wheel falls off. You don't know".

Procedures given

Patients placed a huge value on the procedures given, because of their assumed importance in detecting recurrence. Participants who vocalised their feelings about the internal examination, hated them, as illustrated by participants in Group 4 below.

Leslie (G4): "I've had more people look up my foof in the last five years than I've ever had in my entire life and I hate it.

Myfanwy (G4): I hate it....

Leslie (G4): ...I don't want them and we have to have them [laughs]

Myfanwy (G4): Yeah, it's a necessity. You've just got to go through with it...

Pat (G4): ... I don't care because if they find anything they're going to be...

Myfanwy (G4): Yes well that's it, they can tell you, you know, if there's anything there that shouldn't be. They can straight away get on to it then, can't they..."

Two participants (Tania; Jenny) had experienced tests as well as examinations, and placed more value on these. Other participants (Group 2; Lisa) questioned whether medical tests would provide more reassurance than examinations. Interestingly, Jenny explained that at 10 years follow-up she was informed by the Consultant that internal examinations are a false reassurance, which helped her to 'let go' of her need for regular appointments.

Jenny (I6): I was being given smear tests initially and a follow-up letter so it was all down on a piece of paper that you were clear for another so and so many years ... by the end of the time I was obsessing really ... the examination wasn't even a smear test by the end it was just an internal examination ... Mr [name of Consultant removed] said its false hope in a way you know so urm I let go in the end"

Holistic Care

Patients described heterogeneous effects of diagnosis and treatment, and expressed needs in relation to these. Reported physical effects of treatment included one or more of the following: bladder problems (Pat; Myfanwy); bowel problems (Myfanwy; Pat; Louise; Lisa; Shirley); impeded sex life (Pat); hair loss (Shirley, Elizabeth, Meinir); and financial problems (Louise). Nine participants (Edna; Linda; June; Florence; Betty; Kirsty; Jenny; Tania; Meinir), did not report any lasting *physical* effects of treatment. Two participants explicitly reported to return to 'normal' quickly (Marjorie; Kirsty). Others felt however that you are never 'normal' again (Leslie; Pat; Myfanwy). Participants felt that these needs from diagnosis and treatment would be best met under a holistic multidisciplinary model of care, with patients' needs identified through a simple questionnaire and onward referrals to services that could support them, as illustrated by Louise. Participants valued differing levels of information regarding these needs, and some participants had a preference for a peer support group to meet some of their need for reassurance, as illustrated by participants in group 4.

Pat (G4): "...[brachytherapy] has altered my sex life... a door has closed".

Louise (G5): "... cancer you know causes a wide variety of problems ... if they just gave out a simple questionnaire... [then] pass it onto the relevant bodies"

Leslie (G4): And I think this talking, I mean Pat and Myfanwy talking, I mean, I've not had radiotherapy, I'm a baby to you girls. You know, I only had surgery and I was done. But you know, even for me, to have this sort of peer support would have been brilliant. You know, I think, to talk to other people who've... <Pat and Myfanwy agree> because we've all had the same diagnosis in different stages, haven't we ... this would have been great to do with women who were diagnosed when I was.

Pat (G4): Yes, I agree there! Yes.

...

Leslie (G4): ... it's that reassurance from somebody else and I kind of missed that...

....

Myfanwy (G4): You know, I mean here with these ladies it's easier to talk.

Pat (G4): Yeah it is.

3.3.11 Barriers to Care

In the second theme patients described the barriers they experienced in accessing follow-up care. Two barriers to follow-up care were identified, 1) system failure, 2) low self-confidence.

3.3.11.1 System Failure

Four patients did not receive all follow-up appointments they should have been scheduled for, which hindered their access to reassurance (June, Tania, Lisa, Shirley). Shirley only had follow-up appointments with oncology, rather than with gynaecology as per local follow-up protocol. For Lisa, not receiving a scheduled appointment was simply rectified. However, Tania and June fell through the system on multiple occasions. Tania described the anxiety this caused her, and explained that she continued to 'fight' for her appointments, but June accepted it. Louise asked her General Practitioner for a referral to a counselling service for the post-treatment effects she was experiencing, but believed that a poor quality of care was provided. Therefore, whilst a system was in place to support her psychologically, the actual care provided was not perceived to meet her needs.

June (G2): "... Notes getting lost. Wrong clinic ... I didn't have appointments regularly ... I was told I'd get phone call appointments and never did ... might as well have had a polythene bag over my head ... he said how's the chemotherapy coming along. Well I haven't had any ... never got sorted out it was just like there was a file somewhere and they just used to throw odd letters into it and it'd be mine..."

3.3.11.2 Low Self-confidence

Self-confidence had a large effect on the care received. Some patients felt able to telephone the Specialist Nurse or a Consultant's secretary if appointment letters had not been received, or if they had any concerns or questions regarding psychological and physical symptoms, and continued to pursue this until the issue was resolved for them. However, other patients did not feel confident re-accessing follow-up care between scheduled appointments and/or discussing concerns in their follow-up appointment, and/or keep telephoning the Specialist Nurse or Consultants secretary until the issue was resolved for them, as illustrated below by Pat and Myfanwy.

Pat (G4): "I was concerned ... I never said nothing I just, you know, went on for months really and then I thought no, I'm going to go and see him..."

Myfanwy (G4): And thank you very much because I thought, you know ... Thank you because I, nobody, I don't know. Talking to you... you're too embarrassed sometimes to... you know... you're too embarrassed sometimes to... you know, especially when it's a, it's a man, you know... I've never had chance to ask anybody about it ... I forgot to mention it to the lady doctor... I didn't like to mention it to the man ... if I see the Registrar it's a lady and I will talk with her...."

3.3.12 Individual differences

There is an emerging relationship between demographics and preferences. As can be seen by Table 12 and Table 13, there were both similarities and differences in demographics, and as can be seen through the analysis, similarities and differences in preferences for gynaecology cancer follow-up care. Seven patients had a preference for an increased level of care to meet their needs (Shirley, Leslie, Lisa, Myfanwy, Pat, Jenny, Louise), of these, four reported to having been treated with radiotherapy or brachytherapy (Shirley, Lisa, Myfanwy, Pat), and five

of these reported to be currently experiencing physical effects of treatment (Shirley, Lisa, Myfanwy, Pat, Louise).

Differences in preferences also emerged to be due to at least in part, differences in appraisal of the disease, perceived susceptibility for risk of recurrence, and individual coping, in line with the self-regulation model of illness (Leventhal et al., 1992), and health belief model (Rosenstock, 1974; Becker, 1974; Becker & Rosenstock, 1984). Marjorie, Linda and Leslie were diagnosed with stage 1 endometrial cancer and treated by surgery alone, and Edna whilst not knowing the stage of endometrial cancer she was diagnosed with, was treated by surgery alone. There was huge homogeneity in these participants' preferences for follow-up care despite presenting with similar diagnoses and treatment pathways. Marjorie had a preference for reduced frequency of care as she felt well, "it's almost as if I've had a cold", whereas Leslie and Pat explained how you never feel normal again, and had a preference for peer support groups.

Leslie (G4): "...you're not normal.

...

Pat (G4): none of us are normal after"

Conversely, Edna and Linda both valued their follow-up appointments because of the reassurance provided, but did not state a preference for either 'more' or 'less' follow-up provision. These differences in preferences could not be attributed to duration in follow-up care, with Marjorie and Linda both 0-6 months post treatment, Edna 13-18 months post treatment, and Leslie 4-5 years post treatment.

Florence and Myfanwy were diagnosed with endometrial cancer with an unknown stage but treated with surgery and radiotherapy/brachytherapy, Lisa was diagnosed with stage 1 endometrial cancer and treated by surgery and radiotherapy/brachytherapy, and Pat reported to be diagnosed with endometrial cancer with an unknown stage, but was reported to be treated by surgery, chemotherapy and radiotherapy. These participants' preferences also had large variance, with Myfanwy and Pat (whom both said that they were treated with radiotherapy/brachytherapy) reporting to need more care to meet physical and psychological needs of treatment (and for Myfanwy, and/or possible recurrence). For Myfanwy and Pat, the biopsychosocial impact of diagnosis and treatment was very salient, despite Pat being 4-5

years post treatment. Lisa also had a preference for support groups and more frequent appointments, but these preferences were not as salient and needs not as 'raw' for Lisa as they were for Myfanwy and Pat. Conversely, Florence, whom was also treated with surgery and radiotherapy/brachytherapy, valued the level of care she received (alternate clinic and Specialist Nurse led telephone appointments) but did not have a preference for either 'more' or 'less' care.

Kirsty, Louise, and Tania were all diagnosed with stage 1 cervical cancer and treated by surgery alone. Jenny and Betty were diagnosed with cervical cancer, with an unknown stage. Jenny was treated by surgery alone, whereas Betty was treated by chemotherapy and radiotherapy/brachytherapy. Whilst Kirsty, Louise, Tania and Jenny had similar illness and treatment pathways, their preferences for care were very heterogeneous. Jenny really struggled to let go of follow-up provision and pushed to be followed up for longer than the amended protocol of care (to 5 years post treatment), being followed up for 10 years until she was able to let go after (as opposed to being followed up till post-menopause). Similarly, Louise who was 2-3 years post treatment, reported that the impact of diagnosis and treatment received was very salient, with her currently experiencing pain and reduced quality of life. As such, Louise had a preference for frequent follow-up appointments, as did Kirsty who was 4-5 years post treatment. Conversely, Tania (who was also 2-3 years post treatment) felt that she was able to let go of her need for follow-up care at her next follow-up appointment (which would be 3 years post treatment).

Elizabeth, Shirley, June and Meinir were all diagnosed with ovarian cancer but with different stages of disease. June was diagnosed with stage 1 disease and treated by surgery alone. Meinir was diagnosed with stage 2 disease and Elizabeth was diagnosed with stage 3+ disease, and both treated by surgery and chemotherapy. Shirley did not know her stage of disease but reported on the demographic questionnaire to having received surgery and radiotherapy/brachytherapy, but in her focus group reported to having received chemotherapy. Shirley reported a need for additional follow-up care to address the biopsychosocial effects of her treatment, but Elizabeth felt her effects of treatment were addressed in her follow-up appointments, however she was 3-4 years post-treatment whilst Shirley was 0-6 months post treatment, and did not have a colostomy bag fitted like Shirley did. Shirley also only received follow-up care with an oncologist and not a gynaecologist, which may in part explain these different stated preferences. June also had a preference for 'more'

care but this was because she experienced system failures throughout her follow-up period so did not feel cared for. Conversely, Meinir was happy with the care experienced.

It is worth noting that the majority of patients with the most salient preferences for 'more' care to meet their physical and psychological needs were all in the same focus group (Pat, Leslie, Myfanwy: Group 4).

3.3.13 Summary of patient findings

In summary, because of the reassurance that it provided, patients valued having access to an expert and the individual components of this (the appointment itself; receiving an appointment letter; the Health Care Professional's level of expertise, attitude, and continuity of care; the mode of contact (face-to-face or telephone); contact details for the Specialist Nurse with re-access to the clinic if needed between scheduled appointments; frequency of appointments), the procedures given, and holistic care. However, barriers to care (system failure and low self-confidence) prevented access to this care and so reassurance.

3.3.14 Informal caregiver demographic results

Five informal caregivers were male spouses and two were female (one the patients' sister and one the patients' daughter: see Table 14). The mean age of informal caregivers was 62 years (SD 13) with an age range of 39-74 years. One informal caregiver had been providing care since pre-diagnosis, three had been providing care since diagnosis, one only provided care during the treatment period, and one reported to have only provided care in the follow-up period. One informal caregiver could not recall when they began providing care beyond their normal role. Informal caregiver characteristics are detailed in Table 14.

3.3.15 Informal caregiver thematic results

Informal caregivers felt that the aim of follow-up care was to provide them and the person that they cared for with the following outcome expectancies: reassurance that there was no sign of recurrence, monitor the care recipients physical and psychological wellbeing and provide further treatment if needed (perceived benefits). As Kate explained, given the 'image' of cancer, this fear of recurrence (perceived susceptibility) and a resultant need for reassurance was particularly salient (coping response: Leventhal et al., 1992; Rosenstock, 1974; Becker, 1974; Becker & Rosenstock, 1984).

Kate (I3): “check the physical condition of the the person, urm offer support, make sure the persons well ... check their psychological wellbeing is okay ... Offer further treatment.... cancer is perceived to be this thing you know it attacks you know it’s almost like a person ... like that advert on the tele which is prepare to die cancer...it’s almost like this external like thing that that’s coming to get you and you’ve got to ah you know somehow stave it off ...I think because of that there’s this real kind of fear in people you know that cancers coming back and its coming to get you”

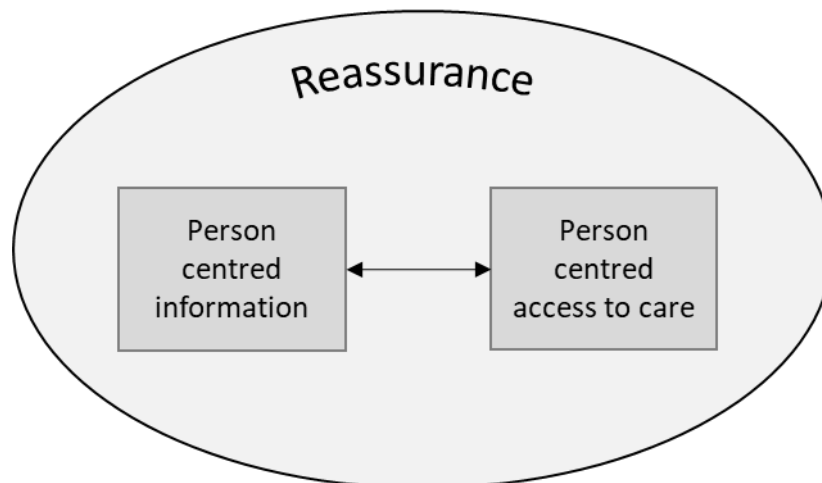
Table 14. Informal caregiver demographic data

Demographic characteristics	N=7
Gender	
Female	2 (29)
Male	5 (71)
Age	
40-50 years	1 (14)
51-60 years	2 (29)
61-70 years	2 (29)
71-80 years	2 (29)
Ethnicity	
English	4 (57)
Welsh	3 (43)
Employment status*	
Employed for wages	3 (38)
Retired	2 (25)
Homemaker	2 (25)
Volunteer	1 (13)
Relationship to patient	
Spouse	5 (71)
Sister	1 (14)
Daughter	1 (14)
Duration providing care to patient	
From pre diagnosis	1 (14)
Since diagnosis	3 (43)
During treatment	1 (14)
During follow-up	1 (14)
Unknown	1 (14)

*Participants provided more than one response so N > 26 and % > 100

Informal caregivers felt that their need for reassurance was met by the provision of person centred information, which in turn was met by the provision of person centred access to care. These iteratively identified themes are depicted in Figure 12.

Figure 12: Thematic theme diagram (informal caregivers)



3.3.15.1 *Person centred information*

It was perceived that information is central to follow-up care, with patients attending follow-up care because of the information (and so reassurance) that this provides, as described by Andrew and Oliver. Informal caregivers believed that everyone is different, and so had a strong preference for information to be centred based on the needs of the patient in terms of its structure, content (verbal and written), and frequency, and so met within the multidisciplinary team, as summed by Jim.

Andrew (G3): "...the information that you get and what we like, it's good. But we're also concerned about the information we don't get [when] ...talking about regular contact, we're also talking about information ... it's very much hand in hand.

Oliver (G3): They cross over. Yeah ... regular contact, information ... continuity er I think they're all sort of linked really in a way ... I don't think you can separate them really.

Jim (I9): "every patient is different and every patient's needs are different no matter what the, the severity of their condition ... it's all down to how ... they're personally

coping ... the system has got to have structure. But it's got to have flexibility within its structure because different people have different needs"

Face-to-face information was valued above the written information given to patients. Interestingly, informal caregivers had a preference to receive detailed face-to-face information, but more basic written information, as summed by Jim.

Jim (I9): It's all down to patient's right to know everything and by knowing everything I think all, all, the only result then is they create too much concern rather than putting people's minds at rest...

Tania (I9): ... All you really wanted was to talk to the people who are looking after you.

3.3.15.2 Person centred access to care

Access to follow-up care provided informal caregivers with the information they desired, and in turn reassurance. Informal caregivers valued the level of expertise of the Health Care Professional, continuity of Health Care Professional seen from pre-treatment to follow-up, the personality and attitude of the Health Care Professional, the medical examinations and procedures the patient received, the patient receiving frequent appointments and being given the time needed in the appointment.

Receiving care from an expert helped the informal caregiver engender trust in the Health Care Professional, as explained by Alfred. The topic of which expert to provide this care was explicitly discussed in four interviews with mixed views (Eamonn, Jim, Alfred, Kate). A strong value was placed on Consultant led care by Jim and Alfred, but Eamonn and Kate also felt confident in the Specialist Nurse's and General Practitioner's ability to provide care. Preferences regarding the suitability of the General Practitioner to provide follow-up care appointments were influenced by prior experience, in accordance with the theory of planned behaviour and health belief model (Ajzen, 1991; Ajzen, 1985; Becker, 1974; Becker & Rosenstock, 1984; Rosenstock, 1974). Alfred felt that the General Practitioner was not suitable to deliver follow-up care, due to his wife's delayed diagnosis, whilst Kate trusted her and her Mums General Practitioner to deliver follow-up care due to a previous gynaecological diagnosis given to her by her General Practitioner, combined with feeling like the General Practitioner 'knows you', but Health Care Professional's in secondary care do not.

Jim (I9): ... I would want more than simply a Specialist nurse ... she needs the full weight of the medical services behind her ...

Alfred (I2): "...I feel very confident in the specialists ...I think she got the best treatment that was available anywhere in the country ... it was months that she was being told (by the General Practitioner] there was nothing wrong with her ... [the General Practitioner] not taking any action ...their knowledge is too urm widespread ... It's not specialised enough, totally not specialised enough ... they wouldn't know what to do..."

Kate (I3): "I've gone for appointments like that I've felt the General Practitioner knows ... the doctor said straight away no that's a blah blah blah ... it's a similar thing isn't it ... you don't have the urm the clinical specialism knowledge ... but you'd get the ... patient contact knowledge trust ... They'd of known exactly who she was ... in villages like this you do anyway"

All informal caregivers valued having access to unscheduled contact with the Specialist Nurse, to provide reassurance in between scheduled appointments, and prompt re-access to the clinic if required, even if they did not need to use this element of the service, as illustrated by Andrew. One informal caregiver and his wife was telephoned by the Specialist Nurse rather than having to telephone her. Jim reported that the support provided was invaluable to his wife's recovery, but because his wife was supported he did not feel that he needed support.

Andrew (G3): "I think it's important that even if you don't use it, it's nice to know it's there because um my wife had a dedicated nurse 24 hours, 7. we didn't need it but it was there. It's nice to have that kind of support"

Jim (I9): ...she was constantly on the phone sort of checking how you were and then

Tania (I9): ...she was ringing you to make sure you were alright. She was hugely important ... it means an awful lot.

...

Jim (I9): Yeah she was tremendous wasn't she.... (her support) neither helped nor hindered me because I I I was fine ... we knew we had it under control didn't we ...

Tania (I9): Yeah, no, I had it under control because of Sharon [laughs]

Jim (I9): You had it under control, yeah.

Tania (I9): You know, I wouldn't have had it under control without her.

Continuity of Health Care Professional seen, from pre-treatment to follow-up was valued by informal caregivers. Informal caregivers felt that through prior experience 1) they felt they knew the Health Care Professional, 2) they felt that the Health Care Professional knew the patient and their clinical history. This facilitated a trust in their expertise, as summed by Kate.

Kate (I3): "you feel ... if your Consultant is not there well how will this other person cope with me you know who are they I don't know who they are will they know me they don't know the decision I've made ...seeing the same person is is fairly important if not of a team of people"

Informal caregivers reported that some Health Care Professionals are harder than others to communicate with, affecting the level of information and so reassurance gained from follow-up care, as illustrated by Oliver. Related to this was the ease with which Health Care Professionals were understood, both in terms of medical jargon used and language barriers. Kate explained how this is particularly important in North Wales.

Oliver (G3): You can get the thing where you get one person who is deadpan and you can't read their thoughts and you haven't got a clue, or another one where their personality brings out, you feel you can, like you can ask questions to"

Kate (I3): "there's a real lack of welsh language service ... you go to an appointment and somebody's speaks says words that don't actually have any meaning to you you piece them together to try and work out you know ... and you ... see people as well that don't speak particularly good English..."

All informal caregivers valued being given the time they needed in the consultation, and not being rushed. All informal caregivers understood that a wait could not be avoided at times, and whilst feeling like it added to the worry of the appointment, understood that those with

appointments before them needed the Health Care Professional's time. However, informal caregivers had mixed valuations of the maximum waiting time as illustrated by Oliver and Eamonn.

Oliver (G3): "...if they're going to make her better, I don't care a monkey's ... treatment is what matters ... once you get to 3 hours you've just about had it then ...

Eamonn: "I would like to see it down to about 15 minutes within reason"

Informal caregivers perceived the gynaecological examination as essential for providing information regarding any signs of recurrence. Oliver explained how his wife used to receive medical tests as part of routine follow-up care, and because of the information these tests provided he had a preference for her to receive them. Two informal caregivers reported that the patient they cared for had received further tests due to concerns regarding recurrence, as illustrated by Andrew. Alfred explained that a further test had been offered to his wife but she had decided not to have it, as felt the symptoms experienced were a result of treatment not a sign of recurrence. However, he portrayed a niggling doubt in his mind.

Oliver "... we're also concerned about the information we don't get ... certain tests ... these scan things you have, you know. And they say "Oh we don't do that anymore" and I'm thinking... if you did them, we'd know for sure what the situation was"

Andrew (G3): "they'd actually said that there was some thickening ... she had to go for a biopsy and we did go back and the result was good"

Alfred (I2): "it is talked about at each urm at each meeting and umm he has said whatever she wants him to do he will do. He will do this follow-up by doing a sigmoidoscopy just to be sure that uh it's not the cancer that's causing this problem. But we're pretty certain that it's um the radiotherapy and scar tissue ... That's the reason I think, is it's either or, it's either this advanced cancer returning or it's urm problems with scar tissue from the radiation that she was given ... these problems with

urm radiation possibly don't show they don't immediately. They take several months to to manifest themselves. Yeah ..."

Frequent appointments were reassuring for the informal caregiver, with more frequent appointments post treatment being valued, and the time expanding between appointments perceived as the patient getting better, providing additional reassurance as illustrated by Group 3.

Andrew (G3): "...you need to regular care and regular assurances that things are OK....things are getting better..."

Sian (G3): It keeps them happy then...

...

Oliver (G3): ...there's nothing better than when you've been on the 3 month times and they say well "We shan't see you now for another 6 months" and you think "Ahhh getting better!" ...

Informal caregivers all felt that the patient they cared for had unmet needs. Kate, Jim, Alfred, and Sian, in part attributed these unmet needs to system failures. System failures prevented access to care, and the information that care provided, and in turn reassurance. For two informal caregivers (Kate, Jim) these system failures were due to poor organisation of appointments and case notes. For Alfred and Sian, these system failures were due to the patients' delayed diagnosis. System failures whilst preventing access to care and so reassurance, further added to the need for reassurance, as illustrated by Kate.

Kate (I3): "...you didn't get that feeling of urm ... we know who you are we know exactly what your urm clinical path is we know where you're at now ... it was so confusing and messy and jumbly ... she has a feeling like she's been missed and you know neglected ... have they overlooked something have I been missed... I think she had loads of needs that weren't met but probably because the care was bungled"

Unmet needs were also attributed to societal change (Oliver, Andrew, Sian), resulting in a perception of reduced availability of informal support networks and less engagement in more

formal support structures like the church. However, informal caregivers also felt that no one understands the experience bar others in the same situation. For this reason, informal caregivers had a preference for the setup of peer support groups for patients and their informal caregivers.

Oliver (G3): "if you go back say 50 years or 40 years ... a lot of support like this would come from the local church organisation ... And there's lots of charities that will do the same thing..."

Andrew (G3): I think it's down to social change as well. You know, families have moved away...

...

Oliver (G3): ... it's interesting talking to Andrew...

Sian: Well this is what I was saying to my sister ... have a class. So that people, you know, can have a chat. People that have had the same thing I mean ... You could talk better with them because they're more understanding.

...

Oliver (G3): ... we're all in the same boat, we've got the same judgements and we're interested ... because it's all happened to us ... But when you go home ... talking to Bill down the road and you start talking about it, he don't want to know ... consequently you're then frightened to talking to other people about it ... that's just the way it goes".

3.3.16 Individual differences

As can be seen by Table 13, informal caregivers supported patients diagnosed with different types and stages of disease, whom received different treatments, were different durations post treatment, and provided support over different periods. Andrews wife was diagnosed with stage 1 endometrial cancer and finished treatment 0-6 months ago. Similarly, Eamonn's wife was also diagnosed and treated for stage 1 endometrial cancer and had been in follow-up for 13-18 months, and Jim's wife was treated for stage 1 cervical cancer and had been in follow-up for 2.5 years. Alfred's wife was diagnosed with cervical cancer with an unknown stage, and had been in follow-up for 7-12 months. Oliver's wife was diagnosed with stage 3 ovarian cancer, and had been in follow-up for 3-4 years. Sian's sister was also treated for ovarian cancer but had been in follow-up for 0-6 months. Kate's mum was diagnosed with

stage 1 ovarian cancer and had been in follow-up for 5 years but now discharged. Kate and Jim supported the patient they provided care for to navigate the health care system with the system failures they experienced. Sian's sister, Eamonn's wife and Alfred's wife were treated with radiotherapy/brachytherapy, and Oliver's wife, Alfred's wife and Shirley's sister treated with chemotherapy (Shirley's sister reported on her demographic questionnaire to receive surgery and radiotherapy/brachytherapy but in her focus group reported to receive chemotherapy). However, preferences for care did not differ by these demographics, but were evident by individual caregiving role, caregiving tasks, the appraisal of the disease, perceived susceptibility and severity of the disease and the outcome expectancies and perceived value and perceived benefits of attending follow-up appointments (Ajzen, 1985; Ajzen, 1991; Becker, 1974; Becker & Rosenstock, 1984; Rosenstock, 1974; Leventhal et al., 1992).

All participants provided emotional and physical support to the person they provided care for, and within this, provided differing caregiving tasks as illustrated by Sian, Kate and Jim.

Sian (G3): ... I keep on telling her oh maybe she's having problems with this bag. Maybe it's this, or you, it could be that ... I've said to her, doesn't matter what time, any time of the morning, early hours of the morning, I'm always there. I can get in the car. Five minutes up the road ..."

Kate (I3): "...I made it part of my job ... to find out exactly what is what and help you you know decipher it all and then to tell the rest of the family what's what"

Jim (I9): "I had to do very little other than be here for her when she needed..."

Some informal caregivers (Oliver, Andrew, Sian and Alfred) valued diagnostic tests. They felt that this would provide them with more 'concrete' information that the patient they provided care for 'would be ok'. As such, provision of diagnostic tests were a coping response to the threat of recurrence (self-regulation model of illness: Leventhal et al., 1992) given the perceived susceptibility and perceived severity of a recurrence, and perceived benefit of these medical tests (health belief model: ; Becker, 1974; Becker & Rosenstock, 1984; Rosenstock, 1974). For Oliver, Sian and Andrew (group 3), the emotional effect of caregiving was consuming, and led them to have a preference for the provision of care to help meet their

emotional needs (coping response: Leventhal et al., 1992). As illustrated below, they reported to put on a brave face to protect the patient despite being concerned that next year the patient would not be with them. These informal caregivers presented with higher levels of informal caregiver burden, and use of emotional labour (external management of emotions, putting on a 'brave face': Hochschild, 1983) and protective buffering, which in the literature has been associated with poorer caregiving outcomes (Langer, Brown & Syrjala, 2009; Rae 1998). Conversely, whilst effected by their role as informal caregiver, Alfred, Jim, Eamonn and Kate presented as coping with this role. These informal caregivers explained that they did not need support from more formal systems and networks. Two of these informal caregivers (Alfred and Kate) had family nearby and reported to be in close contact with them, who in turn provided them with support, as illustrated by Alfred. Jim felt that his wife had, had reassurance that she was ok, and by this nature so did he.

Oliver: "...Who cares for the carer ...

Sian (G3): We go through it as well...

Oliver: Because you do go through some very, it's a traumatic time. And of course you just keep this smile on your face.

Sian: ...I've got to think positive. Every time. And I treated her as if there was nothing wrong and she preferred it like that ... but you've got to be strong.

Oliver (G3): Yes... There's a worry in side like mad but outside you've got to be... strong.

...

Sian (G3): ... At night when I'm at home I could cry ... you sit and you worry ... It makes you feel ill.

Andrew (G3): If there's something happens you start to think ... "Well is she alright, was it something else?"

Oliver (G3): Every time you're at the hospital you think "Will we have bad news?" and you know, this time next year I'll be a widower?

Alfred (I2): "... if I was to need any extra help with the care I do have it on the doorstep. My youngest, my middle daughter ... she's been an absolute brick all the way through this"

3.3.17 Summary of informal caregiver findings

Informal caregivers valued having personalised access to care to meet their need for person centred information. Which if met, met the informal caregivers' need for reassurance. As such, attendance and engagement in follow-up care was part of an informal caregivers' coping response to the threat of a recurrence (self-regulation model of illness: Leventhal et al., 1992).

3.3.18 Synthesis between patients and informal caregiver results

There were similarities and differences in the themes and theme structure identified from patient and informal caregiver data. Analysis of patient data identified two themes each with sub themes: 'Meet my needs please' (with sub themes 'access to an expert', 'procedures given', 'holistic care') which provided reassurance; 'Barriers to care' (with sub themes 'system failure', 'low self-confidence') which prevented reassurance. Whilst analysis of informal caregiver data identified similar themes with the same overarching aim of providing reassurance, a different prevalence and salience of attributes of care were valued, reflected in the differing thematic framework. For informal caregivers, the relationship between reassurance and 'person centred access to care' was mediated by 'person centred information'. Patients and their informal caregivers generally perceived themselves/the person they provided care for, to be susceptible to a disease reoccurrence which if occurred, could result in death (perceived susceptibility and perceived severity: Becker, 1974; Becker & Rosenstock, 1984; Rosenstock, 1974). Patients and their informal caregivers believed follow-up could detect asymptomatic recurrence and reduce the impact of a recurrence (perceived benefits, outcome expectancies: Ajzen, 1985; Ajzen, 1991; Becker, 1974; Becker & Rosenstock, 1984; Rosenstock, 1974). Because of this, patients and their informal caregivers placed a strong value on follow-up care (Ajzen, 1985; Ajzen, 1991). Preferences for specific attributes of care were governed by an individuals' characteristics, perception of the benefit of that attribute in detecting recurrence and providing reassurance (again supporting the self-regulation model of illness: Leventhal et al., 1992; health belief model: Becker, 1974; Becker & Rosenstock, 1984; Rosenstock, 1974; and theory of planned behaviour: Ajzen, 1985; Ajzen, 1991).

3.3.19 Patient and Informal caregiver dyad results

As illustrated in Table 13, five informal caregivers were the patients' spouse (Alfred/Betty, Andrew/Linda, Oliver/Elizabeth, Eamonn/Lisa, Jim/Tania), one informal caregiver was the

patients' sister (Sian/Shirley), and one informal caregiver was the patients' daughter (Kate/Susan).

Two emerging themes were identified when exploring the patient and informal caregiver dyad data together, 1) the effect of the presence of an informal caregiver on service delivery and patient wellbeing, 2) discordant/concordant beliefs regarding perceived risk of cancer recurrence between the patient and the informal caregiver.

3.3.19.1 Informal caregiver presence

Three patients and their informal caregivers (Elizabeth/Oliver, Shirley/Sian, June/Kate) felt that the quality of care received was improved by the presence of an informal caregiver, to support and advocate for the patient. It was explained that patients experience fear and worry in their appointment, so might be in a confused state and so may not ask questions, and/or may misinterpret or not understand everything that is said. These dyads felt that higher levels of care are delivered by the presence of an informal caregiver. However, this was not endorsed by Tania/Jim, and Lisa/Eamonn. Tania did not feel that the quality of her care was improved by Jim, and Jim did not feel like he was an advocate for Tania. It was explained that due to Tania's personality and experience of working in the hospital, and her resulting knowledge of the hospital structure and prior relationship with the Specialist Nurse, she was equipped with the tools to help manage her care and cope with her experience. Lisa did not discuss the caregiving role that Eamonn provided, and Eamonn did not discuss the caregiving role that he provided during the follow-up care period. Lisa presented as being confident and self-reliant.

Elizabeth (G2): my husband is um, very forthright I suppose he will ask the questions as well I think er if he is not satisfied or if er if something is a bit vague he will take it to the point and find out you know and that he wont let things lie

Shirley (G2): I was on my own...

...

June (G2): So maybe, because your husbands there supporting you asking the questions you get a better service than you who go on your own

Shirley (G2): Yeah

June (G2): and don't ask the questions

...

Elizabeth (G2): ... when you are in that state you know you your mind isn't working

Shirley (G2): No its isn't

Elizabeth (G2): to ask questions you know ... the information should be given you know to everyone you know whether you've got the support that I had or whether like ...

Shirley she was on her own ... but she should have had the same amount of information

June (G2): yes exactly she should, Goodness

Elizabeth (G2): Particularly as it sounds as though we were at similar stages as well

June (G2): yes

Kate (I3): "... she's a pretty rubbish patient you know in terms of um saying how she feels what's going on um she's not forthright like that she needs somebody to ah speak up for her and get her to speak up for herself but um also that is true of patients isn't it... I'd hate id hate for people to have to go to those alone that would be awful having been to them ..."

Jim (I9): I had to do very little other than be here for her when she needed. And help her through it when she needed, but she's a very strong personality and had great faith in the system so really there was not a great deal of input from me ... she got lost in the system completely yeah but Tania is fairly tenacious and she sorted it...

3.3.19.2 Perceived risk of cancer recurrence

When exploring the patient and informal caregiver dyad data together, the interplay between patients and their informal caregivers' fear of cancer recurrence emerged (both the perceived susceptibility of a recurrence and the perceived severity of a recurrence: Rosenstock, 1974; Becker, 1974; Becker & Rosenstock, 1984). It was not in the aim of this study to explore perceived risk of recurrence and at a dyadic level. However, it could be seen that the salience of the level of fear of cancer recurrence was presented in some dyads to be at a discordant level and others at a concordant level, with differing effects on quality of life. For example, Oliver, Andrew, and Alfred presented as being very worried about the risk of cancer recurrence in the patient they provided care for, more worried than the patient themselves. For Oliver and Andrew this was consuming. Whereas this risk was presented as more salient in Tania and Lisa compared to their husbands (Jim and Eamonn), but did not present as having

a large impact on their quality of life. The relationship between patient and their informal caregivers perceived risk of recurrence and impact on quality of life should be explored in future research.

3.4 Discussion

The primary aim of the study was to identify and explore patients and their informal caregivers' preferences for gynaecology cancer follow-up, based on their experience of care. The secondary aim of the study was to use the attributes and attribute levels identified to aid the design of a subsequent discrete choice experiment (see Chapter 4). The specific objectives were to:

1. Develop an understanding of the attributes of care valued by patients and their informal caregivers, based on their experience of care.
2. Develop an understanding of why these attributes are valued as they are.
3. Identify attribute levels for each of the identified attributes.
4. Identify and understand hypothetical willingness to pay thresholds for gynaecology cancer follow-up care.
5. Identify and understand how long patients and their informal caregivers would be willing to wait between follow-up care appointments.
6. Develop an understanding of how patients and informal caregivers feel that follow-up care could be improved.

This study identified that patients and informal caregivers' preferences for care were governed by their need for reassurance. In line with the health belief model (Rosenstock, 1974; Becker, 1974; Becker & Rosenstock, 1984) and theory of planned behaviour (Ajzen, 1985; Ajzen, 1991), patients and their informal caregivers perceived the patient to be susceptible to a cancer recurrence (of differing perceived severities). It was perceived by patients and their informal caregivers that follow-up appointments would detect a recurrence (perceived benefit and outcome expectancies) and reduce the severity of a recurrence through earlier onset of treatment, thus providing a large perceived benefit of follow-up provision. As such, attendance and engagement in follow-up care acted as a coping response to the threat of a recurrence (self-regulation model of illness: Leventhal et al., 1992).

For patients, the need for reassurance was met through having access to an expert, the procedures given and holistic care. However, system failures and low self-confidence prevented access to this reassurance. For informal caregivers, the need for reassurance was met through the provision of person centred information, which in turn was met through person centred access to care (objective 1-2).

The themes identified from the analysis of patient data were 'Access to an expert', 'Procedures', 'Holistic care', and 'Barriers to care', the latter with the sub themes 'System failure' and 'Low self-confidence'. Within the themes described above, the following attributes of care were identified, as described in Table 15 (objective 1-3).

Table 15. Attributes identified from analysis of patient data

Themes	Attributes
Access to an expert	<ul style="list-style-type: none"> • Appointment itself • Appointment letter • Level of expertise of the Health Care Professional • Health Care Professional's attitude • Reciprocal communication • Continuity of Health Care Professional seen • Mode of contact (face-to-face or telephone) • Contact details for, and encouraged to call the Specialist Nurse in between appointments with any concerns • Re-access to the clinic if needed • Frequency of appointments
Procedures	<ul style="list-style-type: none"> • Examinations • Medical tests
Holistic care	<ul style="list-style-type: none"> • Holistic multidisciplinary care • Information • Peer support
System failure	<ul style="list-style-type: none"> • Organisation of appointments • Care provision meets needs
Low self-confidence	This is intrinsic to a person, not an attribute describing follow-up care received

The themes identified from the analysis of data generated from informal caregivers were 'Person centred information' and 'Person centred access to care'. Within the themes described above, the following attributes of care were identified, as described in Table 16 (objective 1-3).

Table 16. Attributes identified from analysis of informal caregiver data

Themes	Attributes
Person centred information	<ul style="list-style-type: none"> • Verbal information • Consultation • Written information • Information provision met within the multidisciplinary team • Frequency of care
Person centred access to care	<ul style="list-style-type: none"> • Holistic patient centred care • Expertise of the Health Care Professional • Continuity of Health Care Professional seen from pre-treatment to follow-up • The personality and attitude of the Health Care Professional • Medical examinations • Tests • Frequent appointments • Being given the time needed in the appointment • Unscheduled contact with the Specialist Nurse, to provide reassurance in between scheduled appointments and prompt re-access if required • Reciprocal communication • Organisation of appointments • Peer support • Clinic waiting time

Patients and their informal caregivers valued frequent appointments of a reducing frequency over the duration of care, because of the reassurance it provided by 1) having regular reassurance, 2) the feeling that the time between appointments increased was a reflection on the patient 'getting better'. This therefore aided coping with re-appraisal of perceived susceptibility as the frequency of care provision reduced (self-regulation model of illness: Leventhal et al., 1992; health belief model: Rosenstock, 1974; Becker, 1974; Becker & Rosenstock, 1984). Generally, patients and their informal caregivers were satisfied with the frequency of appointments delivered, follow-up appointments three monthly for the first two years, six monthly in the third year, and yearly in the fourth and fifth year (objective 5).

On the whole, as can be seen from the analysis by the lack of reporting of willingness to pay values, some patients and their informal caregivers struggled to understand this concept, in others it was not discussed in detail, and some would not discuss it in the face of the political climate of the future of the NHS at the time of data collection (BBC News 2014a; BBC News 2014b). As the concept of willingness to pay was not understood or discussed by

all, the discussion around willingness to pay was included in the analysis but not the actual figures that participants identified (objective 4). Additionally, income was not collected in the sociodemographic questionnaire, taking from the meaning that could have been generated from this question.

Through exploration of objectives 1–3 and 5, how patients and their informal caregivers felt that follow-up care could be improved was identified. This was very individual, through presentation of individual needs resulting in patients and their informal caregivers having a preference for personalised holistic care (objective 6).

3.4.1 Previous findings

This study supports previous findings, that gynaecological cancer follow-up patients have a need for reassurance (Bradley et al, 1999; Lydon et al., 2009; Oshima et al, 2011; Sekse et al, 2010). However, for some and as highlighted in the literature, the perceived reassurance from gynaecology cancer follow-up is a false reassurance due to the lack of efficacy of the traditional model of care in detecting asymptomatic recurrence and lack of efficacy of secondary treatment in increasing survival (Clarke et al., 2014; Elit et al., 2009; Fung-Kee-Fung et al 2006; Guerts et al., 2012; Lanceley et al 2013; Leeson et al., 2017; Lim et al., 2004; Nama et al., 2013; Rustin et al, 2010). The findings support the need for a changing model of care stratified by patient need, to meet the differing biopsychosocial needs experienced by patients at this point in their cancer pathway (National Cancer Survivorship Initiative, 2013). Alternative models of care may have a reduced cost to the NHS, allowing reinvestment of resources to better meet the needs of patients who require a more intensive follow-up pathway (Morrison et al, 2018; National Cancer Survivorship Initiative, 2013).

As highlighted above, there is no known prior research exploring the preferences of informal caregivers for gynaecological cancer follow-up, despite the known impact of caregiving on both the patient and informal caregiver (as discussed in Chapter 1). This research study helps to fill a gap in this knowledge. The findings regarding the role informal caregivers play in supporting patient's access the health care system has been previously reported in the literature (Williams, et al, 2013; Wolff & Roter, 2011). As emerged in this study, informal caregivers may feel isolated and feel like they have no one to communicate their feelings and concerns to, and may be "hidden patients" often ignored by the provision of health care (Brodaty & Donkin, 2009).

The findings of this qualitative chapter offer support to the Health Belief Model (Rosenstock, 1974; Becker, 1974; Becker & Rosenstock, 1984) and Theory of Planned Behaviour (Ajzen, 1985; Ajzen, 1991) in explaining preferences for care. The findings build on previous research that has applied the Health Belief model to explore the attitudes, barriers and compliance with hospital follow-up following surgery for colorectal cancer (Cardella et al., 2007). However, whilst Cardella and colleagues (2007) identified similar perceived benefits and barriers to care, the model did not account for all behaviour, with only 70% of clinic visits were attended by patients, only 49% of blood tests and only 62% of abdominal imaging appointments were completed. In a study applying the Theory of Planned Behaviour to explain attendance at GP led follow-up, Ngune and colleagues (2015) identified that similarly attitudes were significantly positively correlated with subjective norms and intention to attend General Practitioner led follow-up. Attitude was also significantly correlated with intention to see a specialist only for follow-up appointments. However, the authors did not report on whether behaviour was predicted by intention or perceived behavioural control (Ngene et al., 2015), and as illustrated above, intention does not always predict behaviour (Ajzen, 2011). This is also a key criticism of utility theory and discrete choice experiments (Quaife, Terris-Prestholt, Di Tanna & Vickerman, 2018).

This chapter also offers support to the self-regulation model of illness (Leventhal et al., 1992). This chapter identified a link between and individuals' illness appraisals and coping response, echoing findings from a systematic literature review and meta-analyses exploring illness representations, coping and illness outcomes in people diagnosed with all forms of cancer (Richardson, Schüz, Sanderson, Scott & Schüz, 2017). The review identified an effect of illness representations on coping strategies used and on a range of health outcomes (anxiety; depression; psychological wellbeing; psychological distress; role functioning; physical functioning and quality of life: Richardson, et al., 2017). Some cognitive illness representations and emotional representations had a small to large effect on coping strategies (cognitive reappraisal; avoidance/denial; problem focused coping), whilst most cognitive illness representations and emotional representations had a small to large effect on outcomes suggesting more support for direct effects rather than effects being mediated by coping (Richardson et al., 2017).

3.4.2 *Strengths and limitations*

Firstly, using a health economic approach whilst acknowledging and seeking to understand the underlying attitudes and beliefs governing these from a health psychology perspective, added significant important findings to this study. Asking open ended inductive questions rather than merely using a reductionist nominal group task (as is commonly used in health economics to generate attributes for a discrete choice experiment: Hiligsman et al., 2013; Powell et al., 2015), ensured that the data generated was a thorough and rich representation of participants' thoughts, feelings, emotions and preferences, rather than a prompted stated preference. Key health psychology theories emerged from the data throughout the iterative data analysis. Patients and their informal caregivers' illness representations and coping strategies offer support to the self-regulation model of illness (Levethal et al., 1992). Patients and their informal caregivers attitudes and beliefs regarding their cancer diagnosis and follow-up care, offer support to the Health Belief Model (Rosenstock, 1974; Becker, 1974; Becker & Rosenstock, 1984) and Theory of Planned Behaviour (Ajzen, 1985; Ajzen, 1991). Using a health economic and health psychology approach was a key asset to this study and significantly adds to the novel contributions this study makes (see page 177-178). Secondly, the study was designed in accordance with the guidance outline by Barbour (2008) and Krueger and Casey (2009), and reported in accordance with the COREQ checklist (Consolidated criteria for reporting qualitative research: Tong, Sainsbury & Craig, 2007), and together with the information provided in Chapter 4 and 5, meet the requirements for the reporting of qualitative research for attribute generation (Coast et al., 2012). Patients were recruited purposefully, and the sample was reflective of the population in terms of incidence rate and five year survival for endometrial, cervical and ovarian cancer (Office for National Statistics, 2013). Whilst this is not a prerequisite for qualitative research, it was important given the overarching aims and objectives of the PhD. Thirdly, the study was conducted using focus groups and face-to-face and telephone interviews to ensure that those who were unable to make a focus group due to co-morbidities, other commitments or the geographical constraints of living in a semi-rural area were able to partake, enabling the sample to be more representative of the population, reducing selection bias (Barbour, 2008; Thompson et al., 2003a; Thompson et al., 2003b). Fourthly, the focus groups were held on the Tenovus Mobile Unit, to help create a relaxed and supportive environment outside of the NHS, and face-to-face interviews were held in the participants' home. This is important when encouraging and

facilitating open discussion about personal and sensitive issues and evaluating services (Krueger and Casey, 2009). Fifthly, the semi-structured interview guide was designed for this research study, and used both a top-down and bottom-up approach to identify preferences for care in accordance with the interdisciplinary nature of this thesis.

Whilst a strength of this research is the co-use of focus groups and interviews (face-to-face and telephone interviews), it is also a weakness. Data elicited in a focus group are embedded in that context and different preferences may have been elicited if focus group participants took part in interviews. For this reason, focus group data from individual participants should not be taken to be representative of individuals' views. For this reason, it has been reported that interviews are more suited to collecting data about individuals' attitudes (Barbour, 2008). However, the reflection and interplay between participants encouraged elicitation of experiences and preferences for a sensitive subject, as has been previously reported in the literature (Crabtree et al., 1993; Guest, Namey, Taylor, Eley & McKenna, 2017; Kitzinger & Barbour, 1999; Krueger & Casey, 2009; Morgan, 1988). Nevertheless participants might feel more comfortable in a 1-1 setting, and in their home environment because it stimulates a more natural every day 'setting' (Norvick, 2008; Shuy, 2002). One telephone interview elicited the same level of detail as face-to-face interviews, however in the other telephone interview the poor telephone line which meant it was hard to hear and understand each other, as has been previously identified in the literature (Carr & Worth, 2001; Sturges & Hanrahan, 2004; Sweet, 2002). Despite the different methods used to elicit data using the same protocol, the overall narratives from participants were consistent regardless of the methods. Vulvar patients and their respective informal caregivers were not recruited given the original recruitment criteria for the feasibility trial that this PhD is linked to (Morrison et al., 2018). However this meant that the experiences and preferences of vulvar patients and their informal caregivers were not identified and explored, and so the results cannot be generalised to vulvar cancer as experiences, needs and preferences of care differ by cancer diagnosis (Morrison et al, 2012). It must be acknowledged that participation bias/nonparticipation bias may occur particularly when recruiting participants to qualitative research, (Johnson & Wislar, 2012; Råheim et al., 2016). This may have been amplified by the conduct of focus groups on the Tenovus cancer care mobile unit. One participant spoke to the Tenovus cancer care employee following the focus group, and another confirmed on the telephone that a Tenovus cancer care employee would be available to talk to after the focus

group. It was unknown why eight screened and eligible patients were not given the recruitment pack. This could be due to the high work load of the Health Care Professionals who screened participants, or due to gatekeeping, but nevertheless is a potential source of bias.

3.4.3 Future research

Future research should explore patients and their informal caregiver's preferences for gynaecological cancer follow-up using a larger sample size, and explore the trade-offs between attributes of follow-up care that patients and their informal caregivers would be prepared to make. This understanding would reduce preference misdiagnosis, and could be used by health care commissioners to reduce the inefficiencies of, and ensure prudent service design, management and delivery of gynaecology cancer follow-up care (Mulley et al., 2012; Welsh Government and NHS Wales, 2016).

3.5 Conclusion

This study aimed to identify and explore patients and their informal caregivers' preferences gynaecological cancer follow-up care, based on their experience of this care. This study identified that patients and their informal caregivers' preferences for attributes of care are based on their differing needs for reassurance. Patients valued having 'access to an expert', 'medical procedures' and 'holistic care', to meet their needs for reassurance. 'System failures' and 'low self-confidence' prevented access to care and so the reassurance that patients felt they needed. Informal caregivers valued 'person centred information' to meet their need for reassurance, which in turn was met through provision of 'person centred access to care'. Future research should explore the acceptability of alternative models of care, and the trade-off's that patients and their informal caregivers are prepared to make between these attributes of care to better meet patients' follow-up needs and at a reduced cost to the National Health Service.

3.6 Novel contributions

3.6.1 Methodological contributions

- Despite guidance on the importance of qualitative research to aid the design of discrete choice experiments, there is still a dearth of transparent reporting of qualitative research for this purpose (Vass, Rigby & Payne, 2017). The methodology used and transparent reporting of this, can undoubtedly be used to aid the design of qualitative research conducted in health economics for this purpose.
- There is a lack of guidance on how to design, conduct, and analyse focus groups to aid the design of a discrete choice experiment, despite this methodology being recommended for this purpose (Louviere et al., 2000). The methodological rigour of the study and the transparency of reporting (Coast et al., 2012; Tong, Sainsbury & Craig, 2007), can be used to aid health economists in the design, conduct and analysis of qualitative research for the purpose of discrete choice experiment design.

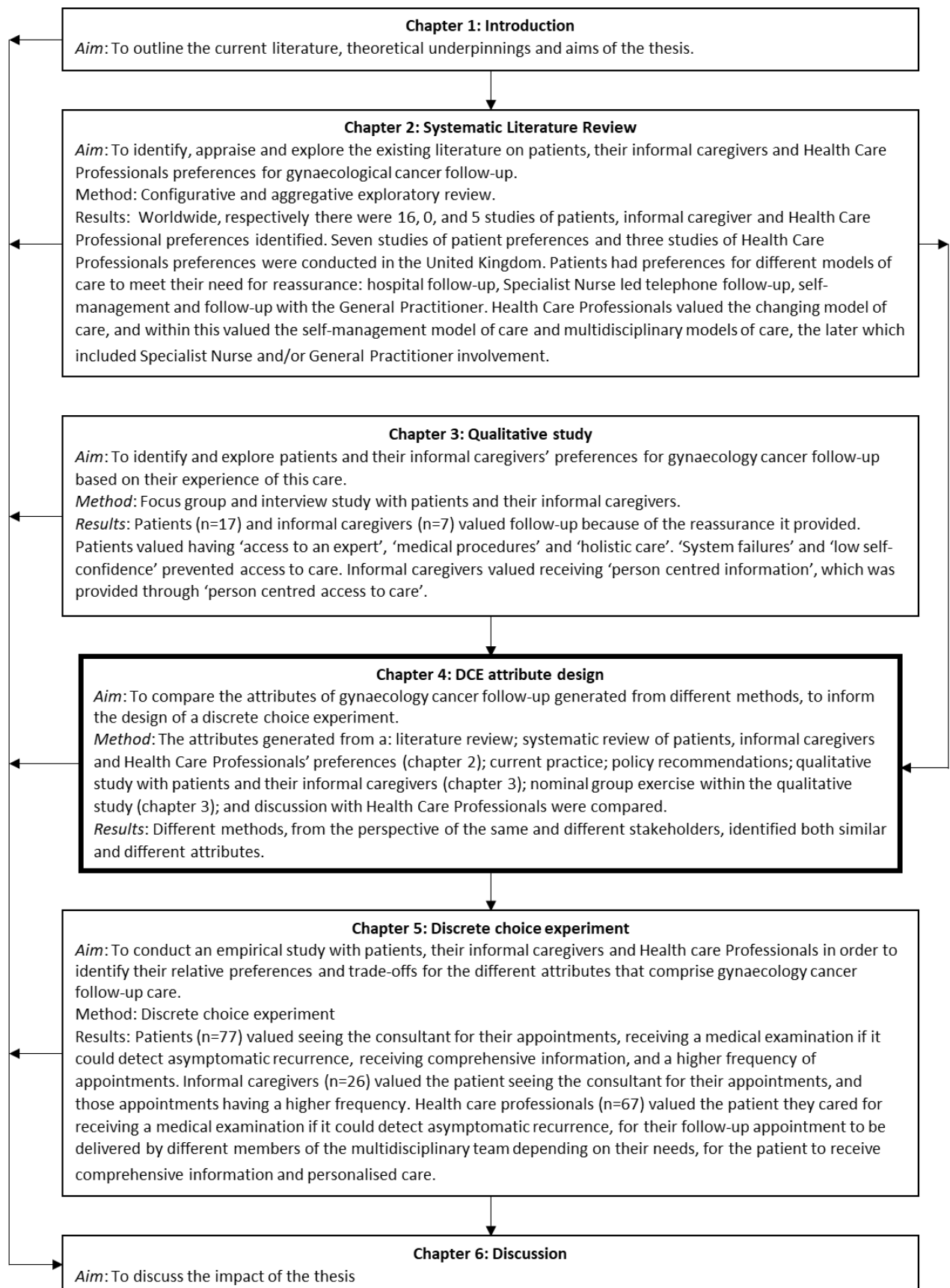
3.6.2 Policy contributions

- This research study adopted an interdisciplinary health psychology and health economic perspective in understanding preferences for care, based on experiences of care. The inductive and deductive nature of the study ensured that attributes valued by stakeholders, and the reason for this were identified. Analysis using the Framework approach ensured that the themes generated described the model of care. As such, this is the first study to identify attributes of care valued by endometrial, ovarian, and cervical cancer patients diagnosed at different stages of disease, whilst valuing their experience of care, attitudes and beliefs. This is of key importance when aiming to understand preferences for care, rather than merely identifying what they are. As such, this study can be used to aid policy makers in their decision making for the prudent changing model of care.
- This is the first study conducted to identify the preferences of informal caregivers for gynaecology cancer follow-up, despite the importance of informal caregiving and so policy focus on understanding the preferences of informal caregivers. Therefore this study provides valuable insight to policy makers.

- The findings from this study were used to aid the design of a subsequent discrete choice experiment (Chapters 4, 5), to further explore patients and their informal caregivers' preferences for gynaecological cancer follow-up care, and the trade-offs between attributes of care that patients and their informal caregivers were prepared to make.

Chapter 4

**Attribute generation in discrete
choice experiments: does the
methods used affect the attributes
identified?**



4.1 Chapter Summary

Chapter 1 identified a need to identify patients, their informal caregivers and Health Care Professionals' preferences for gynaecology cancer follow-up care. A systematic literature review identified a dearth of evidence and that a discrete choice experiment to explore patients, their informal caregivers and Health Care Professionals' preferences for gynaecology cancer follow-up care had not been previously conducted (Chapter 2). It is unknown the extent to which the use of different independent research methods results in the identification of different attributes. Therefore, with the overarching aim of designing a discrete choice experiment, the attributes generated from different methods, and from the perspective of different stakeholders were compared. Attributes generated from the following methods were compared: a literature review; a systematic review of preferences for care (Chapter 2); current practice; policy recommendations; a qualitative study with patients and their informal caregivers (Chapter 3); a nominal group exercise with patients and their informal caregivers (Chapter 3); and informal discussion with Health Care Professionals. Attributes were found to vary within and between stakeholders and were dependant on the methods used to identify them, thus supporting previous literature and highlighting the importance of a clear rationale for the choice of methods used. This paper 1) continues the methodological debate for discrete choice experiment design, and encourages health economists to have a clear rationale for the choice of methods to use when designing a discrete choice experiment, 2) raises important implications for the validity of policy decisions informed by discrete choice experiments.

4.2 Introduction

The aim of a discrete choice experiment is to understand the population of interests' relative preferences for a good or service, and the trade-offs that the population are prepared to make to receive their preferred care. There are five phases to the design of a discrete choice experiment, 1) generation of attributes, 2) definition of attribute levels, 3) identification of the choices/questions, 4) data collection, and 5) analyses (Ryan & Farrar, 2000). This study focuses on the first stage of discrete choice experiment design, the generation of attributes.

4.2.1 *Attribute requirements*

There are theoretical requirements for attributes. To comply with consumer choice theory, it is a requirement that attributes are relevant and important to stakeholders (both providers and consumers of care), plausible, and have the capacity to be traded. The capacity to be traded refers to the ability to trade levels of one attribute for a one unit increase in another, for example: to trade General Practitioner led care for Consultant led care, for a 3 month reduced frequency of appointments (Ryan, 1996).

To comply with random utility theory, all attributes that are important to decision making for a good/service should be included, as not including all important attributes may affect the validity of the findings (Coast et al, 2012; Lanscar & Louviere, 2008). This highlights the importance of conducting methods to ensure identification of all of these attributes. However, this could result in an overly long and complex discrete choice experiment as the number of attributes effects the number of questions that need to be asked, so may be too complex for respondents and so fail to elicit their preferences for care (DeShazo & Fermo, 2002; Lanscar & Louviere, 2008). Therefore, it is acceptable to inform participants that for attributes of care not described, to assume that they do not differ between choices (for example, do not differ between 'Service A' and 'Service B': Bridges et al, 2011). It is important to ensure that attributes not used to describe the model of care, are not associated or correlate with the included attributes. For example, participants may assume that the cost of a service and an improvement in the service provision are connected, so assume increased cost means improved service and use this to inform their decision for which bundle of attributes they prefer (Bridges et al, 2011). Attributes should not be too close to the latent construct they are measuring (for example if the latent construct is utility and the attribute

patient satisfaction). If this was the case, the attribute patient satisfaction could dominate decision making and so provide no information about other attributes which affect it (Coast et al., 2012). One attribute should not dominate decision making for a majority of participants so that no errors in decision making are made (Coast et al, 2012; Flynn et al., 2008). To ensure that the attributes can be manipulated, attributes should not be intrinsic to a person (Coast et al, 2012; Hensher, et al, 2003).

Attributes can be qualitative or quantitative, but require at least one quantitative attribute so that marginal rates of substitution can be explored (the amount of money/time respondents are willing to trade for a one unit increase in another attribute: Bridges et al., 2011).

4.2.2 Methods used to generate attributes

There is a dearth but emerging body of research for the choice and conduct of methods to generate attributes, to ensure that the requirements described above are met. However, generally there is a lack of focus of the importance of high quality evidence and transparency in the conduct and reporting of methods used to generate attributes. The generation of attributes determines the design of the discrete choice experiment, its results and any policy decisions made from these. If attributes valued by participants are not included or participants do not understand the meaning of the attributes, then it affects the validity of the results and any policy decisions made based on these (Coast et al., 2012). Coast and colleagues (2012) emphasise that the decision of the methods used to generate attributes should be clear and transparent and influenced by the available resources, available existing literature and sensitivity of the subject (Coast et al., 2012).

Checklists of items to consider when assessing the quality of a discrete choice experiment ask reviewers to consider how the attributes were generated (Lanscar & Louviere, 2008), or whether the choice of attributes and levels was supported by evidence and is consistent with economic theory (Bridges et al., 2011). The narrative supporting these checklists focus on what the attributes should look like theoretically, and there is no guidance on what quality of evidence is 'good enough', instead simply stating that literature reviews, discussion with stakeholders and qualitative research can be conducted (Bridges et al., 2011; Lanscar & Louviere, 2000). Guidance for the minimum reporting of qualitative research for attribute generation has been reported (Coast et al., 2012). However, a recent systematic

literature review identified that still, few studies are transparently reporting their use of qualitative methods (Vass et al, 2017). Such poor reporting leads some to question the rigour of the resulting discrete choice experiment (Coast & Horrocks, 2007; Coast et al., 2012; Vass et al., 2017).

Currently endorsed methods used to generate attributes for a discrete choice experiment include: narrative literature reviews (Howard, Salkeld, Patel, Mann, & Pignone, 2015; Kunaviktikul, Chitpakdee, Srisuphan, and Bossert, 2014; Robinson, et al., 2015); systematic literature reviews (Lynn, Crealey, Alderdice, and McElnay, 2015); qualitative research (Coast et al., 2012; Vass et al., 2017); discussion with stakeholders (Gerard, Tinelli, Latter, Smith & Blenkinsopp, 2014; Howard et al., 2015; Kateeb et al., 2014; Lynn et al., 2015); the nominal group technique (Hilgsmann et al., 2013; Powell et al., 2015); policy questions (Dixon, Nancarrow, Enderby, Moran & Parker, 2015); existing measures (Viney et al., 2013); and surveys (Gerard et al., 2014).

The use of literature reviews to generate attributes are recommended, as existing literature could define the policy context and characteristics of the service (Abihiro et al., 2014; Bridges et al., 2011; Lanscar & Louviere, 2008). However, it has been argued that literature reviews, whilst recommended, are often inadequate due to a dearth of relevant existing literature, so all attributes that affect an individuals' decision making are not identified (Coast et al., 2012). There is no known discussion around the requirement for the quality and transparency of literature reviews conducted for this purpose, or how attributes can be generated from these. Discrete choice experiments that have used literature reviews to generate attributes have generally not reported details of the literature review, or how they generated attributes from this, but simply stated that a literature review was conducted (Hall, Fiebig, King, Hossain & Louviere, 2006; Howard, et al., 2015; Kunaviktikul et al., 2014; Robinson et al., 2015).

Attributes can be generated directly in response to a policy question (Dixon et al., 2015). However, as argued by Coast and colleagues, this can mean that attributes important to patients' decision making are not included in the discrete choice experiment (Coast et al., 2012).

Qualitative research is recommended for attribute generation (Bridges et al., 2011; Coast & Horrocks., 2007; Coast et al., 2012; Kløjgaard et al., 2012; Louviere, et al, 2000; Lanscar & Louviere, 2008). The use of qualitative research to generate attributes has been reported

to improve the content validity of the discrete choice experiment, as 1) it ensures that attributes that effect the populations decision making for a specified good or service are identified (Mangham, et al, 2008), 2) aids understanding of the perspective, and experience of the target population (Coast & Horrocks, 2007; Hall, et al, 2004), and 3) aids identification of context specific factors which are not specified in the existing literature (Abihiro et al., 2014). There is more detailed recommendations for the use of qualitative research to generate attributes (Coast & Horrocks., 2007; Coast et al., 2012), and a recent systematic literature review has explored the use and reporting of qualitative methods to inform the design and/or aid the interpretation of a discrete choice experiment (Vass, et al, 2017). Coast and colleagues (2012), discuss the qualitative research they used to design eight research studies, what qualitative methods they used and the impact of these. They explain that through the findings of their research, they advocate the use of two stages for the generation of attributes 1) conceptual development and 2) refinement of language to convey meaning. The authors argue that the use of qualitative methods is superior to the use of other methods for attribute generation, as it 1) enables the generation of attributes that are valued by the recipients of care, which prevents mis-specification of attributes valued in decision making by over reliance on experts or the researchers views, 2) enables the discussion of sensitive topics and discussion of factors that are not observed to effect decision making, 3) through rigorous analysis researchers can identify a comprehensive list of attributes valued by participants, 4) attributes that have a small value in decision making can be incorporated into the list of attributes, 5) ensures that the language used conveys the desired meaning, and 6) prevents the use of clinical or research terminology which may be meaningless to the population (Coast et al., 2012).

As discussed above there is an increasing research focus on the development of systematic phased models of attribute generation and refinement. Helder and Boehler (2016) conducted a systematic literature review and from these findings proposed and tested a phased model. Coast and colleagues (2012) discussed the use and benefit of a staged qualitative approach from their experience of conducting eight discrete choice experiments. Whilst Abihiro and colleagues (2014), Kløjgaard and colleagues (2012) and De Brún and colleagues (2018), discussed the phased approach that they used in one discrete choice experiment. Generally, earlier stages are concerned with attribute generation, followed by reduction of attributes in accordance with theoretical requirements, and clarification of

wording to convey the desired meaning (Abihiro et al., 2014; Coast et al., 2012; De Brún et al., 2018; Helter & Boehler, 2016; Kløjgaard, Bech & Sógard, 2012). Some of these staged models include pilot testing (Abihiro et al., 2014; De Brún et al., 2018; Kløjgaard et al., 2012). One of these models discussed the development and refinement of attributes only (Helter & Boehler, 2016), whilst others included the development and refinement of attributes and the levels in which they were specified (Abihiro et al., 2014; Coast et al., 2012; De Brún et al., 2018; Kløjgaard et al., 2012).

It has been recommended that a comparison of the attributes identified from different methods is made (Coast and Horrocks, 2007; Coast et al., 2012; Ryan, Watson, Krucien, and Heidenreich, 2014). Following the conduct of this study, the following methodological comparison has been published. Helter and Boehler (2016) compared a four stage qualitative approach to a more reductionist four stage approach for the purpose of generating attributes for a public health intervention. The qualitative approach was comprised of interviews with experts and focus groups with the public, followed by the researchers' judgement. The more reductionist approach was comprised of a literature review, expert opinion, a ranking exercise with the public, and researchers' judgement. The different sequential methods identified both similar and different findings (Helter & Boehler, 2016). Nine attributes were identified in the qualitative approach, but six in the more reductionist approach. Five of the attributes generated in the reductionist approach were also identified in the qualitative approach. This case study illustrates that different mixed method approaches to identifying attributes for a discrete choice experiment, may identify different attributes (Helter & Boehler., 2016). This raises important questions for the choice of methods to use in attribute generation, echoing concerns previously stated in the literature (Coast et al., 2012). However, the study does not compare the conduct of multiple single methods, and so is essentially comparing attributes generated from a qualitative study to a literature review, which because of their subjectivity is recommended that they should not be used to inform decision making (see below: Gough et al., 2012; Munn et al., 2018; Robinson & Lowe, 2015). Further research, comparing the attributes generated from different methods, and in different decision making contexts is needed.

4.3 Aim

Given the above, this chapter aimed to compare the attributes of gynaecology cancer follow-up generated from different methods, to inform the design of a discrete choice experiment.

4.4 Methods

Attributes generated from: a literature review; a systematic review of patients, informal caregivers and Health Care Professionals' preferences (Chapter 2); current practice; policy recommendations; a qualitative study with patients and their informal caregivers (Chapter 3); a nominal group exercise within the qualitative study (Chapter 3); and discussion with Health Care Professionals are discussed below.

In the context of gynaecology cancer follow-up care it was important to identify attributes that were important to patients (as the direct recipient of care), their informal caregivers (as the 'invisible patient' who provides informal support to the patient), the Health Care Professional (as preferences of Health Care Professionals influence the model of care delivered), and policy makers (given the policy focus of a prudent changing model of care). As such, and as illustrated in table 17, these methods were conducted from the perspective of different stakeholders. The systematic literature review (chapter 2), qualitative study (chapter 3) and ranking exercise (within the qualitative study, chapter 3) were conducted from the perspective of patients. The qualitative study and ranking exercise were conducted from the perspective of informal caregivers (as was the systematic review, but this yielded no empirical papers exploring the preferences of informal caregivers). Exploration of current practice, the systematic review and informal discussion with Health Care Professionals, was conducted from the perspective of Health Care Professionals. Current practice and policy recommendations were explored from the perspective of policy makers. The systematic literature review whilst conducted from the perspective of patients, their informal caregivers and Health Care Professionals, was conducted with the overarching aim of informing policy.

For the purpose of this comparison, each method was conducted in isolation, so the use of one method did not influence the design of a subsequent method (for example, the qualitative study did not influence the informal discussion with Health Care Professionals).

4.4.1 Literature review

Traditional narrative literature reviews are commonly used to generate attributes (Bessen et al., 2014; Burton et al., 2014; Hall et al., 2006; Howard., et al 2015; Kunaviktikul et al., 2014; Robinson et al., 2015). Narrative literature reviews summarise theory and empirical evidence surrounding a given topic. Unlike systematic literature reviews, traditional narrative reviews do not require 1) a review protocol to be written and published a priori, 2) a transparent and explicit search strategy, 3) standardised forms to extract and critically appraise the data, 4) data synthesis. As such, narrative literature reviews are highly subjective and do not provide concrete evidence, so it is recommended that they are not be used to inform health care decision making (Gough et al., 2012; Munn et al., 2018; Robinson & Lowe, 2015). As such, they are not used in this study.

4.4.2 Systematic literature review of patient, informal caregiver and Health Care Professional preferences

The aim of systematic literature reviews is to inform practice, policy and future research (Aromataris & Pearson, 2014; Higgins & Green, 2011; Liberati et al., 2009; Munn et al., 2018; Person, 2004). Use of systematic methods reported explicitly, and chosen with an aim to minimise bias, provide reliable findings in which conclusions to particular questions can be drawn. As such, systematic literature reviews are a trustworthy evidence base in which to inform future research and health care policy decision making (Aromataris & Pearson, 2014; Higgins & Green, 2011; Liberati et al., 2009; Munn et al., 2018; Pearson, 2004; Pearson, Jordan & Munn, 2012; Steinberg, Greenfield, Mancher, Wolman & Graham, 2011).

Contextually, due to insufficient evidence, there is no clinical consensus as to the most effective and cost-effective model of gynaecology cancer follow-up care. The majority of research has focused on medical tests to detect recurrence, however, such tests are not routinely delivered due to the nature of gynaecological cancer, and evidence regarding its detection and survival following diagnosis of a recurrence (see Chapter 1). As such, in this clinical decision making context, development of attributes from systematic literature reviews of effectiveness and cost effectiveness would not of be of benefit.

A systematic literature review of patients, informal caregivers and Health Care Professionals' preferences for gynaecological cancer follow-up care was conducted (see Chapter 2). An objective of this systematic review was to generate attributes for the

subsequent discrete choice experiment (see Chapter 5). The EPPI centre guidance was used to guide the methodology and conduct of the systematic review (Gough et al., 2012), and the PRISMA checklist for the reporting of systematic reviews was followed (Moher, et al., 2009: see Chapter 2 for detailed methodology).

4.4.3 Current practice

An audit of current practice was used to identify the current model of care delivered in the United Kingdom (Leeson et al., 2013). The audit was read and re-read, to identify the attributes comprising the current care model of care.

4.4.4 Policy guidance

Guidance regarding the changing model of cancer follow-up was used to generate attributes valued by policy makers (National Cancer Survivorship Initiative, 2013). The guidance was read and re-read, to identify the attributes comprising the changing model of follow-up care within the United Kingdom.

4.4.5 Focus groups and interviews with patients and their informal caregivers

Focus groups and interviews (when participants were unable to make a focus group) were held with patients (n=17) and their informal caregivers (n=7) with the aim of generating attributes valued by participants based on their experience of this care. A semi-structured interview guide was used (see Figure 13), and the data was analysed using the Framework approach in accordance with the aim of the study (Richie & Spencer, 1994). Results were analysed separately for patients and their informal caregivers (the detailed methodology and results are presented in Chapter 3).

Figure 13: Semi-structured interview guide

<p style="text-align: center;"><u>Semi-structured interview guide</u></p> <ul style="list-style-type: none">• What is the purpose/aim of gynaecology cancer follow-up• What are the attributes of gynaecology cancer follow-up• What attributes of gynaecology cancer follow-up are important to you (individual ranking task, then nominal group discussion) <p>Case studies:</p> <p>After each case study was read out by the researcher, participants were asked: “What do you think her needs are, and how can follow-up care meet those needs?”</p> <p>Case study 1: Maggie is fifty years old and was diagnosed with early stage endometrial cancer last year. She goes to see the consultant every 3 months to have an internal examination to check that the cancer hasn’t returned. Maggie found the first couple of weeks after her hysterectomy hard, but has since returned to work and is living the life she lived before.</p> <p>Case study 2: Ann is seventy years old and was diagnosed with an advanced form of ovarian cancer four years ago. Ann goes to see the Specialist Nurse once a year for an internal examination to check that the cancer hasn’t returned. Ann has struggled with daily living since she was treated for the cancer. Because of her treatment she has had problems with her bowel functioning, which she still suffers with.</p> <p>Participants were then asked, thinking about these case studies,</p> <ol style="list-style-type: none">1) What is good about the follow-up care that you/the person you care for receives?2) Are any of your needs/the person you care for’s needs not met by their follow-up care appointments? Which ones?3) What aspects of follow-up care do you think could be improved?4) How long would you like between follow-up appointments?5) Hypothetically, if you had to pay for follow-up after treatment care, how much would you be willing to pay per follow-up visit?
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4.4.6 Nominal group task (within focus groups and interviews)

The nominal group technique is a stepped process used in health care research to identify and prioritise participants’ responses to a question (Vella, Goldfrad, Rowan, Bion & Black, 2000). It can be used to identify the importance of possible attributes to make a decision for the attributes to include in the discrete choice experiment (Bridges et al., 2011). As such, there is

an emerging use of ranking exercises for this purpose (Hiligsman et al., 2013; Powell et al., 2015). As illustrated by Figure 13, firstly, patients and informal caregivers were asked to identify attributes of their follow-up care. Secondly, participants were asked to rank as an individual (to ensure that their independent voice was heard), and in a group setting (to aid identification of the attributes valued by the majority) their perceived importance of attributes of care (see Figure 13).

4.4.7 Informal discussion with Health Care Professionals

An informal discussion was held with different Health Care Professional's (senior Clinical Medical Oncologists, a Specialist Nurse and a senior Primary Care Practitioner), with the aim of generating attributes of gynaecology cancer follow-up that they perceived to be of the most benefit to patients (see Figure 14). For illustrative purposes, Health Care Professionals were given an example of an attribute and respective levels. Health Care Professionals were asked to rank the attributes they identified in order of importance.

Figure 14: Informal discussion with Health Care Professionals

<p style="text-align: center;">Informal discussion with Health Care Professionals.</p> <ol style="list-style-type: none">1. What are the 5 aspects of gynaecological cancer follow-up care that you think are the most important for patients?2. And what aspect levels do you think should be used to describe these?
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4.5 Results

The attributes generated from the aforementioned sources are presented in Table 17. The purpose of this study was not to advocate which method is 'best', but to highlight the differences in findings and to question the impact this may have on the subsequent discrete choice experiment and ultimately, policy decisions. Different methods resulted in the identification of both similar and different attributes. Attributes valued by stakeholders differed both between and within stakeholder groups (patients, informal caregivers, Health

Table 17. Attributes of gynaecology cancer follow-up generated from each method

	Systematic literature review (patients)	Systematic literature review (HCP's)	Current practice	Policy guidance ⁺	Discussion with HCP	Qualitative study (patients)	Qualitative study (caregivers)
Stakeholder	P, PM	HCP, PM	HCP, PM	PM	HCP	P	C
Consultation itself	X	X	X		X	X	X
Time spent in consultation							X
Organisation of appointments					X	X	X
Holistic patient centred care	X	X		X	X	X	X
Care stratified by risk of recurrence				X			
Information	X			X		X	X
Reciprocal communication	X	X		X	X	X	X
Medical Exam	X	X	X		X	X	X
Tests	X	X	X			X	X
Expertise of HCP	X	X	X	X	X	X	X
Continuity of care				X	X	X	X
Attitude of Health Care Professional	X	X				X	X
Protocol based care			X		X		
Transparent protocol based care				X	X		
Mode (face-to-face location/telephone/self-management)	X	X	X	X	X	X	
Patient involved in decision making			X	X			
Peer support						X	X
Contact details for Health Care Professional if concerns			X	X	X	X	X
Prompt re-access if required			X	X	X	X	X
Frequency of appointments	X	X		X	X	X	X
Duration (number of years have appointments for)		X	X		X	X	
Clinic waiting time	X						X
P=patients C=informal caregivers HCP=Health Care Professionals PM=policy makers							

Care Professionals, and policy makers). This finding raises the important question regarding the methods used to develop attributes for a discrete choice experiment.

4.5.1 Systematic review of patient, informal caregiver and Health Care Professionals' preferences

The systematic review generated attributes of care valued by patients and Health Care Professionals, but from the overarching perspective of policy makers and health care commissioners given the overarching aim to influence policy and practice (Aromataris & Pearson, 2014; Higgins & Green, 2011; Liberati et al., 2009; Munn et al., 2011; Person, 2004).

Within the timeframe of the PhD programme, the descriptive map was conducted before the conduct of this methods review, but the synthesis was conducted after (see Chapter 2 for difference between a descriptive map and synthesis). Therefore attributes identified from the descriptive map were used for this mixed method comparison (see Tables 4-7 in Chapter two for a breakdown of the attributes generated from individual empirical papers of patients and Health Care Professionals preferences). The descriptive map of patients' preferences for care identified the following attributes, as described in Table 18, translated to the language used to describe attributes in this mixed method comparison. The descriptive map identified no studies of the preferences of informal caregivers. The descriptive map of Health Care Professionals preferences for care identified the following attributes of follow-up, as described in Table 19, translated to the language used to describe attributes in this mixed method comparison. Studies identified in the systematic literature review: were not of high methodological quality; generally used quantitative methods of preference identification, and so used a top down approach; did not aim to identify all attributes valued by the sample concerned; did not recruit endometrial, ovarian and cervical cancer patients with different stages of disease; and finally, no studies explored the preferences of informal caregivers.

4.5.2 Current practice

As highlighted in Chapter 1, there is no consensus regarding the model of gynaecology cancer follow-up that is most effective and so should be delivered. An audit of current practice conducted in 2012 identified that the model of care delivered in the United Kingdom differs

by local protocol, and is delivered over a specified duration by an expert in gynaecological oncology (with patient involvement in decision making regarding which Health Care

Table 18. Attributes generated from systematic review (patient's perspective)

Attributes generated	
Expertise of health care professional	<ul style="list-style-type: none"> • Standard Consultant/hospital Doctor led care • Specialist Nurse led care • General Practitioner led care
Mode	<ul style="list-style-type: none"> • Face-to-face • Telephone follow-up • Self-management
Consultation itself	<ul style="list-style-type: none"> • Consultation itself • Monitor signs of recurrence
Reciprocal communication	<ul style="list-style-type: none"> • Health Care Professional listen to concerns • Health Care Professional answers questions • Onward referrals
Attitude of Health Care Professional	<ul style="list-style-type: none"> • Health Care Professional listen to concerns • Health Care Professional answers questions
Holistic patient centred care	<ul style="list-style-type: none"> • Holistic care • Survivorship care plan • Holistic discussion with Health Care Professional • Manage treatment related morbidities
Information received	
Medical examination	
Diagnostic tests	<ul style="list-style-type: none"> • Location of CA125 testing • Delivery of results from diagnostic tests
Frequency of appointments	
Financial cost of attending appointment	Identified from study conducted in Thailand. Attribute not used for purpose of comparison as was not directly applicable to UK context
Travel time	Identified from study conducted in Thailand. Attribute not used for purpose of comparison as was not directly applicable to UK context
Preferences for care at provincial hospital	Identified from study conducted in Thailand. Attribute not used for purpose of comparison as was not directly applicable to UK context
Clinic waiting time	

Table 19. Attributes generated from systematic review (Health Care Professionals perspective)

Expertise of Health Care Professional seen	<ul style="list-style-type: none"> • Hospital doctor • Specialist Nurse • Nurses • General Practitioner
Mode of care	<ul style="list-style-type: none"> • Face-to-face • Self-management • Telephone appointments • Location of appointments
Holistic care	
Consultation itself	
Medical Examination	
Tests	
Reciprocal communication	<ul style="list-style-type: none"> • Health Care Professional listens to concerns • Health Care Professional answers questions
Attitude of Health Care Professional	<ul style="list-style-type: none"> • Health Care Professional listens to concerns • Health Care Professional answers questions
Frequency of appointments	
Duration of care	

Professional is seen for appointments), either face-to-face in the hospital/over the telephone/via patient initiated care. However, is generally conducted face-to-face in the hospital. The audit did not report on whether a medical examination took place but this was an assumed part of the consultation, with tests delivered, if any, varying by location and type of gynaecological cancer diagnosis. The audit did not report on frequency of care but frequency of differing tests. Patients generally had prompt re-access to the clinic if there were concerns regarding recurrence in between scheduled appointments, but were often not given contact details for Health Care Professionals with whom to raise these concerns (Leeson et al., 2013). See Table 17 for attributes generated from current practice. Exploration of current practice was essential in ensuring that the attributes included in the discrete choice experiment were relevant and plausible (Ryan, 1996).

4.5.3 Policy guidance

As described in Chapter 1, there is a policy emphasis on the changing model of aftercare. There is a policy focus on holistic patient care stratified by risk of cancer recurrence and cancer related needs. It is recommended that there are three levels of care, 1) supported self-management with open access if the patient has any concerns, 2) shared care between the patient and Health Care Professional, 3) complex care managed with the multidisciplinary team. Central to each stratification is transparent information and reciprocal communication between the patient and health care provider. See Table 17 for attributes generated from policy recommendations. Policy recommendations (from the perspective of the policy maker) were therefore of key importance in the generation of attributes, ensuring that the attributes were relevant and plausible (Ryan, 1996).

4.5.4 Qualitative study with patients

Focus groups and interviews were conducted to identify the preferences of patients based on their illness appraisals, experience, attitudes and beliefs. Analysis using the Framework approach (Richie & Spencer, 1994) identified that patient's valued gynaecology cancer follow-up due to the reassurance it provided. Two themes were identified each with sub-themes. The theme 'meet my needs please', with the sub themes 'access to an expert', 'procedures give' and 'holistic care' were valued because of the reassurance they provided. The theme 'barriers to care', with the sub themes 'system failure' and 'low self-confidence' prevented access to care and therefore reassurance from being obtained. Within these themes, multiple attributes of care were identified. See Table 15 for a breakdown of the attributes generated from the qualitative study with patients. See Chapter 3 for a detailed discussion of the results, and so detailed discussion of attributes valued by patient participants. This qualitative study provided information on the attributes valued by patients, which is essential in discrete choice experiment design (Ryan, 1996).

4.5.5 Qualitative study with informal caregivers

Focus groups and interviews were conducted to identify the preferences of informal caregivers based on their experience of care. Analysis of results using the Framework

approach (Richie & Spencer, 1994), identified that informal caregivers valued follow-up care due to the reassurance it provided. Informal caregivers felt their need for reassurance was met through the provision of 'person centred information' which in turn was met through having 'person centred access to care'. Multiple attributes of care were identified within these themes. These attributes are illustrated in Table 16, and discussed in detail in Chapter 3. The preferences of patients were assessed as being of most importance as they are the target of follow-up care, with an informal caregiver's role being to support patients, in part with accessing this care. Due to the caregiving experience, informal caregivers may become an 'invisible patient' and there is a policy focus on understanding the preferences of informal caregivers (see Chapter 1 for further discussion). Therefore it was important to value the preferences of informal caregivers when designing the discrete choice experiment.

4.5.6 Nominal group task (within focus groups and interviews)

Participants struggled with the prioritisation aspect of the task. During discussion of the task it was clear that some participants did not understand the task (for example ranking informal care provision as well as NHS delivered care). Participants' prioritisation often did not reflect the attributes generated from the more open deductive questions. For example, one participant wrote down and verbalised one attribute she valued, but later apologised saying she did not know what to put. This participant, and others, had strong preferences, the strength of which were not reflected in this exercise. Additionally, more confident participants (and their respective rankings) took dominance in the group prioritisation exercise. Omitting attributes valued by participants in their decision making for follow-up care violates a theoretical requirement of discrete choice experiments (Coast et al, 2012; Lancsar & Louviere, 2008). Results identified from this aspect of the nominal group task are considered to lack validity and reliability. As a meaningful consensus was not reached, results from the prioritisation task are not be presented.

4.5.7 Informal discussion with Health Care Professionals

This method was used to generate attributes of follow-up care that Health Care Professionals perceived to be of most benefit to patients. Due to the resources available this was an informal discussion. Health Care Professionals individually identified and prioritised attributes in order of value. Attributes identified and prioritisation of these differed between Health Care

Professionals. See Table 17 for attributes generated from informal discussion with Health Care Professionals. This informal discussion provided information on the attributes valued by Health Care Professional's, and by its nature generated attributes which are plausible in the clinical setting which is essential in discrete choice experiment design (Ryan, 1996).

4.6 Discussion

This chapter aimed to compare the attributes of gynaecology cancer follow-up generated from different methods, to inform the design of a discrete choice experiment. This chapter continues the methodological debate for attribute generation and the impact this has on the design of the discrete choice experiment, the results and ultimately policy decisions made from these, thus meeting research recommendations (Coast & Horrocks, 2007; Coast et al., 2012; Ryan et al., 2014).

Different methods identified both similar and different attributes, both within and between an individual stakeholders perspective. Overall, 22 attributes were identified. Two attributes (care stratified by risk of recurrence; time spent in consultation) were identified by one method; five attributes (clinic waiting time; patient involved in decision making: protocol based care; transparent protocol based care; peer support) were identified by two methods; one attribute (organisation of appointments) was identified by three methods; four attributes (attitude of Health Care Professional; information; continuity of care; duration (number of years have appointments for)) were identified by four methods; three attributes (tests; contact details for Health Care Professional if concerns; prompt re-access if required) were identified by five methods; six attributes (consultation itself; holistic patient centred care; reciprocal communication; medical examination; mode of care; frequency of appointments) were identified by six methods; and one attribute (expertise of Health Care Professional) was identified by seven methods. This demonstrates that:

- 1) The use of different methods may generate different attributes, and therefore discrete choice experiment design, findings and any policy making decisions based on these.
- 2) The choice of methods used should be evidenced based and guided by a clear rational within the available resources.

In this context, qualitative research and an informal discussion with Health Care Professional's was essential in identifying attributes valued by stakeholders. The use of the nominal group technique was not valuable as patients and their informal caregivers were unable to prioritise attributes as a group. Even if patients and their informal caregivers could have done this, different focus groups and interviews may have identified different attributes and with different rankings. The systematic literature review was of less benefit, as the individual studies identified generally did not use a bottom up approach, so the attributes identified and valued were generally those perceived by researchers and policy makers as important to explore. Exploration of current practice and policy recommendations were invaluable in identifying plausible attributes within the changing model of care from the perspective of the health care provider. Given that non-systematic literature reviews should not be used to inform decision making in health care, a literature review was, and should not be conducted to generate attributes.

4.6.1 Existing research

The findings of this study support findings from a previous methodological comparison of a four phase qualitative approach compared to a four phase reductionist approach. Similarly results identified that the conduct of different methods resulted in the identification of both similar and different attributes (Hetler & Boehler, 2016). However, Helter and Boehler (2016) compared a phased approach not single methods, whereby attributes generated from qualitative research were compared to attributes generated from a narrative literature review (which should not to be used to inform health care decision making: Gough et al., 2012; Munn et al., 2018; Robinson & Lowe, 2015), before subsequent methods were employed for subsequent phases. Therefore this study provides further knowledge of the potential impact of different methods to generate attributes. This finding also confirms previously published recommendations that the choice of methods used to inform attribute development, should be informed by a clear rationale with consideration to the skills available within the research team and resource availability (Coast et al., 2012).

In this clinical context, the use of qualitative research was invaluable in ensuring that attributes identified were valued by stakeholders, echoing previous research recommendations (Coast et al., 2012; Louviere et al, 2000). Exploration of current practice

and policy recommendations was also invaluable in identifying plausible attributes valued by policy makers. The use of literature reviews to identify attributes for a discrete choice experiment is common (De Brún et al., 2018; Hiligsmann et al., 2013; Howard et al., 2015; Kunaviktikul et al., 2014; Robinson, et al., 2015), but as illustrated is a source of bias. The use of systematic literature reviews to generate attributes for a discrete choice experiments is also endorsed (Lynn et al., 2015), but in this context, because of the methodology employed within empirical studies identified, was not as valuable as the qualitative study with patients and their informal caregivers, and the informal discussion with Health Care Professionals, in identifying attributes valued by stakeholders. The use of the nominal group technique was not of value in this context, despite being used in other contexts (Hiligsman et al., 2013; Powell et al., 2015).

4.6.2 Strengths and limitations

To our knowledge, this is the first comparison of attributes identified from the conduct of multiple independent methods and from the perspective of different stakeholders. As such, this methodological study provides valuable insight and discussion regarding the importance of the choice of methods used for the generation of attributes to be included in a subsequent discrete choice experiment.

The following limitations must be acknowledged. It is acknowledged that the conduct of this comparison in a different clinical field, and using different methods, by researchers with different methodological and disciplinary perspectives may have yielded different results. Given the timing of completion of the systematic literature review, the findings from studies identified in the descriptive map were used in this mixed method comparison, as opposed to the synthesis. However, the synthesis identified similar attributes to the descriptive map, but due to the nature of the synthesis this was more refined. The informal discussion with Health Care Professional's, by its nature, was not an independent research study but a brief discussion. An advantage to this method of attribute generation was that it was brief and did not require ethics approval. However, the data was not rich and as was not as meaningful as the data generated from the qualitative study with patients and their informal caregivers. It is acknowledged that if the same qualitative methods were used to generate attributes from Health Care Professional's as they were for patients and their informal caregivers, it may have

generated different attributes of care. However, this was not possible within the resources of this PhD funded study.

4.6.3 Future research

This methodological comparison was conducted in the field of gynaecology cancer follow-up care. Further research is needed to explore whether similar results are identified in different clinical fields.

4.7 Conclusion

This chapter aimed to compare the attributes generated from recommended and frequently endorsed methods of attribute generation, in the clinical context of gynaecology cancer follow-up. This chapter identified that the independent conduct of different methods to inform discrete choice experiment design, and from the perspective of different stakeholders, identified both similar and different attributes. Therefore the attributes in a DCE, are a result of the methods used to identify them. This highlights the importance of formative work in DCE design, questions the validity of DCE's, their findings and any policy decisions made based on these. Because of these findings it is recommended that the choice of methods used should be evidenced based and guided by a clear rationale within the available resources.

4.8 Novel contributions

4.8.1 Methodological contributions

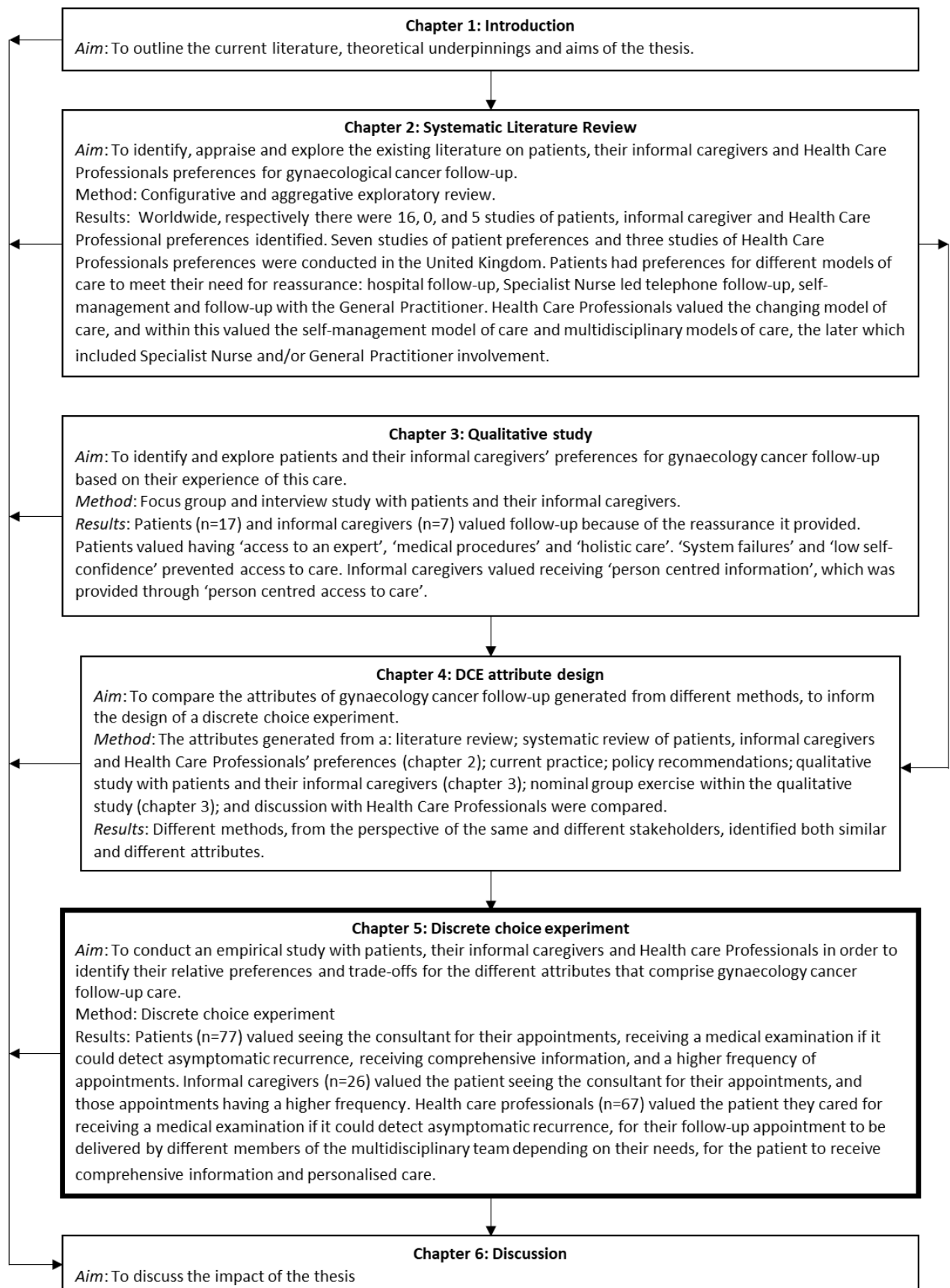
- There has been no previous methodological comparisons of attributes generated from different independent methods, and from the perspective of different stakeholders. This chapter identified that different methods, conducted from the perspective of similar and different stakeholders, generated both similar and different attributes.
- This chapter demonstrates the importance of a clear rational and transparent methodology in the choice of methods to inform discrete choice experiment design.
- This chapter highlights the importance of further methodological comparisons in different clinical populations to continue this methodological debate.

4.8.2 Policy contributions

- Discrete choice experiments are conducted with the aim of understanding relevant stakeholders relative preferences for a good or service, and the trade-offs that stakeholders are prepared to make for a one unit increase in care provision (for example, willing to trade 3 months between appointments to receive personalised over generalised care).
- Different methods are employed by researchers to generate attributes for a discrete choice experiment. This chapter identified that the independent conduct of different methods identified similar and different attributes, both within and between the perspectives of different stakeholders. Different attributes by their nature would produce different results in the discrete choice experiment. This raises huge questions for health care decision making based on findings from a discrete choice experiment.

Chapter 5:

**Patients, their informal caregivers’
and Health Care Professionals’
preferences for gynaecology cancer
follow-up care: a discrete choice
experiment**



5.1 Chapter Summary

In Chapter 1 (introduction) and Chapter 2 (systematic literature review) a need to identify patients, their informal caregivers and Health Care Professionals' preferences for care emerged, with the overarching aim of improving the efficiency of the provision of gynaecological cancer follow-up care. Chapter 3 (qualitative study), identified attributes of care valued by patients and their informal caregiver's. The attributes identified from the qualitative study (Chapter 3), a nominal group exercise within this, along with the attributes identified from a literature review, the systematic literature review (Chapter 2), an informal discussion with Health Care Professional's, current practice and policy documents were discussed, compared and contrasted (Chapter 4). This methodological comparison identified that the method used to generate attributes had an effect on the attributes generated, highlighting a need for the choice of methods used to be evidence based, be informed by a clear rational and within the resources available (Chapter 4). This chapter describes the development and conduct of a discrete choice experiment, designed from the aforementioned methodological comparison, to identify patients (n=77), their informal caregiver's (n=26) and health care professionals (n=67) preferences for gynaecological cancer follow-up. The discrete choice experiment identified that when follow-up was described by the following attributes: 'personalised care'; 'information received'; 'medical examination'; 'Health Care Professional seen'; and 'frequency of appointments', patients valued in order of importance, 'Health Care Professional seen' ($p = 0.001$), the 'frequency of appointments' ($p = 0.000$), 'medical examination' ($p = 0.000$), and 'information received' ($p = 0.001$). Patients were willing to trade 17.4 months to see a Consultant over their General Practitioner, 6.8 months between appointments to receive a medical examination that can as opposed to cannot detect asymptomatic recurrence, and 4.3 months to receive comprehensive over basic information. Whereas informal caregiver's valued, in order of importance, 'frequency of appointments' ($p = 0.000$) and 'Health Care Professional' seen ($p = 0.000$). Informal caregivers were prepared to wait a further 12.7 months for the patient they provided care for to be seen by a Consultant over General Practitioner. Dissimilarly, Health Care Professionals valued, in order of importance, 'medical examination' ($p = 0.000$), 'Health Care Professional seen' ($p = 0.000$), 'information received' ($p = 0.000$), and 'personalised care' ($p = 0.000$). Nevertheless, the findings from this study provide decision makers and health commissioners a meaningful and important insight into preferences of these three stakeholder groups. Together with the

findings from the systematic literature review (Chapter 2) and qualitative research study (Chapter 3), the results from this study can be used to help improve the efficiency of gynaecological cancer follow-up, with the need for a prudent changing model of care (see Chapter 1 and Chapter 6 for discussion).

5.2 Introduction

There is a lack of evidence for the efficacy of the traditional model of gynaecology cancer follow-up care delivered within the NHS, in detecting recurrence and increasing survival (Clarke et al., 2014; Elit et al., 2009; Fung-Kee-Fung et al 2006; Guerts et al., 2012; Jefford et al., 2013; Jeppesen, 2017; Lanceley et al 2013; Leeson et al., 2017; Leeson et al., 2013; Lim et al., 2004; Nama et al., 2013; Rustin et al, 2010; Vistad et al, 2017). Additionally, patients report unmet needs within the follow-up period and have asked for alternative models of care to meet their need for medical reassurance post-treatment (Bradley et al, 1999; Hodgkinson et al, 2007; Lydon et al., 2009; Miller et al, 2003; Morrison et al., 2012; Oshima et al, 2011; Sekse et al, 2010). Given the above there is a need for a changing model of follow-up to better meet patients' needs (Leeson et al., 2017; National Cancer Survivorship Initiative, 2013; Welsh Government & Welsh NHS, 2016).

There is an increased focus on the provision of patient centred services, designed based on patients' preferences for care (Coalition for Collaborative Care, 2014; Mulley et al., 2012; The Kings Fund, 2010; Wales Cancer Network, 2016). Given the role and impact of informal caregiving on the informal caregiver as well as the patient, there is also a need to understand the preferences of informal caregivers from a policy maker's perspective (Brodaty & Donkin, 2009; Department of Health, 2012; Romito et al, 2013; Stafford & Judd, 2010). Given the role influence and effect of Health Care Professional's preferences on the model of care delivered and consumed, there is also a need to understand Health Care Professional's preferences for care (Buchannan, 1988; Stacey et al., 2017; van Hezewijk et al., 2011).

To date, no discrete choice experiments have been conducted to explore preferences for gynaecology cancer follow-up, despite the information they provide on relative preferences and trade-off's for care, and despite the importance of understanding preferences from each of these stakeholders perspectives in informing health care decision making within the ever finite resources of the NHS (Ryan, 2004). Therefore, this chapter aimed to conduct an empirical study with patients, their informal caregivers and Health care Professionals in order to identify their relative preferences and trade-offs for the different attributes that comprise gynaecology cancer follow-up care. Specifically, the objectives were to:

- 1) Identify patients, their informal caregiver's and Health Care Professionals relative preferences for the different attributes of gynaecological cancer follow-up care and the levels in which they were defined
- 2) Identify the trade-off's that patients, their informal caregiver's and Health Care Professionals are prepared to make for their desired levels of care
- 3) Compare and contrast the preferences of patients, their informal caregivers and Health Care Professionals.

5.3 Method

5.3.1 Design

The study was approved by the School of Psychology, Bangor University and NHS ethics and R&D committees. Recruitment documents and the questionnaire booklet were piloted with patient and informal caregiver representatives and in a demographically mixed, opportunistically recruited sample of the researcher's family and friends to ensure readability and ease of understanding (INVOLVE, 2012). The study met the eligibility criteria for adoption onto the NISCHR CRC portfolio (portfolio number 20257).

5.3.2 Participants

Gynaecological cancer follow-up patients (and their respective informal caregivers), were eligible to take part in the study if: they had been treated with curative intent and were receiving follow-up at the recruiting hospitals; had, had at least one follow-up appointment; had no sign of active disease at their last follow-up appointment; were over 18 years of age; could speak English or Welsh; and had the capacity to consent. Patients were not excluded if their informal caregiver did not consent.

Health Care Professionals who worked with gynaecological cancer patients and/or their informal caregiver's, or who had an interest in gynaecological cancer were eligible to participate.

5.3.3 *Sample size*

Sample size calculations are a growing field (de Bekker-Grob, Donkers, Jonker & Stolk., 2015). The study identified a minimum sample size of 30, given the guidance for regression analysis outlined by Ryan et al (2008).

5.3.4 *Recruitment*

Two hundred and twenty three patients were screened for eligibility between December 2015 and April 2016 and 114 were deemed eligible to take part. Eighty seven patient consent forms were returned to the researcher (recruitment rate 76%). Ten questionnaires were incomplete, and 77 complete.

Informal caregivers were opportunistically recruited by the patient they provided care for, so the number of informal caregivers given the recruitment pack is unknown. Twenty seven informal caregiver consent forms were returned to the researcher. One was incomplete and 26 complete.

Health Care Professionals working in secondary care were invited to take part in the study at the North Wales Gynaecological Cancer Educational Evening in November 2015, and study day in March 2016 via an interactive presentation using Microsoft PowerPoint and participation handsets. Sixty nine Health Care Professionals attended the educational evening and 11 the study day session. Seventy five secondary Health Care Professionals consented to take part in the study (94% recruitment rate). Due to the recruitment method, the number of primary Health Care Professional's invited to take part in the study is unknown, as the number of practices the research study was sent to, and number of eligible professionals within each practise is unknown. Overall, 84 questionnaires were 'returned' by Health Care Professionals, 17 of these were incomplete and 67 were complete. Of these, 58 were completed by secondary Health Care Professional's and 9 were completed by primary Health Care Professional's.

In the population of patients receiving gynaecology cancer follow-up, 53% are diagnosed with endometrial cancer, 25% are diagnosed with ovarian cancer, 16% are diagnosed with cervical cancer and 6% are diagnosed with vulvar cancer (<https://www.cancerresearchuk.org/health-professional/cancer-statistics/statistics-by-cancer-type>). Comparatively, of the sample recruited to the study, 55% were diagnosed with

Table 20. Sample demographics compared to population demographics

Cancer type	Population incidence rate	Incidence rate x 5*	Population 5 year survival	Number of patients alive 5 years post diagnosis	Percentage of patients in follow-up	Sample recruited† n (%)
Endometrial	8,984 per year	44920	79%	35487 alive at 5 years post diagnosis	53%	42 (55%)
Ovarian	7,270 per year	36350	46.2%	16794 alive at 5 years post diagnosis	25%	24 (31%)
Cervical	3,126 per year	15630	67.4%	10535 alive 5 years post diagnosis	16%	9 (12%)
Vulvar	1,339 per year	6695	63.6%	4258 patients alive 5 years post diagnosis	6%	1 (1%)
Total	20719	103595		67074	100%	76 (99%)

*Incidence rate x 5 as patients are traditionally followed up for 5 years within the local health board

†n=1 missing data (total patient sample 77)

endometrial cancer, 31% were diagnosed with ovarian cancer and 1% were diagnosed with vulvar cancer (see Table 20). Therefore, compared to the population, the sample was under representative of cervical and vulvar cancer patients.

5.3.5 Materials

5.3.5.1 Patient and Informal caregiver materials

The recruitment booklet contained the research invite letter, information sheet, request for more information, consent form, questionnaire (see measures), debrief form, and two freepost envelopes. Within the patient recruitment pack was an envelope containing the informal caregiver recruitment pack. A Welsh language version of the recruitment booklet was available for participants. Materials and measures were printed on different coloured paper to reduce participant burden (Bray, 2015).

The research invite letter outlined the aim of the study, asked the potential participant to read the recruitment pack, provided contact details for the researcher and thanked them for taking the time to read this information.

The information sheet was a two page document that outlined the study aims; why the patient/informal caregiver was invited to participate; whether participation was required; what was required if they chose to participate; whether there was payment for participation; possible benefits and disadvantages to participation; confidentiality; the complaint procedure; ethical and R&D approval; who the study was organised and funded by; and what would happen to their questionnaire responses. Potential participants were informed that this research was being conducted as part of a PhD study.

The 'Request for more information' sheet asked participants to complete and return their contact details if they wanted the researcher to contact them with more information.

The consent form asked potential participants to sign that they had read and understood the participant information sheet and had, had the opportunity to have any questions answered; understood that participation was voluntary; agreed that participation and the data they provided was strictly confidential, however, if they disclosed harm to an individual that by law, this information had to be reported to the appropriate authority; consented for data to be stored securely in accordance with the Data Protection Act (1998); consented for their data to be looked at for the purpose of audit and monitoring by members of the auditing and monitoring team at Bangor University and the NHS Trust; that they

consented to taking part in the study; that they consented for their General Practitioner to be informed that they were taking part in the study; that they consented to Tenovus Cancer Care being given their contact details so that the charity could contact them to see if they could offer any support (if participants did not agree to this, they were still able to take part in the study: see ethical considerations). There were two copies of the consent form, one was kept by the participant and one was returned and then stored by the researcher.

The debrief form thanked participants for their participation and outlined: the aim of the study; that the data they provided would be analysed to understand what aspects of care they valued; to contact the researcher if they would like an overview of the results; that the results would be published and hopefully used to improve gynaecological cancer follow-up care; a prompt to contact their General Practitioner or Tenovus cancer care if following participation they would like to talk to someone about their cancer needs.

5.3.6 Materials for Health Care Professionals recruiting patients

The eligibility screening form was a tick box exercise for ease and to reduce error. Eligibility screening stickers contained the study title, name of principal investigator, ethics number and phrase 'screened for eligibility'. The 'screening, eligibility and recruitment pack record' contained clinic information (date of clinic; time of clinic; name of person who screened clinic), screening information (has the eligibility screening form been put in the respective patients' file Y/N; tally charts for the number of patients screened, number of patients eligible, number of patients not eligible with reasons, and number of patients given recruitment booklet to), as well as monitoring information (has a copy been given to the researcher, date, whom sent).

5.3.7 Materials for the recruitment of Secondary care Health Care Professionals

All materials and measures (see below), were contained within the 'Health Care Professional presentation'. Health Care Professionals provided their responses on handheld participation devices (see Figure 15 and description of handsets below). The information sheet outlined the aim of the study, what was required of participation, consent and confidentiality. Potential participants were asked to respond by pressing a button on their participation handset if they consented to taking part in the study, before the questionnaire began. The debrief form explained the aim of the study, how the participants' data would be used and contact details

for the chief investigator. A paper copy of the participant information sheet and debrief form was also given to all potential participants.

The use of a presentation and participation handsets provide an opportunity for participants to ask any questions whilst the questionnaire is being completed, and aids a timely response thus increasing recruitment rates (<http://www.sussex.ac.uk/tel/learningtechnologies/prs/tp>). The participation handsets are owned by Bangor University and supplied by Turning Technologies (<http://www.turningtechnologies.co.uk/>). Participants' responses were anonymous and were saved as a data set for later statistical analysis.

Figure 15: Participation handsets



5.3.8 Materials for the recruitment of Primary care Health Care Professionals

The expression of interest form was designed by NISCHR CRC Primary care workers, and contained a brief overview of the study. The participant information sheet had the PiCRIS (Primary care research incentive scheme) format, and outlined the study title; NIHR Portfolio reference number; type of study; study design; study aims and objectives; study target; eligibility criteria; health and care research wales workforce activities; practice activities; research team activities; and reimbursement of practice costs.

5.3.9 Materials for the Chief Investigator

A contact form detailing participant contact, and a case report form detailing all contacts was used as per good clinical practice (National Institute for Health Research, 2013).

5.3.10 Measures

5.3.10.1 Socio demographic questionnaire

Patients answered 24 questions regarding their demographics, cancer pathway and caregiving experience. For example, age, employment status and household income, cancer diagnosis, follow-up care received, and relationship to informal caregiver. Informal caregiver's answered 10 questions regarding their demographics and caregiving experience. For example, age, employment status, household income and how long had been providing care for. Health Care Professionals answered five questions regarding their demographics. For example, gender, professional role, and experience supporting gynaecological cancer follow-up care patients

5.3.10.2 Discrete choice experiment

As discussed in Chapter 4, discrete choice experiments aim to identify stakeholder's relative preferences for a hypothetical good or service. In accordance with the theoretical underpinnings of discrete choice experiments (consumer choice theory, Lancaster's theory of value and random utility theory: Lancaster, 1966; Manski, 1977; McFadden, 1974), it is assumed that individuals are rational decision makers who make stable decisions about which good or service they prefer, based on the perceived ability of the good or service to maximise their utility. In discrete choice experiments, a good or service is described by the attributes that comprise it, with a slight shift in attributes causing a shift in utility and hence preference for the model of care (Ryan, 1996). It has been recommended that discrete choice experiments are used to inform health care decision making (Ryan, 2004). There are five stages in the design and conduct of a discrete choice experiment, 1) attribute generation, 2) defining levels for the attributes, 3) identifying the choices to present, 4) collecting responses, and 5) analysing participants' responses (Ryan & Farrar, 2000).

The attributes generated from the systematic review (Chapter 2), qualitative study (Chapter 3), discussion with Health Care Professionals, current practice (Leeson et al., 2013) and policy guidance (National Cancer Survivorship Initiative, 2013), were discussed at depth with STY in relation to attribute and attribute level requirements (Bridges et al., 2011; Coast

et al., 2012; Flynn et al., 2008; Lanscar & Louviere, 2008; Ryan, 1996). It was recognised that it was important to not exclude any attributes important to decision making, but this was balanced by the need to adhere to the available resources (Bridges et al, 2011; Coast et al, 2012; Lanscar & Louviere, 2008). Through iterative discussion, the attributes generated from the aforementioned methods that met attribute criterion were able to be collapsed into five attributes with respective levels (Table 21). A research trial in which this thesis is connected to was also used to confirm the final choice of attributes (Morrison et al., 2018). In the qualitative work patients struggled with the concept of willingness to pay. Therefore the quantitative attribute was frequency of care (an attribute shown to be valued). These attributes were verified by the Principal Investigator (SL) as being plausible.

Table 21. Attribute development

Attributes generated from mixed methods	From mixed methods	Iterative discussion	Final choice of attributes
Consultation itself	X		Personalised care
Time spent in consultation	X		Personalised care
Organisation of appointments	X	They should be organised as standard. Not being organised is a system error	
Holistic patient centred care	X		Personalised care
Care stratified by risk of recurrence	X		Personalised care
Information	X		Information received
Reciprocal communication	X	Cannot be experimentally manipulated	
Medical Exam	X	Medical Exam	Medical examination
Tests	X	Not plausible in the UK	
Expertise of Health Care Professional	X		Expertise of Health Care Professional
Continuity of care	X	Not plausible	
Attitude of Health Care Professional's	X	Cannot be experimentally manipulated	
Protocol based care	X	Cannot be experimentally manipulated – this is standard	

Transparent protocol based care	X	Cannot be experimentally manipulated – it is the aim of future care to have clear pathways	
Mode (face-to-face location/telephone/self-management)	X		Personalised care
Patient involved in decision making	X		Personalised care
Peer support	X		Personalised care
Contact details for Health Care Professional if concerns	X		Information received
Prompt re-access if required	X	Cannot be experimentally manipulated – this will always be what happens	
Frequency of appointments	X		Frequency of care
Duration (number of years have appointments for)	X	Patients and informal caregivers felt this was for Health Care Professional's to decide	
Clinic waiting time	X	Not plausible – clinics do not aim to have a waiting time	

Attributes and levels were defined using the language used by patients in the qualitative research (Coast et al., 2012). In the supplementary guidance the definition for each attribute and level was explained, and participants were asked to assume that any aspect of follow-up care that was not described in the choices did not change (see appendices: Bridges et al., 2011). Piloting of the discrete choice experiment and supplementary guidance (as well as other study documentation), with patients and their informal caregivers, as well as in a sample of the researchers family and friends, took place to confirm the attributes, levels, explanations, wording of these and ease of understanding.

The above preliminary work resulted in the identification of five attributes in which four attributes had two levels ($\text{Levels}^{\text{Attributes}}: 2^4$), and one attribute had four levels ($\text{Levels}^{\text{Attributes}}: 4^1$). The combination of the number of attributes and levels (2^4 and 4^1) resulted in a full factorial design with 64 possible scenarios ($2^4 \times 4^1 = 64$). Table 22 below describes the attributes, attribute levels, definition of attribute levels, effect coding used and rationale for level definition in the study.

Table 22. Attributes, attribute levels, definitions and effect coding used in the study

Attributes	Attribute levels	Attribute levels definition (effect coding)	Rational for level definition and coding
1.Personalised care	1. Generic care 2. Person centred care	I have the same medical care as everyone else who has had gynaecological cancer (base) The care I receive is specific to my physical and mental needs	There is a policy focus on a more personalised model of care, stratified by patients' needs. The qualitative study identified a strong theme for holistic personalised care, and Health Care Professional's valued this attribute for patients. Generalised care is the current standard care, so was defined as the base.
2.Information received	1. Basic information 2. Comprehensive information	I receive basic information (base) I receive comprehensive information	There is an increasing policy focus on increased information provision so that care recipients are more informed. The qualitative study identified that patients and informal caregivers valued different amounts and transparency of information. Within BCUHB, basic information is currently delivered as standard, so was defined as the base.
3.Medical examination	1. Cannot detect asymptomatic recurrence 2. Can detect asymptomatic recurrence	I have an internal examination even if it can't detect whether the cancer has come back. I don't have any symptoms (base) I have an internal examination if it can detect whether the cancer has come back. I don't have any symptoms.	Medical examinations are given at all follow-up appointments despite the lack of evidence for their efficacy. The qualitative study identified that patients and informal caregivers place a lot of value on the medical examination because of its assumed importance in detecting recurrence. Informal discussion with Health Care Professional's highlighted the importance of a medical examination if there was evidence of efficacy to support this, whilst another Health Care Professional valued this for patients because of the amount of reassurance it provided, albeit false reassurance. Medical examinations are administered as standard despite a lack of evidence to their efficacy, so this was defined as the base.

4. Health Care Professional seen	1. GP 2. Specialist Nurse 3. Consultant 4. Multidisciplinary team	I see the GP for my appointments (base) I see the Specialist Nurse for my appointments I see the Consultant for my appointments I see different people in the multidisciplinary team for my appointments depending on my needs	This is a key theme within the changing model of follow-up as identified from policy documents, with patients and informal caregivers valuing care provided from Health Care Professionals with differing levels of expertise. Some Health Care Professional's thought that this attribute was important for patients care. The qualitative study identified that the General Practitioner would be the least preferred option, so this was defined as the base.
5.Frequency of appointments	1. Every 6 months 2. Every 12 months	I have an appointment every 6 months (6) I have an appointment every 12 months (12)	The qualitative study identified that patients and informal caregivers valued the frequency of appointments received. Patients and informal caregivers felt duration of care provision should be decided by Health Care Professional. This was the quantitative attribute, enabling trade-offs to be explored. The levels 6 months and 12 months were chosen as the same interval between levels is required. Because this attribute is the quantitative attribute, it does not have a base level.

To reduce participants' burden, a fractional factorial design (FrFD) was used to create a manageable number of choice sets (Hahn & Shapiro, 1966). The minimum number of choice sets (questions) for the combination of attributes and respective levels (Levels^{Attributes}: 2⁴ and 4¹) was eight. Using this catalogue (Hahn & Shapiro, 1966) ensured that there was orthogonal (that the occurrence of one attribute level was not dependant on the occurrence of another attribute level), and level balance (that each attribute level was presented the same number of times). This resulted in a discrete choice experiment that contained eight generic unlabelled pairwise choices (described as 'Follow-up Care A' vs 'Follow-up Care B'). It was important not to include a choice for the status quo or an opt out option, as 1) the aim was to explore preferences for the changing model of care, 2) research has shown that patients have a preference for the model of care delivered, and 3) the formative work and personal communication with treating clinicians identified that patients value follow-up and so attend their follow-up appointments (see Chapter 3; Salkeld, Ryan & Short, 2000; Personal communication, 2015). The pairwise choices were unlabelled, as the attributes comprising it defined the name of the model of care. There were no implausible attribute combinations or overlapping attributes. Participants were asked to choose which hypothetical model of care they preferred, 'Follow-up Care A' or 'Follow-up Care B' (a pairwise choice discrete choice experiment), described by the attributes and respective levels that comprised it. An example of a pairwise choice is illustrated in Figure 16.

The wording of the discrete choice experiment was amended to suit the perspective of different populations. For example, "I have the same medical care as everyone else who has had gynaecological cancer" was changed to "I would like the person I care for to have the same medical care as everyone else who has had gynaecological cancer" for informal caregivers, and to "Patients have the same medical care as everyone else who has had gynaecological cancer" for Health Care Professionals. Informal caregivers and Health Care Professionals were asked to make their valuations based on **what they perceived to be of most benefit to the patient(s) they cared for**.

To ensure that any attributes that effected decision making were not excluded, participants were informed that "Any other aspects of follow-up care that you think are important and are not listed, do not change. For example, duration of appointments" (Bridges et al, 2011).

Figure 16: Example of a pairwise choice task

Follow up A	Follow up B
I have the same medical care as everyone else who has had gynaecological cancer	The care I receive is specific to my physical and mental needs
I receive basic information	I receive comprehensive information
I have an internal examination even if it can't detect whether the cancer has come back. I don't have any symptoms	I have an internal examination if it can detect whether the cancer has come back. I don't have any symptoms
I see the GP for my appointments	I see the specialist nurse for my appointments
I have an appointment every 12 months	I have an appointment every 6 months

Tick here if you prefer Follow up A <input style="width: 40px; height: 20px;" type="checkbox"/>	OR	Tick here if you prefer Follow up B <input style="width: 40px; height: 20px;" type="checkbox"/>
---	----	---

5.3.11 Procedure

5.3.11.1 Procedure for patients and their informal caregivers

Health Care Professionals (Specialist Nurses/Research Nurses/Research Officers) screened patients attending relevant gynaecology and oncology outpatient clinics against the eligibility criteria, then confirmed the eligibility of the patient with the treating clinician. If eligible, the Health Care Professional gave the patient the recruitment pack. Where possible, eligible participants completed the recruitment pack in the waiting room before or after their appointment, with the Research Nurse/Research Officer nearby to assist in any clarification. When this was not possible, the recruitment pack was taken home with the potential participant for completion and return of documentation in their own time.

If the patient/informal caregiver returned the request for more information, the researcher contacted them to explain the research study further and to answer any questions, providing more detailed instructions to aid completion of the questionnaire if required. If the patient/informal caregiver expressed an interest in the research study but did not return a

consent form and questionnaire booklet, the researcher contacted the potential participant to enquire whether they had any questions or needed any assistance in completing the discrete choice experiment.

If the participant provided the relevant consent, the participants General Practitioner was informed of their participation via letter. If the participant provided the relevant consent, Tenovus Cancer Care were given the participants' contact details so that they could offer any support required (see ethical considerations). All patients and informal caregivers providing consent were entered into a prize draw with the winner chosen at random, to receive one of two £50 or £25 vouchers (Giuffrida & Torgerson, 1997).

5.3.11.2 Procedure for secondary Health Care Professionals

At the North Wales Gynaecology Cancer Education Evening November 2015, and a BCUHB Gynaecology oncology study day in March 2016, the researcher presented the study to attendees. Attendees if they wished consented and participated during the presentation using Participation Handsets.

5.3.11.3 Procedure for primary care Health Care Professionals

NISCHR CRC Primary Care workers sent an expression of interest form and information sheet to PiCRIS practices within BCUHB. A few weeks later NISCHR CRC Primary Care workers sent these primary care practices a questionnaire for completion (paper version of presentation). Recruitment reminders were sent as and when required. Primary care professionals returned the questionnaire directly to the researcher or indirectly via NISCHR CRC. Completion of the questionnaire counted towards the surgeries PiCRIS award.

5.3.12 Ethical considerations

Given the sensitive nature of the questionnaire and the emotions elicited in the qualitative research study (Chapter 3), consent was obtained to inform participants' General Practitioner and Tenovus Cancer Care of the participants' contact details, so support, if required, could be offered.

To reduce participant burden, the socio-demographic questionnaire was shortened from that described in Chapter 3 (see appendices). The number of potential attributes and attribute levels in the discrete choice experiment was reduced to five attributes (4x2, 1x4), to

limit the number of possible choice sets. It took the patient and informal caregiver representatives between 10 and 20 minutes to complete the recruitment booklet. The consent form, request for more information and questionnaire booklet were on different coloured paper and there were two separate booklets for participants, an English language booklet and a Welsh language booklet.

Non anonymised data was stored in a locked filing cabinet in a locked room, with the researcher being the only key holder. Anonymised data was stored on the researchers encrypted university laptop or in a locked filing cabinet, which the researcher was the only key holder for.

5.4 Data Analysis

Participants' demographic data were analysed to explore the characteristics of the sample. Data collected from the discrete choice experiment was inputted into Stata version 13 and analysed using random effects logistic regression techniques (Ryan, Bate, Eastmond & Ludbrook, 2001).

As discussed in Chapter 1, random utility theory underpins discrete choice experiments. In accordance with this theory, choice is indicative of preference, with individuals making a choice for the bundle of attributes that maximises their utility for health. Choice is a result of observable factors (the attribute level specification) and random components which cannot be observed (McFadden, 1986). To account for this, and because it allows for multiple responses from participants (participants responded to 8 pairwise choices), the discrete choice experiment was analysed using a random effects logit model (Bridges et al., 2011). The random effects logit model is described in Figure 17. The variables were effects coded. The attribute frequency of appointments (6 months/12 months)' was coded as a continuous variable, and the attributes 'personalised care (generic care/person centred care)'; 'information received (basic information/comprehensive information)'; 'medical examination (if it cannot detect asymptomatic recurrence/if it can detect asymptomatic recurrence)' and 'Health Care Professional seen (General Practitioner/Specialist Nurse/Consultant/different members of the multidisciplinary team depending on needs) as categorical variables. In accordance with the assumptions of discrete choice, and as illustrated in Figure 17, utility for a model of care is a sum of the utility of the attributes that comprise it.

Figure 17: Random effects logit model

$$\Delta \text{ Utility} = \alpha + \beta_1 \text{PerCare} + \beta_2 \text{Info} + \beta_3 \text{MedExam} + \beta_4 \text{HCP} + \beta_5 \text{FreApp} + \varepsilon_1 + \varepsilon_2$$

Where:

$\Delta \text{ Utility}$	= the change in utility in moving from scenario A to scenario B
α	= constant term
$\beta_1 - \beta_5$	= the beta coefficients of the model to be estimated
PerCare	= the difference in personalised care between follow-up A and follow-up B
Info	= the difference in information received between follow-up A and follow-up B
MedExam	= the difference in efficacy of medical examination between follow-up A and follow-up B
HCP	= the difference in Health Care Professional seen between follow-up A and follow-up B
FreApp	= the difference in frequency of appointments between follow-up A and follow-up B
ε_1	= the error term due to differences amongst observations
ε_2	= the error term due to differences amongst participants

Using this model, the results were analysed to identify, 1) which attributes significantly affected, and so were important to participants decision making for which bundle of attributes they preferred, 2) the relative preference of different attributes (the order in which they were valued), 3) the relative preference of individual attributes (the valuation of the levels within each attributes), 4) how participants were willing to trade between attributes, to have their desired preferences (marginal rate of substitution: MRS).

Within the regression analysis, the relative preference of individual attributes (the valuation of the levels within each attributes) were estimated by calculating the difference of the attribute's β -coefficient between the most preferred level of the attribute and the least preferred level of the attribute (Hauber et al., 2016). The attributes that significantly affected participants decision making for which bundle of attributes they preferred, was indicated by the p value for each respective attribute. The sign (+/-) of the β -coefficients is indicative of the direction of preference.

Marginal rates of substitution (MRS) demonstrate the amount of an attribute that participants are willing to trade (or give up) to obtain a one unit increase in another attribute.

The MRS is calculated by dividing the co-efficient of each statistically significant attribute, by the co-efficient for frequency of care (the attribute defined by a continuous quantitative scale).

Confidence intervals for the β -coefficients and MRS were estimated using non-parametric bootstrapping methods, by running a simulation of 1000 iterations in Stata version 13.0 (Hole, 2006; Yeo et al., 2012). Bootstrapping was chosen above other methods to estimate confidence intervals because it is the only method that is robust to ignoring unobserved heterogeneity when present (Hole, 2006).

When comparing the relevant preference of attributes, the co-efficient for 'frequency of care' was multiplied by 6 (months), a valid interval between appointments, to ensure a meaningful comparison.

5.4.1 Hypotheses

It was hypothesised that participants would prefer higher levels of personalised care (receiving person centred care over generic care), information received (comprehensive information over basic information), medical examination (a medical examination that can detect asymptomatic recurrence over a medical examination that cannot detect asymptomatic recurrence), and Health Care Professional seen (seeing members of the multidisciplinary team depending on needs, over the Consultant, over the Specialist Nurse, over the General Practitioner). It was hypothesised that patients would have a preference for a lower interval between scheduled appointments. These hypotheses were made based on the findings from the systematic literature review (Chapter 2) and qualitative research study (Chapter 3). This apriori hypothesis was compared with the results from the logic regression model, as a test for internal validity (Yeo et al., 2012).

5.5 Results

5.5.1 Patient Results

5.5.1.1 Demographic results (patient)

The mean age of patient participants was 65.8 years (SD = 12.3, range 27-95). Forty two (55%) patients had a diagnosis of endometrial cancer, 24 (31%) a diagnosis of ovarian cancer, 9 (12%) a diagnosis of cervical cancer, 1 (1%) a diagnosis of vulvar cancer, and 1 (1%) did not provide

a response. Seventy one (92%) patients were treated with surgery, 18 (23%) with radiotherapy, 11 (14%) with brachytherapy and 19 (25%) with chemotherapy. Eighteen (24%) patients were in their first year of follow-up care, 29 (38%) patients were 1-2 years post treatment, 11 (14%) patients 2-3 years post treatment, 6 (8%) patients 3-4 years post treatment and 10 (13%) patients 4-5 years post treatment (for these and other demographic results see Table 23). The model of follow-up care that patients receive varies by the Health Care Professional (Health Care Professional) that delivers the appointment; location of appointment; content of appointment; and frequency of appointment (Table 23).

Table 23. Patient (n=77) demographic data

Question	N (%)
Age Mean: 65.8 years (SD = 12.3) Range:27-95	
21-30 years old	1 (1)
31-40 years old	1 (1)
41-50 years old	7 (9)
51-60 years old	13 (17)
61-70 years old	25 (33)
71-80 years old	25 (33)
81-90 years old	4 (5)
91-100 years old	1 (1)
Employment Status*	
Employed for wages	17 (22)
Self employed	3 (4)
Retired	51 (66)
Homemaker	4 (5)
Unable to work	2 (3)
Full time carer	2 (3)
Income	
Nil or loss	1 (1)
£1-£79 a month/£3,999 a year	5 (6)
£80 - £149 a month/£4,000-£7,999 a year	16 (21)
£150 - £229 a month/£8,000 to £11,999 a year	13 (17)
£230 - £329 a month/£12,000 to £16,999 a year	16 (21)
£330 to £459 a month/£17,000 to £23,999 a year	10 (13)
£460 to £759 a month/£24,000 to £36,999 a year	4 (5)
£710 or more a month/£37,000 or more a year	5 (6)
£1 to £150	1 (1)
Missing data	6 (8)
Live with*	
Spouse	3 (4)

Family	10 (13)
Care home	1 (1)
Homeless	1 (1)
Alone	28 (36)
Husband	32 (42)
Son	1 (1)
Daughter	1 (1)
First diagnosis of gynaecological cancer	
Yes	76 (99)
No	1 (1)
Type of cancer diagnosed with	
Endometrial	42 (55)
Ovarian	24 (31)
Cervical	9 (12)
Vulvar	1 (1)
Missing data	1 (1)
Stage of cancer diagnosed with	
1	24 (31)
2	9 (12)
3	4 (5)
Not known	24 (31)
Incomplete or missing data	16 (21)
Has the cancer recurred	
No	76 (99)
Yes	1 (1)
How did you realise it had recurred?	No response
Treatment received*	
Surgery	71 (92)
Chemotherapy	19 (25)
Radiotherapy	18 (23)
Brachytherapy	11 (14)
Other	1 (1)
Duration in Follow-up care	
0 – 6 months	5 (7)
6 – 12 months	13 (17)
1 – 2 years	29 (38)
2 – 3 years	11 (14)
3 – 4 years	6 (8)
4 – 5 years	10 (13)
5+ years	3 (4)
Number of follow-up appointments received	
0 - 5	18 (23)
6 – 10	36 (47)
10+	4 (5)
Unknown	12 (16)
Missing data	7 (9)

Location of appointments*	
Hospital treated at	71 (92)
Local hospital	5 (7)
Telephone	8 (10)
Treated in different area of the country, have follow-up appointments at major hospital	2 (3)
Health Care Professional that delivers the appointments*	
Consultant Gynaecologist	52 (68)
Consultant Oncologist	29 (38)
Specialist Nurse	22 (29)
General Practitioner	3(4)
Frequency of Appointments	
3 monthly	37 (48)
4 monthly	5 (7)
6 monthly	22 (29)
12 monthly	11 (14)
Between 2 – 4 monthly	1 (1)
Other (frequency not given)	1 (1)
Content of Appointments*	
Internal examination	68 (88)
External examination	38 (49)
Medical Tests	4 (5)
Asked how I am feeling	69 (90)
Needs Assessment	5 (7)
Referred to services	5 (7)
Opportunity to ask questions	61 (79)
Relationship to informal caregiver	
Spouse	29 (38)
Parent	5 (7)
Sibling	7 (9)
Daughter	5 (7)
Other family member Daughter in law	3 (4)
Friend	7 (9)
No one was an informal caregiver to me	2 (3)
Missing data	19(25)
Live with informal caregiver	
Yes	30 (39)
No	24 (31)
Missing data	23 (30)
*n= >100 as multiple responses provided	

Table 24 shows the results from the logit regression model.

Table 24. Patient (n = 77) results from the logit regression model

Attribute	Attribute level	Patient study sample (n=77)				
		β -coefficient	95% CI†	P-value	MRS values‡ (months)	95% CI†
1. Personalised care	1.Care is generalised (omitted category)	0.0278	-0.0601 to 0.1294	0.534		
	2.Care is personalised	-0.0278	-0.1294 to 0.0601	0.534		
2.Information received	1.Information received is basic (omitted category)	-0.1440	-0.2566 to -0.0644	0.001*	4.2921	1.6394 to 8.2227
	2.Information received is comprehensive	0.1440	0.0644 to 0.2566	0.001*		
3.Medical examination	1.Internal examination if it <i>can't</i> detect whether the cancer has come back. No symptoms (omitted category)	-0.2292	-0.3671 to -0.1682	0.000*	6.8316	4.1001 to 12.2595
	2.Internal examination if it can detect whether the cancer has come back. No symptoms	0.2292	0.1682 to 0.3671	0.000*		
4. Health care professional seen	1.Health care professional seen for appointments: GP (Omitted category)	-0.5947	-0.9376 to -0.4602	0.000*	17.3979	10.6602 to 28.9233
	2.Health care professional seen for appointments: specialist nurse	0.1600	-0.0559 to 0.4130	0.121		
	3.Health care professional seen for appointments: consultant	0.5037	0.3821 to 0.7996	0.000*		
	4.Health care professional seen for appointments: different people in the multidisciplinary team depending on needs	-0.0690	-0.2889 to 0.1517	0.475		
5.Frequency of appointments	Frequency of appointments (months)	-0.0671	-0.1098 to -0.0448	0.000*		
	Constant	.0560	-0.1346 to 0.2707			
	Number of observations =	616				
	Number of individuals =	77				
	Wald chi2(7)	75.67				
	Log likelihood ratio	-378.3258				
	*p<0.05 †95% confidence intervals generated using non-parametric bootstrapping ‡MRS values = β -coefficient for attribute/ β -coefficient for frequency of appointment					

5.5.1.2 *Frequency of appointments*

To ensure a meaningful comparison of the relative preference for 'frequency of appointments', the β co-efficient was multiplied by 6 (months).

$$-0.0671 \times 6 = 0.4046$$

Logit regression analysis (patients)

Out of the five attributes, the β -coefficients from four attributes ('Health Care Professional', 'medical examination', 'information received', and 'frequency of appointments') were statistically significant ($p < 0.05$), demonstrating that these attributes were important to patients decision making for which bundle of attributes they preferred. When ranked in order of importance, the attribute Health Care Professional ($p = 0.001$) was valued most by patients followed by frequency of appointments ($p = 0.000$), medical examination ($p = 0.000$), and information received ($p = 0.001$: see Table 24).

The estimated β -coefficients and p values for 'medical examination', 'information received' and 'frequency of appointments' confirmed the apriori hypothesis. The coefficient for the attribute 'medical examination' was positive, indicating that patients had a preference for having a medical examination if it could detect asymptomatic recurrence, over not being able to detect asymptomatic recurrence. The positive sign on the β -coefficient for the attribute 'Information received' indicates that patients had a preference for comprehensive information over basic information. The negative sign on the β -coefficient for 'Frequency of appointments' indicate that patients had a preference for more frequent appointments. The β -coefficient for the attribute 'Health Care Professional' indicates that patients had stated a preference for seeing the Consultant over the General Practitioner for their follow-up appointments, but the p value for seeing the 'Specialist Nurse' and 'multidisciplinary team depending on needs' was not significant, indicating that seeing these professionals for their appointments was not important to patients' decision making for which bundle of attributes they preferred. The negative sign on the β -coefficient for the statistically insignificant attribute 'Personalised care' indicates that everything being equal, patients had a preference for generic over personalised care (see Table 25).

Results from the marginal rate of substitution demonstrate that patients were willing to wait:

- 1) An additional 4.3 months (CI 1.67 – 8.22) between follow-up appointments to receive comprehensive over basic information.
- 2) An additional 6.8 months (CI 4.10 – 12.26) between follow-up appointments to receive an internal examination if it can detect asymptomatic recurrence over if it cannot detect asymptomatic recurrence.
- 3) An additional 17.4 months (CI 10.66 – 28.92) between follow-up appointments to see the Consultant over the General Practitioner for their follow-up appointments.

Table 25: Attributes ranked in order of importance to patients (1=most important; 4=least important)

No.	Ranked attributes in order of importance
1.	Health Care Professional
2.	Frequency of appointments
3.	Medical examination
4.	Information received

5.5.2 Informal caregiver Results

5.5.2.1 Demographic Results (informal caregiver)

There were nine female informal caregivers and 17 male informal caregivers. The mean age of informal caregivers was 62.1 years (SD = 12.1), with an age range of 42-84 years. Sixteen (59%) informal caregivers were the patients' spouse, eight (27%) another family member and three (11%) a friend. Twenty two (81%) informal caregivers had provided care for the patients since the time of their diagnosis, three (11%) provided care when the patient was receiving their treatment, one (3%) during the recovery period post treatment (from straight after their treatment till 3 months post treatment), and one (3%) during the follow-up period only (for these and other demographic results see Table 26).

Table 26. Informal caregiver (n=26) demographic data

Question	N (%)
Gender	
Male	17 (65)
Female	9 (35)
Age	
Mean: 62.1 years (SD = 12.1)	
Range: 42-84 years	
40 – 50 years old	6 (23)
51 – 60 years old	5 (19)
61 – 70 years old	7 (27)
71 – 80 years old	6 (23)
80 – 90 years old	2 (8)
Employment Status*	
Employed	10 (38)
Retired	12 (46)
Out of work looking for work	1 (4)
Out of work not looking for work	1 (4)
Unable to work	1 (4)
Full time informal caregiver	1 (4)
Income	
Nil or loss	2 (8)
£1-£79 a month/£3,999 a year	1 (4)
£80 - £149 a month/£4,000-£7,999 a year	3 (11)
£150 - £229 a month/£8,000 to £11,999 a year	5 (19)
£230 - £329 a month/£12,000 to £16,999 a year	4 (15)
£330 to £459 a month/£17,000 to £23,999 a year	1 (4)
£460 to £759 a month/£24,000 to £36,999 a year	3 (11)
£710 or more a month/£37,000 or more a year	6 (23)
Missing	1 (4)
Relationship to Patient	
Spouse	16 (61)
Mum	3 (11)
Daughter	1 (4)
Mother in law	1 (4)
Daughter in law	1 (4)
Sister	2 (8)
Friend	2 (8)
Live with*	
Spouse	22 (85)
Family	6 (23)
Alone	1 (4)
No response	1 (4)
Duration of care	
Since the time of their diagnosis	21 (81)
When they had their treatment	3 (11)

In the recovery period after their treatment	1 (4)
During their follow-up period	1 (4)

Table 27 shows the results from the logit regression model

5.5.2.2 *Frequency of appointments*

To ensure a meaningful comparison of the relative preference for ‘frequency of appointments’, the β co-efficient was multiplied by 6 (months).

$$-0.1230 \times 6 = 0.738$$

5.5.2.3 *Logit Regression analysis (informal caregivers)*

Table 27 shows the results from the logit regression model. Out of the five attributes, the β -coefficients from two attributes were statistically significant ($p < 0.05$), and ranked in order of importance were: frequency of appointments’ ($p = 0.000$) and ‘Health Care Professional’ ($p = 0.000$: see table 28). This demonstrates that the attributes ‘frequency of appointments’ ‘Health Care Professional seen’ were important to informal caregivers decision making as to which bundle of attributes they preferred.

The estimated β -coefficients for frequency of appointments had the expected sign and significance, as stated a priori. The negative sign on the β -coefficient for the attribute ‘Frequency of appointments’, indicated that informal caregivers had preference for the patient to be given more frequent appointments. The positive β -coefficient for the attribute ‘Health Care Professional’ indicates that informal caregivers had a preference for the patient to see the Consultant for their follow-up appointments over the General Practitioner (The p value for seeing the ‘Specialist Nurse’ and ‘multidisciplinary team depending on needs’ was not significant, indicating that these attribute levels were not important to informal caregivers’ in their decision making for which bundle of attributes they preferred, thus not supporting the a priori hypothesis). The positive sign on the statistically insignificant attributes ‘personalised care’, ‘medical examination’ and ‘Information received’, indicates that, everything being equal, informal caregivers had a preference for the patient they provided

Table 27. Informal caregiver (n = 26) results from the logit regression model

Attribute		Informal caregiver study sample (n=26)				
		β -coefficient	95% CI†	P-value	MRS values‡ (months)	95% CI†
1. Personalised care	1. Care is generalised (omitted category)	-0.1194	-1.3989 to 0.0373	0.155		
	2. Care is personalised	0.1194	-0.0373 to 1.3989	0.155		
2. Information received	1. Information received is basic (omitted category)	-0.0104	-1.1562 to 1.1572	0.902		
	2. Information received is comprehensive	0.0104	-1.1572 to 1.1562	0.902		
3. Medical examination	1. Internal examination if it <i>can't</i> detect whether the cancer has come back. No symptoms (omitted category)	-0.1279	-1.3054 to 1.0174	0.127		
	2. Internal examination if it can detect whether the cancer has come back. No symptoms	0.1279	-1.0174 to 1.3054	0.127		
4. Health Care Professional seen	1. Health care professional seen for appointments: GP (Omitted category)	-0.9198	-4.7942 to -0.6027	0.000*	12.6772	7.3276 to 21.5946
	2. Health care professional seen for appointments: specialist nurse	0.1194	-1.1203 to 3.4517	0.524		
	3. Health care professional seen for appointments: consultant	0.6395	0.3143 to 2.1268	0.000*		
	4. Health care professional seen for appointments: different people in the multidisciplinary team depending on needs	0.1609	-0.9127 to 3.7344	0.392		
5. Frequency of appointments	Frequency of appointments	-0.1230	-0.5624 to -0.0835	0.000*		
	Constant	208	-0.731 to 0.5847			
	Number of observations =	26				
	Number of individuals =	34.82				
	Wald chi2(7)	208				
	Log likelihood ratio	-117.6268				
	*p<0.05 †95% confidence intervals generated using non parametric bootstrapping ‡MRS values = β -coefficient for attribute/ β -coefficient for frequency of appointment					

care for to receive personalised over generic care, a medical examination if it could detect asymptomatic recurrence compared to if it could not detect asymptomatic recurrence, and comprehensive over basic information (see Table 28).

As the β -coefficient for the attribute 'Frequency of appointments' was significant, MRS were calculated with this as the denominator. Calculation of the MRS demonstrates that informal caregivers were willing to trade an additional 12.7 months between follow-up appointments, for the patient they cared for to see the Consultant over the General Practitioner for their follow-up appointments.

Table 27. Attributes ranked in order of importance to informal caregivers (1=most important; 2=least important)

No.	Ranked attributes in order of importance
1	Frequency of appointments
2	Health Care Professional seen

5.5.3 Health Care Professional Results

5.5.3.1 Demographic results (Health Care Professional)

There were n=51 (76%) female Health Care Professional's and n=13 (19%) male Health Care Professional's. Three Health Care Professionals (4%) did not provide a response. Fifty eight (87%) Health Care Professional's worked in secondary care, and nine (13%) Health Care Professional's worked in primary care. Forty four (64%) Health Care Professional's worked with gynaecology cancer patients (for these and other demographic results see Table 29).

Table 30 shows the results from the logit regression model

5.5.3.2 Frequency of appointments

To ensure a meaningful comparison of the relative preference for 'frequency of appointments', the β co-efficient was multiplied by 6 (months).

$$-0.0322 \times 6 = 0.1932$$

Table 28. Health Care Professional (n=67) demographic data

Question	N (%)
Gender	
Male	13 (19)
Female	51 (76)
No response	3 (4)
Practitioner in:	
Secondary Care	58 (87)
Primary Care	9 (13)
Role in Secondary care:	
Consultant (A)	9 (13)
Specialist Trainee 1 – 6 (B)	11 (16)
Core Trainee 1 and 2 (C)	0 (0)
Foundation Year 1 and 2 (D)	0 (0)
Specialist Nurse (E)	3 (4)
Nurse (F)	11 (17)
Health Care Assistant (G)	1 (1)
Other (H)	23 (34)
Missing data	2 (3)
Role in Primary Care:	
General Practitioner (A)	7 (10)
Practice Nurse (B)	0 (0)
Advanced Nurse Practitioner (C)	0 (0)
Practice Manager (D)	0 (0)
Other (E)	0 (0)
Training	
Currently Training	15 (22)
Completed training 0-5 years ago	11 (16)
Completed training 5-10 years ago	7 (10)
Completed training 10-20 years ago	11 (16)
Completed training over 20 years ago	20 (30)
No response	3 (4)
Work with gynaecological cancer patients or their informal caregivers	
Yes	43 (64)
No	23 (34)
No response	1 (1)
Close family member or friend has been diagnosed with cancer	
Yes	48 (72)
No	18 (27)
No response	1 (1)

Table 29. Health Care Professional (n=67) results from the logit regression model

Attribute	Attribute level	Health Care Professional study sample (n=67)		
		β -coefficient	95% CI†	P-value
1.Personalised care	1.Care is generalised (omitted category)	-0.2668	-1.4407 to -0.1551	0.000
	2.Care is personalised	0.2668	0.1551 to 1.4407	0.000
2.Information received	1.Information received is basic (omitted category)	-0.3241	-1.5126 to -0.2285	0.000
	2.Information received is comprehensive	0.3241	0.2285 to 1.5126	0.000
3.Medical examination	1.Internal examination if it <i>can't</i> detect whether the cancer has come back. No symptoms (omitted category)	-0.5391	-1.7724 to -0.4648	0.000
	2.Internal examination if it can detect whether the cancer has come back. No symptoms	0.5391	0.4648 to 1.7724	0.000
4.Health care professional seen	1.Health care professional seen for appointments: GP (Omitted category)	-0.7466	-4.2569 to 0.4868	0.000
	2.Health care professional seen for appointments: specialist nurse	0.1720	-0.1454 to 3.6603	0.297
	3.Health care professional seen for appointments: consultant	0.0619	-0.1766 to 1.2445	0.583
	4.Health care professional seen for appointments: different people in the multidisciplinary team depending on needs	0.5127	-0.6147 to 0.8716	0.000
5.Frequency of appointments	Frequency of appointments	-0.0322	-0.4196 to .01502	0.141
	Constant	-0.1100	-0.3671 to 0.1470	
	Number of observations =	536		
	Number of individuals =	67		
	Wald chi2(7)	92.99		
	Log likelihood ratio	-283.8084		
*p<0.05				
†95% confidence intervals generated using non-parametric bootstrapping				

5.5.3.3 *Logit Regression Analysis (Health Care Professional)*

Out of the five attributes, the β -coefficients from four attributes were statistically significant ($p < 0.05$), demonstrating that these attributes were important to Health Care Professional's decision regarding which bundle of attributes they preferred. In order of importance Health Care Professional's valued 'medical examination' ($p = 0.000$), 'Health Care Professional seen' ($p = 0.000$), 'information received' ($p = 0.000$), and 'personalised care' ($p = 0.000$: as demonstrated in Table 30).

The estimated β -coefficients for the attributes 'medical examination', 'information received' and 'personalised care' all had the expected and significant positive sign as stated apriori. The β -coefficient for the attribute 'medical examination' was positive, indicating that Health Care Professional's had a preference for patients to be given an internal examination if could as opposed to could not detect asymptomatic recurrence. The positive sign on the β -coefficient for the attribute 'information received' indicates that Health Care Professional's had a preference for patients to receive comprehensive over basic information. The positive sign on the β -coefficient for 'personalised care' indicates that Health Care Professional's had a preference for patients to receive personalised over generic care. The positive sign of the β -coefficient for 'Health Care Professional' indicates that Health Care Professional's had a preference for the patient to see different Health Care Professionals within the multidisciplinary team depending on their needs, over the General Practitioner (The p value for seeing the 'Specialist Nurse' and 'Consultant' was not significant, indicating that these attribute levels were not important to Health Care Professional's decision making for which bundle of attributes they preferred). The negative sign on the statistically insignificant attribute 'Frequency of appointments' indicates that everything being equal, Health Care Professional's had a preference for patients to receive more frequent over less frequent appointments (see Table 31).

As the β -coefficient for the attribute 'Frequency of appointment' was insignificant ($p > 0.05$), MRS were not calculated.

Table 30. Attributes ranked in order of importance to Health Care Professional's (1=most important; 4=least important)

No.	Ranked attributes in order of importance
1.	Medical examination
2.	Health Care Professional seen

3.	Information received
4.	Personalised care

5.5.4 *Synthesis of Patient, Informal caregiver and Health Care Professional results*

Results showed that patients, their informal caregivers and Health Care Professional's placed differing values on attributes of follow-up care (see Table 32). The only attribute valued significantly by all three stakeholder groups (patients, their informal caregiver and Health Care Professionals) was 'Health Care Professional seen'. Patients and informal caregivers valued seeing the Consultant for appointments, whereas Health Care Professional's had a preference

for care to be delivered by different members of the multidisciplinary team depending on the patients' needs. Patients, their informal caregivers and Health Care Professional's had a preference for care not to be delivered by the General Practitioner. The attribute 'personalised care' was important to Health Care Professional's decision making for which bundle of attributes they preferred, with Health Care Professional's having a preference for personalised care and not generic care. However, this attribute was not important to patients and their informal caregivers decision making for which bundle of attributes they preferred. Conversely, the provision of information was important to patients and Health Care Professional's decision making for which bundle of attributes they preferred, with both stakeholders valuing the provision of comprehensive information and not valuing the provision of basic information. However, information received was not important to informal caregivers in their decision making for which bundle of attributes they preferred. Interestingly, the attribute 'medical examination' was important to patients and Health Care Professional's in their decision making for which bundle of attributes they preferred, but not informal caregivers. Both patients and their Health Care Professional's valued the provision of a medical examination if it could detect a recurrence in the absence of symptoms, but did not value the provision of a medical examination if it could not detect a recurrence in the absence of symptoms. The attribute 'frequency of appointments' was important to patients and their informal caregiver's, but not Health Care Professional's, in their decision making for which bundle of attributes they preferred. Both patients and their informal caregiver's valued more frequent appointments and did not value less frequent appointments.

The marginal rate of substitution was calculated when the attribute 'frequency of appointments' was significant, to identify the amount of time between appointments respondents would be willing to trade in order to have a one unit level improvement of care. Results showed that patients were willing to trade 17.4 months between appointments to see a Consultant over a General Practitioner for their appointments (other attribute levels were not significant), 6.8 months to receive a medical examination

if it could detect asymptomatic recurrence as opposed to could not, and 4.3 months to receive comprehensive over basic information. Similarly, informal caregiver's were willing to trade 12.7 months between appointments for the patient they cared for to see a Consultant over a General Practitioner for their appointments (other attribute levels were not significant). Health Care Professionals did not significantly value the attribute 'frequency of appointments', so calculating MRS using this insignificant attribute would have been meaningless.

Table 31. Synthesis of the attributes valued by patients, their informal caregiver's and Health Care Professional's

Ranked order*	Valued by Patients	Valued by informal caregiver's	Valued by Health Care Professionals
1	Health Care Professional seen	Health Care Professional seen	Medical examination
2	Medical examination	Frequency of appointments	Health Care Professional seen
3	Information received		Information received
4	Frequency of appointments		Personalised care

*Ranked in order of importance from 1 the most important to 4 the least important.

5.6 Discussion

To our knowledge this is the first discrete choice experiment conducted to elicit preferences for the model of gynaecology cancer follow-up care. As such, this study provides important information for decision makers as to the value of different attributes of care, within the levels in which they were defined. The aim of the study was to 1) identify patients, their informal caregiver's and Health Care Professionals relative preferences for the different attributes of gynaecological cancer follow-up care and the levels in which they were defined, 2) Identify the trade-off's that patients, their informal caregiver's and Health Care Professionals were prepared to make for their desired levels of care, and 3) compare and contrast the preferences of patients, their informal caregiver's and Health Care Professional's.

This study identified that in order of preference patients valued 'Health Care Professional seen'; 'Frequency of appointments', 'Medical examination'; and 'Information received'. However, surprisingly the attribute 'personalised care' did not significantly impact on patients' decision making for which bundle of attributes they preferred. Conversely, informal caregivers' valued 'frequency of appointments' and 'Health Care Professional'. However did not significantly value 'personalised care', 'information received' or 'medical examination' in their decision making for which bundle of attributes they preferred. Both

patients and their informal caregiver's valued care delivered by the 'Consultant' but did not value care delivered by the 'General Practitioner', whilst whether care was delivered by the 'Specialist Nurse' or 'different members of the multidisciplinary team depending on their needs', was not important to patients and informal caregivers in their decision making. Interestingly, patients were prepared to trade 17.4 months between appointments to see the 'Consultant' for their appointment over the 'General Practitioner', and informal caregiver's were prepared to trade 12.7 months between appointments for the patient to see the 'Consultant' for their appointments over them seeing the 'General Practitioner' for their appointments. The amount of time patients and informal caregivers were prepared to trade for this one unit increase in attribute level is an indication of the strength of their preference for care to be delivered by the 'Consultant' over the 'General Practitioner'. Interestingly, as indicated by the marginal rate of substitution, patients were prepared to trade less time between appointments to have a medical examination if it could as opposed to could not detect asymptomatic recurrence (6.8 months), and comprehensive over basic information (4.3 months). Conversely, Health Care Professional's significantly valued in order of importance, 'medical examination', 'Health Care Professional seen', 'information received', and 'personalised care' in their decision making for which bundle of attributes they preferred. Surprisingly, 'frequency of care' did not significantly impact on Health Care Professional's decision making for which bundle of attributes they preferred. Interestingly, Health Care Professional's valued care delivered by 'different members of the multidisciplinary team depending on needs', and did not value care delivered by the 'General Practitioner', whilst care delivered by the 'Consultant' and 'Specialist Nurse' was not important to Health Care Professional's decision making for which bundle of attributes they preferred (Objectives 1-3).

5.6.1 Comparison to previous research

In comparison to the findings from the systematic literature review (Chapter 2) and the qualitative study (Chapter 3), the findings from this study are particularly interesting. The qualitative study identified that patients and their informal caregivers had a strong preference for personalised care. However, in this study the attribute 'personalised care' was not important to patients and their informal caregivers' decision making for which bundle of attributes were preferred. In this study, the attribute personalised care was defined as: "This describes having your care personalised to your physical and mental needs, with referrals to other services to support you if you need this", with the possible choices "I have the same medical care as everyone else who has had gynaecological cancer" and "The care I receive is specific to my physical and mental needs". This explanation is transparent, and the wording used was based on that identified in the qualitative study. There are some differences in the demographics of patients recruited

between the qualitative study and discrete choice experiment. For example, regarding the content of care, in the discrete choice experiment 90% of participants reported that they were asked how they were feeling compared to 76% in the qualitative study. Discordant results between this study and the qualitative study were also found for other attributes. Within the attribute 'Health Care Professional seen', seeing different members of the multidisciplinary team depending on needs, and seeing the 'Specialist Nurse' was not important to patients and informal caregiver's decision making for which bundle of attributes they preferred. Whereas in the qualitative study, generally patients and their informal caregiver's valued care delivered in the multidisciplinary team and by the Specialist Nurse. The systematic literature review (Chapter 2) identified that care led by the Specialist Nurse and General Practitioner were valued. However, in both the qualitative study (Chapter 3) as a whole and this study, patients and their informal caregiver's did not value follow-up appointments delivered by the General Practitioner. Whilst in the qualitative study (Chapter 3), patients and informal caregiver's placed a strong value on the frequency of care, it was felt that the frequency of care should be decided by Health Care Professional's based on their needs, but in this study, patients and informal caregiver's used this attribute (as defined by 6 monthly or 12 monthly appointments) to make a decision about which bundle of attributes they preferred. It is not uncommon for the use of different methods to identify different results, and whilst the differential findings are considered a question of internal validity of the discrete choice experiment, in health care research generally, differential mixed method findings are considered a strength as provide different insight and so understanding of the phenomenon (Tariq & Woodman, 2010).

The finding that patients and their informal caregiver's valued care delivered by the Consultant and did not value care delivered by the General Practitioner, confirms existing patient research regarding the value of Consultant led care because of the reassurance that seeing the 'top dog' for their appointment provides, and the view that a General Practitioner, a generalist, would not be able to provide this reassurance (Bradley et al, 1999; Kew et al, 2009; Lydon et al., 2009). However, previous research has shown that when patients have experience of seeing the Specialist Nurse for their gynaecology cancer follow-up appointments, they have a preference for this (Cox et al, 2008). It must be acknowledged that only 22 (29%) patients had experience of care delivered by the Specialist Nurse in the current study, which may explain these different findings. Findings regarding the value of the medical examination to patients and their informal caregiver's also confirms previous research regarding the value placed upon medical reassurance through medical examinations and tests (Bradley et al., 1999; Kew et al., 2007/Kew et al., 2009; Lydon et al., 2009).

This study confirms previous research into Health Care Professional's preferences for care. The systematic literature review (Chapter 2) identified that Health Care Professionals valued both the self-

management model of care and multidisciplinary models which included Specialist Nurse and/or General Practitioner involvement, due to their perceived need for a changing model of care. Comparatively this discrete choice experiment identified that Specialist Nurse led care was not important to Health Care Professionals decision making for which bundle of attributes were preferred, but that care led by the General Practitioner was not valued. It must be emphasised that the findings from the systematic review were 'involvement from', not 'led by' the Specialist Nurse or General Practitioner, which may explain these differential findings. The finding that 'frequency of care' was not important to Health Care Professional's decision making for which bundle of attributes they preferred is interesting given previously published findings regarding, and the reality of, over-burdened clinics (Lydon et al., 2009; findings from the systematic literature review in Chapter 2).

The finding that patients and Health Care Professionals valued the provision of a medical examination if it could detect a recurrence in the absence of symptoms, but did not value the provision of a medical examination if it could not detect a recurrence in the absence of symptoms is interesting. Informal discussion with Health Care Professionals (Chapter 4) generated the wording of this attribute, and was agreed by the Principal Investigator (SL) who confirmed the final choice of attributes. However on reflection, in discussion with Health Care Professionals and methodologists, there is a slight misspecification of the wording of this attribute that was not detected a priori. Asymptomatic recurrence can be detected by internal examination, however, asymptomatic recurrence can also not be detected by internal examination. The cancer has to be large enough to feel or see, to be detected. When the cancer is large enough to feel or see it is likely to be symptomatic (Personal communication, 2019). Therefore, clinically, the wording of this attribute is not entirely representative of the efficacy of an internal examination when the patient is asymptomatic. Nevertheless, the findings in relation to this attribute are interesting. This attribute did not govern patients and Health Care Professionals' decision making (other attributes were also important to decision making); patients had a stronger preference to see the Consultant over the General Practitioner for their appointments, compared to an internal examination that could as opposed to could not detect asymptomatic recurrence; this attribute was not important to informal caregivers decision making for which model of care they preferred.

The finding that patients, informal caregivers and Health Care Professional's preferences for care are both concordant and discordant supports previous research (Harrison et al, 2017; Shin et al., 2013). This discordance could be attributed to the difference in decision making: patients and their informal caregivers made a decision based on their experiences, attitudes and beliefs regarding their own/the person they provide care for's, care. Whereas Health Care Professionals made a decision based on what they perceive to be best for patients generally, not Mrs Jones for example. Nevertheless, this is an

interesting finding, and adds to the complexity of decision making based on the findings from a discrete choice experiment conducted from both singular and multiple stakeholder's perspectives.

5.6.2 Strength and limitations

There are several strengths to this research study. Firstly, this is the first discrete choice experiment to explore preferences for gynaecology cancer follow-up care, and from the perspective of both of those delivering and consuming care. This is of particular importance given the cost of follow-up care to the NHS, the lack of efficacy for the current model of care, the focus on the 'changing model of follow-up', prudent health care and patient involvement in decision making, as well as patients and their informal caregiver's unmet needs (see Chapter 1). Limited research has explored the preferences of gynaecology cancer patients as a whole, with some research focusing on a sub group of patients (for example early stage endometrial cancer patients), despite the similarities in current protocols of care (see Chapter 2); no existing empirical research has explored informal caregiver's preferences (bar Chapter 3); and there is a dearth of research regarding Health Care Professional's preferences for care (see Chapter 2). Secondly, the choice of methods used to generate attributes were informed by a clear rationale to ensure that they met the requirements for attributes (Coast et al, 2012; Ryan, 1996). The choice of methods used, and the conduct of these methods to a high standard ensured the attributes presented in the discrete choice experiment were valid, a common pitfall in the design of discrete choice experiments (see Chapter 4 for further discussion). Thirdly, the method used to recruit secondary Health Care Professional's yielded a particularly high recruitment rate (94% of secondary Health Care Professional's recruited), with also a high level of recruitment of patients (76%; Treweek et al., 2013). The use of interactive technology at study days/evenings (for the recruitment of secondary Health Care Professional's), and the use of research nurses and research officers (for the recruitment of patients) was an asset to this research study.

Whilst a strength of this research project is the number of patients and secondary Health Care Professionals recruited within the resource timeframe, a weakness is the number of patients, Health Care Professionals and particularly informal caregiver's recruited to ensure statistical efficiency in the discrete choice experiment. A sample size that it is too small may lead to imprecise estimates of effects. The study identified a minimum sample size of 30, given the guidance for regression analysis outlined by Ryan et al (2008). However, in accordance with Orme's rule of thumb which considers the number of questions, attributes and levels in its sample size calculation, a sample size of 125 is recommended for this discrete choice experiment (Johnson & Orme, 2003; Orme, 1998). Therefore, while these results are of interest, they should be interpreted with caution.

$$N > 500c / (t \times a)$$

c=Largest number of levels for main effects

t=largest number of tasks

a=number of alternatives

$$N > 500 \times 4 / (8 \times 2)$$

$$N = 125$$

Based on the recruitment figures obtained in Chapter 3, it was predicted that the recruitment of informal caregivers would be 50% of that of patients recruited (or more given that there was less burden participating in this questionnaire study compared to a qualitative study), however, it was only 34%. The primary care research team within NISCHR CRC predicted that 50 primary Health Care Professionals would have participated. Given that the timing of recruitment was at the end of the PiCRIS year and the ease of completion, the NISCHR CRC research team were not sure why recruitment levels were so low. Study reminders were sent to PiCRIS practices. Secondly, all participants were recruited from follow-up clinics or study days conducted within BCUHB, affecting the generalisability of the findings. It must be acknowledged that previous research using the same population (gynaecology cancer patients and their informal caregiver's recruited within Betsi Cadwaladr University health board), has shown that patients and their informal caregiver's have misconceptions regarding the efficacy of gynaecology cancer follow-up in detecting disease recurrence in the absence of symptoms, and then improving outcomes (see Chapter 3). As such, different preferences and trade-offs may have been elicited if patients and their informal caregiver's were fully informed of the aims of gynaecology cancer follow-up and the efficacy of the current model of care in detecting recurrence. Due to ethical and resource constraints, it was not possible to inform patients and their informal caregiver's of the evidenced based efficacy of the current model of care delivered and consumed. As a measure of stated preferences, it is assumed that if respondents were presented with the same choices in a real life situation, they would make the same choices. However, in real life these choices are not present so external validity cannot be confirmed.

5.6.3 *Future research*

Given the above, future research should explore why all of the attributes generated and used to define follow-up were not important to decision making for which bundle of attributes were preferred. Given the preliminary research used to generate attributes, this was a surprising finding. Following the above, further research, using a population that are informed as to the efficacy of the current model of care,

should be conducted to further explore the preferences of patients, their informal caregiver's and Health Care Professional's given the importance of this for the prudent changing model of care.

5.7 Conclusion

This study aimed to identify patients, their informal caregivers and Health Care Professional's preferences for gynaecology cancer follow-up care. This study identified that stakeholders had both concordant and discordant preferences. Patients valued seeing the Consultant for their appointments, a higher frequency of appointments, receiving a medical examination if it could detect asymptomatic recurrence and receiving comprehensive information. Informal caregiver's valued the patient having a higher frequency of appointments, and the patient seeing the Consultant for their appointments. Health Care Professionals valued the patient they cared for receiving a medical examination if it could detect asymptomatic recurrence, for their follow-up appointment to be delivered by different members of the multi-disciplinary team depending on their needs, for the patient to receive comprehensive information and personalised care. Patients were willing to accept a longer duration between appointments for care to be delivered by a Consultant over General Practitioner, for a medical examination that can as opposed to cannot detect recurrence in the absence of symptoms, and for comprehensive over basic information. Informal caregivers were also willing to accept a longer duration between appointments for the patient they provided care for, to receive their appointments with a Consultant as opposed to their General Practitioner. Future research should explore why all the attributes generated from rigorous and multiple methods were not important to stakeholders decision making for which bundle of attributes were preferred, and then further explore preferences for gynaecological cancer follow-up using a sample that are informed about the limitations of the current model of care. This is of key importance given the strategic focus on prudent health care in Wales, and need for a changing model of care from a policy perspective.

5.8 Novel Contributions

5.8.1 Methodological contributions

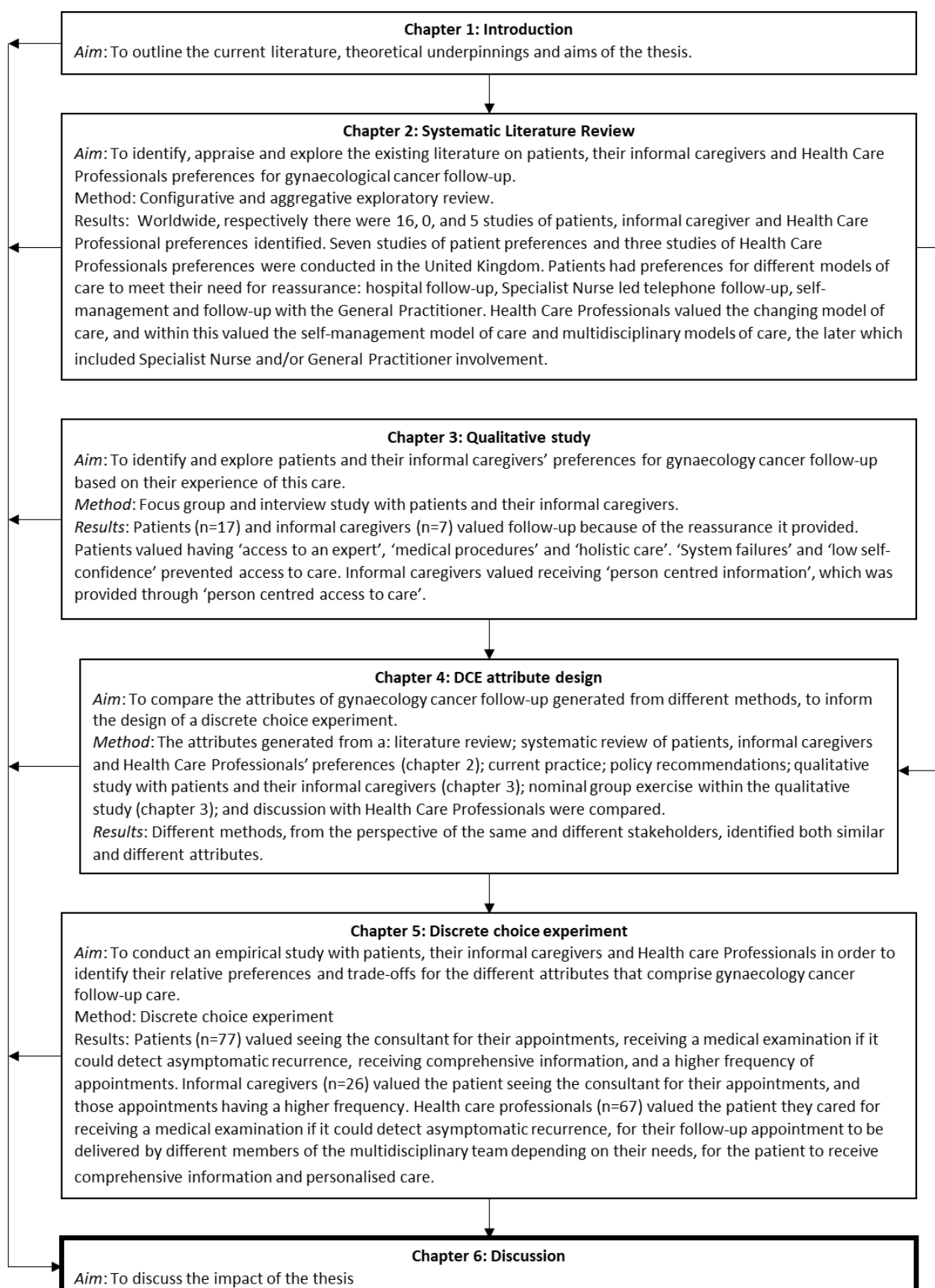
- This discrete choice experiment compared the relative preferences for gynaecology cancer follow-up, from the perspective of patients, their informal caregiver's and Health Care Professionals. The comparison of preferences from different stakeholders perspectives is novel but of high importance given the importance of 'preference misdiagnosis', patient involvement in decision making, impact of informal caregiving, and impact of Health Care Professional's preferences on the model of care delivered (Mulley et al., 2012; Revenson et al., 2016; de Bock et al., 2004; van Hezewijk et al., 2011).

5.8.2 Policy contributions

- No previous discrete choice experiments that have explored patients, their informal caregiver's and Health Care Professional's preferences for gynaecological cancer follow-up care (see Chapter 2).
- Discrete choice experiments can be used to aid health care decision making (Ryan, 2004). Exploring patients, their informal caregiver's and Health Care Professional's preferences for gynaecology cancer follow-up can aid health care decision making within the prudent changing model of follow-up (Department of Health, 2012; Mulley et al., 2012; Welsh Government & Wales NHS, 2016).
- It was of particular importance to conduct this research given the efficacy of the traditional model of follow-up from the patients and health care provider's perspective and the policy focus on the changing model of care (Leeson et al., 2017; National Cancer Survivorship Initiative, 2013).

Chapter 6:

Discussion



6.1 Main findings

There is a policy focus on a changing model of gynaecological cancer follow-up care given its limited efficacy and efficiency, from both a patient and health care provider perspective (Clarke et al., 2014; Elit et al., 2009; Fung-Kee-Fung et al 2006; Hodgkinson et al, 2007; Jefford et al., 2013; Jeppesen, 2017; Lanceley et al 2013; Leeson et al., 2017; Leeson et al., 2013; Miller et al, 2003; Morrison et al., 2012; Nama et al., 2013; National Cancer Survivorship Initiative, 2013; Rustin et al, 2010; Vistad et al, 2017). This is of particular importance in Wales, given the strategic focus on prudent health care (Welsh Government and NHS Wales, 2016). There is no NICE guidance or medical consensus regarding the model of care that should be delivered, resulting in a post code lottery of care provision (Leeson et al., 2013). However, a detailed protocol for the delivery of endometrial cancer follow-up was published by the British Gynaecological Cancer Society in 2017, advocating a stratified model of follow-up: Sundar et al., 2017).

Involvement of, and understanding the preferences of stakeholders is central to the development of evidence based guidelines for gynaecology cancer follow-up care. The first principle of prudent health care within the Welsh NHS is that the public, patients and Health Care Professionals are co-producers across service design, management and delivery, within the aim to achieve health and well-being (Welsh Government and NHS Wales, 2016). There is a focus on patient involvement in decision making regarding the care that should be delivered at both a commissioning and individual patient level, given the impact on patient outcomes and the efficiency of care delivery (Wales Cancer Network, 2016; Coalition for Collaborative Care, 2014; Mulley et al., 2012; The Kings Fund, 2010). This is of particular importance when resources are finite and needs infinite. Misdiagnosis of patients' preferences for care are prevalent, and has an impact on the outcomes and efficiency of care (Stacey et al., 2011; Harrison et al., 2017; Mulley et al., 2012). Given the role and impact of informal caregiving, there is an increasing awareness and focus on the need to understand informal caregivers' preferences for care (Department of Health, 2012). Even when there is guidance for the model of care delivered, the preferences of Health Care Professionals influence the model of care delivered (van Hezewijk et al., 2011). It was therefore of most importance to explore patients, their informal caregivers and Health Care Professional's preferences for care, at a time of increasing conversation for the changing model of follow-up care (National Survivorship Initiative, 2013; Personal communication, 2019), and strategy focus on a prudent health care within Wales (Welsh Government & NHS Wales, 2016).

The overarching aim of the thesis was to understand patients, their informal caregivers and Health Care Professional's preferences for gynaecology cancer follow-up care. Within this, the thesis aimed to:

- 1) To systematically identify, extract, appraise and explore the existing literature on patients, their informal caregiver's and Health Care Professionals preferences for gynaecological cancer follow-up care.
- 2) To identify and explore patients and their informal caregivers' preferences for gynaecology cancer follow-up, based on their experience of this care.
- 3) To compare the attributes of gynaecology cancer follow-up generated from different methods, to inform the design of a discrete choice experiment
- 4) To conduct an empirical study with patients, their informal caregivers and Health care Professionals in order to identify their relative preferences and trade-offs for the different attributes that comprise gynaecology cancer follow-up care

A systematic literature review (Chapter 2) was conducted to identify existing literature on gynaecological cancer patients, their informal caregivers and Health Care Professional's preferences for follow-up care. A qualitative study (Chapter 3) was conducted to identify and explore patients and their informal caregivers' preferences for gynaecology cancer follow-up, based on their experience of this care. Attributes generated from the systematic literature review and qualitative study were compared along with attributes generated from a literature review, policy guidance, current practice, informal discussion with Health Care Professionals and nominal group task, to see if the method used to identify attributes affected the attributes generated (Chapter 4). A discrete choice experiment was designed (Chapter 5) based on the findings from Chapter 4, to identify patients, their informal caregivers and Health Care Professional's relative preferences and trade-off's for the attributes that together defined gynaecology cancer follow-up care.

The findings from the thesis, and their corresponding chapters are summarised below:

6.1.1 Chapter 2

Aim: To systematically identify, extract, appraise and explore the existing literature on patients, their informal caregiver's and Health Care Professionals preferences for gynaecological cancer follow-up care.

Findings: A systematic literature review that explored 20 years of empirical research identified 19 research studies. Of these, 14 studies explored patient's preferences, no studies explored informal caregiver's preferences, three studies explored Health Care Professional's preferences, and two studies explored patients and Health Care Professionals preferences. Eight of these studies aimed to explore care delivered

by the National Health Service. Existing empirical evidence identified that patient's value follow-up for its assumed ability to detect recurrence and provide reassurance in the absence of disease. Because of this, some patients had a preference for the traditional model of follow-up, whilst other patients had a preference for Specialist Nurse led telephone follow-up, patient initiated care or follow-up delivered by the General Practitioner.

Health Care Professionals felt that the aim of care was to detect recurrence, and to monitor and treat any effects of treatment. Given the need for a changing model of care, Health Care Professional's valued self-management and multidisciplinary models of care, the latter including involvement from the Specialist Nurse and/or General Practitioner.

6.1.2 Chapter 3

Aim: To identify and explore patients and their informal caregivers' preferences for gynaecology cancer follow-up, based on their experience of this care.

Findings: A focus group and interview study conducted with 17 patients and 7 informal caregivers identified that preferences for care were governed by a need for reassurance. For patients, reassurance was gained through the provision of having access to an expert, medical procedures and holistic care. However, system failures and low self-confidence prevented access to reassurance. For informal caregivers, reassurance was met through the provision of person centred information, which was met through the delivery of person centred access to care.

6.1.3 Chapter 4

Aim: To compare the attributes of gynaecology cancer follow-up generated from different methods, to inform the design of a discrete choice experiment

Findings: Attributes generated from a literature review, systematic literature review (Chapter 2), current practice, policy guidance, informal discussion with Health Care Professional's, qualitative study (Chapter 3) and nominal group task (Chapter 3) were compared. Findings identified that the conduct of different methods, identified both similar and different attributes, both within and between individual stakeholder perspectives.

6.1.4 Chapter 5

Aim: To conduct an empirical study with patients, their informal caregivers and Health care Professionals in order to identify their relative preferences and trade-offs for the different attributes that comprise gynaecology cancer follow-up care

Findings: A stated preference discrete choice experiment was completed by 77 patients (recruitment rate: 76%), 26 informal caregivers (recruitment rate unknown) and 67 Health Care Professional's (recruitment rate of secondary Health Care Professional's: 94%, recruitment rate of primary Health Care Professionals is unknown). The relative preference for different attributes of gynaecology cancer follow-up care ('personalised care', 'information received', 'medical examination', 'Health Care Professional seen', and 'frequency of appointments') differed between stakeholders. Patients and informal caregivers valued seeing the consultant for appointments, whereas Health Care Professional's had a preference for care to be delivered by different members of the multidisciplinary team dependant on a patient's needs. Patients, their informal caregivers and Health Care Professional's had a preference for care not to be delivered by the General Practitioner. Health Care Professional's had a preference for personalised care and not generic care. Patients and Health Care Professional's valued the provision of comprehensive information and did not value the provision of basic information. Patients and Health Care Professional's valued the provision of a medical examination if it could detect asymptomatic recurrence, and did not value the provision of a medical examination if it could *not* detect asymptomatic recurrence. Patients and their informal caregivers valued more frequent appointments and did not value less frequent appointments. Patients were willing to trade 17.4 months between appointments to see a Consultant over a General Practitioner for their appointments, 6.8 months to receive a medical examination if it could detect asymptomatic recurrence as opposed to could not, and 4.3 months to receive comprehensive over basic information. Informal caregivers were willing to trade 12.7 months between appointments for the patient to see a Consultant over a General Practitioner for their appointments.

Together, the chapters in this thesis identified that patients, informal caregivers and Health Care Professional's valued gynaecology cancer follow-up. Patients and their informal caregivers valued follow-up because of the reassurance it provided in the absence of detection of recurrence. The specific attributes of care valued, to meet patients and their informal caregivers need for reassurance differed by the method used to identify preferences. Health Care Professionals felt the aim of follow-up was to detect recurrence, discuss concerns, monitor recovery and treat any symptoms and side effects of treatment. The attributes valued by patients, their informal caregivers and Health Care Professionals were discordant to varying degrees.

6.1.5 Patient preferences

The systematic literature review (Chapter 2) identified that patients have a preference for differing models of care because of the reassurance it provided: the traditional model of care, Specialist Nurse led

care, self-management or General Practitioner led care. The focus group and interview study (Chapter 3) also identified that patients had a preference for differing models of care, with some participants preferring the traditional model of care, whilst others preferring Specialist Nurse led care and others General Practitioner led care. However, the qualitative study (Chapter 3) identified that patients did not have a preference for the self-management or patient initiated model of care because of the assumed importance of follow-up and its ability to detect recurrence. These findings are in line with the health belief model (Rosenstock, 1974; Becker, 1974; Becker & Rosenstock, 1984) and theory of planned behaviour (Ajzen, 1985; Ajzen, 1991) that the perceived benefits (or outcome expectancies and the value of this outcome) of follow-up are strongly valued given the perceived susceptibility of a recurrence and perceived severity of the recurrence. Subsequently, participating in follow-up care is a coping response to the emotional and cognitive perceived threat of recurrence (the self-regulation model of illness: Leventhal, 1992).

The qualitative study (Chapter 3) further identified that patients had a preference for their individual needs to be met through having access to an expert, experiencing procedures to detect recurrence and the provision of patient led holistic care. Within this, patients valued the frequency in which they received follow-up appointments, generally having a preference for more frequent appointments at the onset of follow-up. A reducing frequency of appointments was perceived by patients as an indicator that they were 'getting better'. Therefore attending follow-up appointments could be considered as part of patients' coping response to the cognitive and emotional illness representation of the stressor (the perceived risk of recurrence: Leventhal et al., 1992). In accordance with this belief, patients felt that the frequency of appointments should be decided by the Health Care Professional, based on the care the Health Care Professional felt that the patient needed. In both support and contradiction of these findings, when the attributes, 'personalised care', 'medical examination', 'health care professional', 'information' and 'frequency of appointments' were used to describe the model of care, all attributes bar 'personalised care' influenced patients' decision making as to which bundle of attributes/model of care was preferred (Chapter 5).

In contradiction to the findings of the systematic literature review (Chapter 2) and qualitative study (Chapter 3), the discrete choice experiment (Chapter 5) identified that patients valued care delivered by the Consultant and did not value care delivered by the General Practitioner. Whether care was delivered by different members of the multidisciplinary team or Specialist Nurse was not important to patients' decision making for which bundle of attributes they preferred.

Additionally, the discrete choice experiment (Chapter 5) identified that the provision of personalised over generic care was not important to patients' decision making for which model of care

they preferred. However, the qualitative study (Chapter 3) identified that personalised care (centred on the needs of the patient) was highly valued by the majority of patients.

6.1.6 Informal caregiver preferences

The focus group and interview study (Chapter 3) identified that because of the reassurance follow-up appointments provided, informal caregivers valued the provision of person centred information (centred on the needs of the patient and the informal caregiver), met through the provision of person centred access to care (centred on the needs of the patient and the informal caregiver). Similarly to the findings regarding patients' preferences, these findings offer support to the health belief model (Rosenstock, 1974; Becker, 1974; Becker & Rosenstock, 1984), theory of planned behaviour (Ajzen, 1985; Ajzen, 1991) and the self-regulation model of illness (Leventhal et al., 1992), that follow-up care is strongly valued due to informal caregivers perceived susceptibility of recurrence and the perceived severity of this, with follow-up care being assumed to provide the information needed that there is no sign of recurrence (perceived benefit/outcome expectancies) which is highly valued. Attendance and engagement in follow-up care therefore acts as coping response to the illness representation of a perceived threat of recurrence.

Within the themes 'person centred information' and 'person centred access to care', attributes of care valued by informal caregivers included: the expertise of the Health Care Professional, continuity of care, the Health Care Professional's personality/attitude, medical investigations (examinations and tests), frequency of appointments and being given the time needed in these. Conversely, the discrete choice experiment (Chapter 5) identified that whilst 'information received', 'personalised care', 'medical examination', 'Health Care Professional', and 'frequency of appointments' were used to describe the model of care, only the attributes 'frequency of appointments' and 'Health Care Professional' influenced informal caregivers decision making for which model of care they preferred. Within this, informal caregivers valued care delivered by the Consultant and did not value care delivered by the General Practitioner, whilst care delivered by different members of the multidisciplinary team and care delivered by the Specialist Nurse did not significantly affect decision making. This differential finding could be explained in part by the finding from the qualitative study (Chapter 3), that informal caregivers valued continuity of care as well as the expertise of the Health Care Professional, with a value on consultant led care for scheduled appointments but strong value on unscheduled contact with the Specialist Nurse in between these appointments. However, does not explain the finding regarding information received, personalised care or medical examination. As described in Chapter 5, these insignificant findings could be explained by the small sample size.

6.1.7 Health care professionals' preferences

The systematic literature review (Chapter 2) identified that Health Care Professionals had a strong preference for a changing model of follow-up care. This belief was due to the lack of efficacy of the current model of care and the burden unnecessary appointments created on the clinic. Because of this some Health Care Professionals had a preference for self-management/patient-initiated care with information provision (that contained information about signs of recurrence and contact details if concerns), whilst others had a preference for multidisciplinary care that included medical procedures. Health care professionals had mixed preferences as to who should deliver follow-up care, with some health care professionals valuing Consultant led care, with others valuing involvement from the Specialist Nurse or General practitioner. Health care professionals also had mixed preferences regarding the frequency of appointments. Health care professionals delivering vulvar cancer follow-up generally had a preference for frequent appointments, whilst Health Care Professionals delivering ovarian cancer follow-up had a preference for less frequent care to reduce the burden on clinics. This could be a reflection on the differential rates of, and presentation of recurrence and the efficacy of treatment following identification of an ovarian and vulvar cancer recurrence (Rustin et al., 2010; Guerts et al., 2012; Leeson et al., 2017; Oonk et al., 2003). Nevertheless and interestingly, the discrete choice experiment (Chapter 5) identified that whilst 'information received', 'personalised care', 'medical examination', 'Health Care Professional', and 'frequency of appointments' (as detailed above) were used to describe the model of aftercare, the attributes: 'medical examination', 'Health Care Professional', 'information delivered' and 'personalised care' significantly affected Health Care Professionals' decision making regarding which bundle of attributes they thought was best for patients. However, the attribute 'frequency of appointments' did not. Interestingly, Health Care Professionals valued care delivered by different members of the multidisciplinary team but did not value care delivered by the General Practitioner, whilst care delivered by the Consultant and care delivered by the Specialist Nurse was not important to Health Care Professionals' decision making for which model of care they thought was best for patients.

6.1.8 Synthesis between patients, their informal caregivers and health care professionals' preferences

Patients, their informal caregivers and Health Care Professionals had both concordant and discordant beliefs regarding the aim of gynaecology cancer follow-up care, and preferences of care to meet these needs. Concordantly, and in line with previous research in cancer follow-up generally, the systematic literature review (Chapter 2) and qualitative study (Chapter 3) identified that patients and their informal caregivers valued follow-up care because of the reassurance it provided in the absence of detecting recurrence. As discussed in the introduction (Chapter 1), and as highlighted by one participant in the

qualitative study (Chapter 3), the reassurance that this provides is a false reassurance as disease recurrence for endometrial, ovarian and cervical cancer is generally symptomatic.

Concordantly, as identified in the discrete choice experiment (Chapter 5), patients, their informal caregivers and Health Care Professionals all valued the Health Care Professional that delivered follow-up appointments, in their decision making for which bundle of care they stated a preference for. Patients, their informal caregivers and Health Care Professionals did not value the idea of follow-up care appointments delivered by the General Practitioner. However, patients and their informal caregivers valued care delivered by the Consultant, whilst Health Care Professionals did not, only valuing care delivered by the multidisciplinary team. The discrete choice experiment (Chapter 5) identified that for patients the strength of preference for Consultant led care, was stronger than the preference for a medical examination if it could as opposed to could not detect asymptomatic recurrence. However, the systematic literature review (Chapter 2) and qualitative study (Chapter 3) identified that patients valued care delivered in the multidisciplinary team and care delivered by the Specialist Nurse. The qualitative study (Chapter 3) identified that informal caregivers valued scheduled care delivered by the Consultant, but not other professionals supporting this finding. The systematic literature review (Chapter 2) identified that Health Care Professionals valued care delivered in the multidisciplinary team, which included involvement from, not care led by, the Specialist Nurse and General Practitioner, thus in part supporting these findings.

Concordantly, as identified in the discrete choice experiment (Chapter 5), the provision of a medical examination was important to patients and Health Care Professionals, but not informal caregivers in their decision making for which model of care they preferred. Patients and Health Care Professionals valued the provision of a medical examination if it could detect asymptomatic recurrence, and did not value the provision of a medical examination if it could not detect asymptomatic recurrence. As identified in Chapter 2 (the systematic literature review) and Chapter 3 (the qualitative study), patients and their informal caregivers place a large value on the medical examination because of its assumed importance in detecting disease recurrence.

Concordantly, as identified in the discrete choice experiment (Chapter 5), patients and Health Care Professionals significantly valued information provision in their decision making for which bundle of attributes they preferred, valuing comprehensive over basic information. Discordantly, information provision was not important to informal caregivers in their decision making for which model of care they preferred. Although this differential finding is likely due to an insufficient sample size of informal caregivers (see Chapter 5). In the qualitative study (Chapter 3), information was not a key theme in the analysis of patient data, but was part of the theme 'access to care'. Conversely, information provision was a key attribute of care valued from the informal caregiver's perspective. The systematic literature review

(Chapter 2) identified that patients valued receiving information regarding symptoms of recurrence, and Health Care Professionals had a preference for patients to receive written and verbal information about signs of recurrence, and who to contact should they experience any of these. The definition used to describe basic information in the discrete choice experiment was:

“a one page sheet about the signs and symptoms of the cancer coming back and the contact details for your key worker/specialist nurse”

The definition used to describe comprehensive information in the discrete choice experiment was:

“Comprehensive information means a ten page booklet that also tells you about the physical and mental effects of the cancer for you and your informal caregiver, how you can make changes to your lifestyle to reduce the risk of the cancer coming back, and a list of local support groups and charities that can help you and your informal caregiver”.

Concordantly, as identified in the discrete choice experiment (Chapter 5), the frequency of care was important to patients and their informal caregiver’s decision making for which model of care they preferred, but discordantly, the frequency of care was not important to Health Care Professionals decision making for which model of care they thought was best for patients. Patients and their informal caregivers valued the provision of more frequent over less frequent appointments. Results from the qualitative study (Chapter 3) in part support this. The qualitative study identified that patients and their informal caregivers both valued the provision of frequent follow-up appointments, particularly towards the beginning of the follow-up period, but valued a reducing frequency of follow-up appointments because of the perception that reduced appointments mean that the patient is ‘getting better’. Interestingly whilst the qualitative study (Chapter 3) identified that patients and their informal caregivers valued more frequent appointments, patients and their informal caregivers felt that the frequency of follow-up appointments should be decided by the Health Care Professional, based on the Health Care Professionals perception of their need. However, Health Care Professionals did not value this attribute in their decision making for which model of care was best for patients. The systematic literature review synthesis (Chapter 2) did not identify any studies that explored desired frequency of follow-up provision, but one study identified that patients valued the current frequency of care, and one study identified that Health Care Professional’s valued regular follow-up for vulvar cancer patients. This discordant finding is particularly interesting as it highlights the problem of the agency relationship in health care, preference misdiagnosis, and patients and their informal caregiver’s misperceptions of the efficacy of follow-up care (Mulley et al., 2012). This discordant finding could be explained by health economic and health psychology theory. For Health Care

Professionals, this preference is likely to be a reflection of their clinical knowledge, that recurrence of gynaecological cancer is generally symptomatic, and that patients present with symptoms in between scheduled appointments. Therefore do not perceive this attribute as a benefit to increasing the utility gained from gynaecology cancer follow-up, in accordance with discrete choice theory (Lancaster, 1966; McFadden, 1974) and the health belief model (Rosenstock, 1974; Becker, 1974; Becker & Rosenstock, 1984). For patients and informal caregivers, this preference may be a reflection of their perceived importance of regular consultations in detecting any recurrence (as identified in the systematic literature review and qualitative study: Chapter 2-3). Therefore in accordance with discrete choice theory perceive frequent appointments as maximising their health (Grossman, 1972). In accordance with the findings from the qualitative study (Chapter 3), and in support of the Health Belief Model (Rosenstock, 1974; Becker, 1974; Becker & Rosenstock, 1984) and Theory of Planned Behaviour (Ajzen, 1985; Ajzen, 1991; Ajzen & Fishbein, 1970) this might be due to patients and their informal caregivers perceived susceptibility of a recurrence, and perceived ability of follow-up to reduce the perceived severity of a recurrence on their life. Therefore attendance at follow-up could be in part, a coping response (Self-regulation model of illness: Leventhal et al., 1992).

Concordantly, as identified in the discrete choice experiment (Chapter 5), patients and their informal caregivers did not value the provision of personalised care in their decision making regarding which model of care they preferred, but discordantly, Health Care Professionals did. This finding could be explained by the small sample size (Chapter 5), but is nevertheless surprising given the strength of patients and their informal caregivers' preferences for personalised care, as identified in the qualitative study (Chapter 3). Personalised care was not a theme in the systematic literature review (Chapter 2), from the perspective of patients or Health Care Professionals, but this could be due to the deductive nature of studies identified.

Overall it can be seen that patients, their informal caregivers and Health Care Professionals have both concordant and discordant preferences for gynaecology cancer follow-up care, both within and between stakeholder groups. This is interesting but not surprising given the individual nature of illness representations and the individual factors that affect this (Leventhal et al., 1992), and the individual nature of experiences, attitudes and beliefs governing behavioural intentions in accordance with social cognitive models of behaviour (Ajzen, 1985; Ajzen, 1991), and the knowledge and experience of Health Care Professionals, as the provider of care. Nevertheless, the concordance and discordance identified between patients, their informal caregivers and Health Care Professionals' preferences for gynaecology cancer follow-up, raises important questions for commissioners and researchers, and highlights the

problem of the agency relationship and preference misdiagnosis in gynaecology cancer follow-up (Mulley et al., 2012).

The differential findings between studies could be explained by the use and benefit of different methods to answer the research question (Tariq & Woodman, 2010), the deductive nature of the limited studies identified in the systematic literature review (Chapter 2) and the relatively small sample size recruited in the discrete choice experiment (Chapter 5: see strengths and limitations). Nevertheless, illustrates the differential findings that can be obtained from mixed method research. This raises important questions for policy makers in the interpretation of evidence to guide decisions, and the choice of studies which should be funded to generate valid and reliable findings to inform policy decisions.

6.2 Novel Contribution

The studies reported in this thesis make multiple novel contributions to both policy and health economics.

6.2.1 Contribution to methodology

The systematic literature review (Chapter 2) used a configurative and aggregative approach which aimed to explore which is novel in health economics, where systematic literature reviews generally aim to test. The use of transparent reporting, in accordance with the PRISMA guidance (Moher et al., 2009) aids the conduct of future exploratory systematic literature reviews conducted within the field, and the conduct of future systematic reviews to generate attributes for a discrete choice experiment.

The search terms used in this review identified that relevant empirical papers were poorly indexed and so were not identified when only using terms relating to follow-up (as has been the case in previous systematic reviews of follow-up care), but were identified by terms that described the model of care, for example nurse. This finding highlights the need for more extensive, broader scoping searches in the development of the search criteria.

It is recommended that qualitative research is conducted to generate attributes for a discrete choice experiment (Louviere, et al, 2000; Lanscar & Louviere, 2008; Bridges et al., 2011; Coast & Horrocks., 2007; Coast et al., 2012; Kløjgaard et al., 2012). However, the conduct of qualitative research for this purpose is often reported only to a basic level questioning the rigour of such studies, the validity of the discrete choice experiment, its results, and any changes to decision making made based on these (Vass et al., 2017; Louviere et al., 2000; Coast et al., 2012; Coast & Horrocks, 2007). The methodological rigour of the qualitative research study (Chapter 3) and the transparency of reporting (meeting the COREQ guidance: Tong, et al, 2007), can be used to aid health economists in the design, conduct and analysis of qualitative research for the purpose of discrete choice experiment design.

To date, there have been no published comparisons of attributes generated from multiple independent methods, and from the perspective of different stakeholders, despite this research recommendation, given the potential impact on the validity of the discrete choice experiment and decision making based on this (Ryan et al., 2014; Coast et al., 2012; Coast & Horrocks, 2007). Chapter 4 provides a comparison of attributes generated from different independent methods and from the perspective of the same and different stakeholders. Different methods identified both concordant and discordant attributes, within and between different stakeholder perspectives. Therefore this methodological comparison further highlights the importance of a clear rational and transparent methodology in the choice of methods used to inform discrete choice experiment design (Coast et al., 2012). This novel contribution is considered to be of significant importance to health economics methodology.

Throughout the thesis, the generation and comparison of preferences from different stakeholders perspectives is novel but of high importance given the prevalence and impact of preference misdiagnosis, the need for patient involvement in decision making, the role and impact of caregiving, and impact of Health Care Professional's preferences on the model of care delivered (Brodaty & Donkin, 2009; Department of Health, 2012; Mulley et al., 2012; Stacey et al., 2011; van Hezewijk et al., 2011). The systematic literature review (Chapter 2) compared the preferences of patients and Health Care Professionals (there were no existing studies of informal caregivers preferences); the qualitative study (Chapter 3) compared the preferences of patients and informal caregivers; the mixed method comparison (Chapter 4) compared the attributes generated from methods conducted from the perspective of patients, informal caregivers, Health Care Professionals, and policy makers; the discrete choice experiment (Chapter 5) compared the preferences of patients, their informal caregivers and Health Care Professionals.

This thesis took a health economic and health psychology approach to identify and understand preferences for care. Acknowledging illness regulations, experiences, attitudes and beliefs underlying stated preferences has undoubtedly strengthened the contributions of this thesis. The transparently reported interdisciplinary and mixed method approach can be used by health economists to strengthen their research studies and respective outcomes, enabling them to make more meaningful contributions to the clinical field.

6.2.2 Contributions to policy

There is a need to understand patients, their informal caregivers and Health Care Professional's preferences for gynaecology cancer follow-up care from a policy perspective (Buchannan, 1988; de Bock

et al., 2004; Leeson et al., 2017; Lydon et al., 2009; Morrison et al., 2012; Mulley et al., 2012; Revenson et al., 2016; Stacey et al., 2011; National Cancer Survivorship Initiative., 2013; van Hezewijk et al., 2011; Wales Cancer Network, 2016; Welsh Government and NHS Wales, 2016; Wolff & Roter, 2011; Williams et al., 2013: see chapter 1 for an in-depth discussion).

Systematic literature reviews are a trustworthy evidence base in which to inform policy making (Person, 2004; Aromataris & Pearson, 2014; Liberati et al., 2009; Higgins & Green, 2011; Munn et al., 2018). Chapter 2 is the first systematic literature review to consider patients, their informal caregivers and Health Care Professional's preferences for gynaecology cancer follow-up care. The descriptive map provides a methodic summary of the existing research, highlights gaps in the existing research and provides a context for interpreting the results of the synthesis (EPPI-centre, 2010). A systematic synthesis of existing research conducted in the United Kingdom explored patients and Health Care Professionals' preferences for care (no studies exploring the preferences of informal caregivers were identified). This systematic literature review identified that patients valued follow-up care because of the reassurance provided in absence of detection of disease recurrence. Because of the reassurance provided, patient's valued the provision of hospital led care. However, Specialist Nurse led telephone follow-up was valued by some patients, particularly those with experience of this care. Some patients valued the hypothetical idea of General Practitioner led or self-management/patient-initiated care. Results identified that Health Care Professionals felt the aim of care was to detect recurrence, monitor and treat any side effects, and collect data. Due to the inefficiencies of the current model of hospital led care and burden on the clinic, generally Health Care Professionals had a preference for the changing model of care (however, some had a preference for hospital led care). Within the changing model of care, Health Care Professionals valued self-management and multidisciplinary models of care, with specialist nurse and/or General Practitioner involvement. The systematic literature review identified a dearth of existing high quality evidence exploring patients and Health Care Professional's preferences for gynaecology cancer follow-up, and no previous research exploring informal caregiver's preferences despite their importance (Chapter 2). The systematic literature review identified a need for further research, conducted to a high methodological quality and using an inductive and deductive approach to ensure plausible and policy relevant attributes of care are identified, whilst valuing patients and their informal caregivers' experiences and preferences for care.

Given the findings of the systematic literature review and its limited use to aid health care decision making for the changing model of follow-up care, two further research studies were conducted, a qualitative study (Chapter 3) and a discrete choice experiment (chapter 4). The qualitative study was used to generate attributes and levels for the discrete choice experiment, but also makes novel contributions

in its own right due to the interdisciplinary approach taken. Identification and exploration of patients and their informal caregivers' preferences for gynaecology cancer follow-up based on their experience of this care provides information to policy makers of why attributes of care were valued, aiding policy development and health care practice change. Use of an inductive and deductive approach ensured that the themes were generated from the data but described the attributes of care. Endometrial, ovarian and cervical cancer patients diagnosed with different stages of disease, whom received different treatments and were currently receiving follow-up care were recruited. The study identified that patients and their informal caregiver's preferences for gynaecology cancer follow-up are embedded in their need for reassurance, and perceived ability of follow-up care to detect any recurrence and reduce the severity of any disease detected. To meet their need for reassurance, patients valued having access to an expert, medical procedures and holistic care. However, system failures and a low self-confidence prevented some patients from accessing care and reassurance. To meet their need for reassurance informal caregivers valued the provision of person centred information, which was met through the delivery of person centred care. This study was reported in accordance with the COREQ checklist (Tong et al., 2007), recruited gynaecology cancer patients and their informal caregivers whom had received different cancer pathways and used both an inductive and deductive approach to data generation. This study therefore makes significant novel contributions to aid health care decision making.

Discrete choice experiments can be used to inform health care policy decision making (Ryan, 2004). Despite the importance of the changing model of gynaecology cancer follow-up, no prior discrete choice experiments had been conducted. The aim of the discrete choice experiment was to understand patients, their informal caregivers and Health Care Professionals' relative preferences and trade-off's for the different attributes that comprise gynaecology cancer follow-up care, and compare and contrast preferences identified between stakeholders. Differing methods of data collection, and derived from the perspective of different stakeholders, can be used to generate attributes for a discrete choice experiment (Harrison et al., 2017; Vass et al., 2017; Hetler & Boehler, 2016). However, there is no published research comparing the attributes generated from multiple methods, and from the perspective of different stakeholders (Ryan et al., 2014; Coast et al., 2012; Coast & Horrocks., 2007). This thesis (chapter 4) identified that the method used to generate attributes, affects the attributes generated. This raises important issues for policy decisions that are made based on the findings from a discrete choice experiment. Attributes generated from the systematic literature review, qualitative study with patients and their informal caregivers, informal discussion with Health Care Professionals, current practice and policy recommendations were used to design the discrete choice experiment (Chapter 5). The discrete choice experiment identified that patients valued Consultant led follow-up, a higher frequency of

appointments, a medical examination if it could detect asymptomatic recurrence, and being delivered comprehensive information. Patients were willing to trade 17.4 months between appointments to see the Consultant over the General Practitioner, but less time between appointments to have a medical examination if it could as opposed to could not detect asymptomatic recurrence (6.8 months), and comprehensive over basic information (4.3 months). The amount of time patients were willing to trade to receive their desired level of care is an indication of the strength of their preference for care. Informal caregivers valued the patient receiving appointments of a higher frequency and valued the patient seeing the Consultant for their appointments. Informal caregivers were willing to trade 12.7 months between appointments for the patient to receive appointments delivered by the Consultant over the General Practitioner. Conversely, Health Care Professionals had a preference for the patient to receive a medical examination if it could detect asymptomatic recurrence, for their appointment to be delivered by different members of the multidisciplinary team dependant on the patients' needs, for the patient to receive comprehensive information and personalised care. The discrete choice experiment was reported in accordance with the checklist for reporting discrete choice experiments in health care (Bridges et al., 2011), and substantial mixed methods research was used to generate the attributes used to describe the model of care. The discrete choice experiment makes significant novel contributions to aid policy makers and health care commissioners in their health care decision making.

Exploration of preferences from a health economic and health psychology perspective, rather than merely a health economic perspective as outlined in the original grant proposal, adds to the significant contributions to policy that this thesis has made. Acknowledging the effect and impact of illness appraisals, attitudes, beliefs, experiences, others and contextual factors on preferences for care, has shaped each element of the thesis, and adds to the significant policy contribution this thesis makes (see strengths and limitations for further discussion).

Identification of preferences for care from the perspective of multiple stakeholders is relatively novel but particularly important from the perspective of policy change. Understanding the preferences of patients is of particular importance given the Welsh NHS strategy on prudent health care, policy focus on patient centred services to improve patient outcomes, the prevalence and impact of preference misdiagnosis and the effect this has on the efficiency of care (Coalition for Collaborative Care, 2014; Dixon, 2012; Mulley et al., 2012; The Kings Fund, 2010; Wales Cancer Network, 2016; Welsh Government and NHS Wales, 2016). As identified in the systematic literature review (Chapter 2), prior to the studies conducted within this thesis, there have been no studies of informal caregivers' preferences for care. However, because of their role and the impact of caregiving on the caregiver and the patient, there is a policy focus on understanding and involving informal caregivers as well as patients in the decision making

for patients' care (National Alliance for caregiving and AARP, 2015; Hodgen, Greenfield, Nugus & Kierman, 2013; Williams, et al, 2013; Wolff & Roter, 2011; Leung, et al, 2014; Luszczynska, et al, 2013; Courtens, et al, 1996; Department of Health, 2012). The preferences of Health Care Professionals influence the model of care they deliver, even when there are guidelines in place for the model of care that should be delivered (van Hezewijk et al., 2011). Health Care Professionals believe they understand the preferences of the patient they provide care for, however, patients and Health Care Professionals' preferences for care are often discordant to varying degrees, with preference misdiagnosis prevalent and leading to inefficiencies in care delivery (Stacey et al., 2011; Harrison et al., 2017; Mulley et al., 2012). It was therefore important to understand patients and their informal caregivers' preferences based on their experience of care. Exploration of patients, informal caregivers and Health Care Professionals' preferences throughout the thesis (chapter 2-3, 5) provides policy makers and health care commissioners with information regarding concordant and discordant preferences between stakeholders. This knowledge can be used to reduce preference misdiagnosis and improve the outcomes and efficiency of gynaecology cancer follow-up care (Mulley et al., 2012; Welsh Government and NHS Wales, 2016).

In conclusion, it is considered that each study conducted as part of the thesis makes a novel contribution to both health economics methodology and policy. These novel contributions, at this point in time, are of particular importance given strategic focus on prudent health care within the NHS in Wales, the inefficiencies of the current model of care, policy focus on a changing model of care and finite resources of the National Health Service (Bradley et al, 1999; Hodgkinson et al, 2007; Leeson et al., 2017; Lydon et al., 2009; Miller et al, 2003; Morrison et al., 2012; Oshima et al, 2011; Sekse et al, 2010; National Cancer Survivorship Initiative, 2013; Welsh Government and NHS Wales, 2016).

6.3 Comparison to previous research

The aim of the thesis was to identify patients, their informal caregivers and Health Care Professionals' preferences for gynaecology cancer follow-up care. The findings presented in the empirical chapters of this thesis confirm or conflict with current understanding, but all expand on our knowledge, and therefore elicit a range of thoughts and recommendations for future care and stakeholder involvement in health care decision making more generally.

Overall, this thesis makes new and improved contributions to the literature on preferences for gynaecology cancer follow-up care (for example, Lydon et al., 2009; Kew et al., 2007; Kew et al., 2009). The finding from the systematic literature review (Chapter 2) and qualitative research study (Chapter 3),

that patients value gynaecology cancer follow-up because of the reassurance it provides in the absence of asymptomatic disease detection, confirms previous findings identified in systematic reviews of cancer follow-up generally, and qualitative and quantitative studies of gynaecology cancer follow-up (Oshima et al, 2011; Sekse et al, 2010; Lewis et al., 2009b; Lydon et al., 2009; Bradley et al, 1999). However, emphasises that patients and their informal caregivers are not informed about the limitations of the traditional model of follow-up care, and the false reassurance that the traditional model of care can provide (Leeson et al., 2017). If patients are not informed, they are not co-producers of care, are unable to make informed decisions about the care they receive, and their preferences are misinformed. This might be why patients and informal caregivers did not have a preference to receive only the care needed, not more, not less, in line with research regarding the effectiveness and cost-effectiveness of gynaecology cancer follow-up, and principles of prudent health care (Welsh Government and NHS Wales, 2016).

Existing research has shown that patients place a greater value on a gynaecological examinations than more scientific medical tests such as pelvic ultrasounds, CT scans, chest x-rays, and blood tests (Griemel et al., 2011; Kew et al., 2009). The systematic review (Chapter 2) and the qualitative study (Chapter 3) also identified that patients (and in addition their informal caregivers) place a significant value on gynaecological examinations. However, conversely to previous research, the qualitative findings identified that more value was placed on scientific medical tests compared to examinations, due to the additional reassurance it was felt these provided. The provision of scientific medical tests was not explored in the discrete choice experiment as the delivery of such tests are implausible as part of standard follow-up in the United Kingdom given the evidence base, finite resources and focus on a changing model of care to reduce inefficiencies and improve outcomes (Lesson et al., 2017). However, form part of routine follow-up care practice within some European countries (Vistad et al., 2012).

Previous research regarding preferences for whom should deliver follow-up care is inconsistent, as is also reflected in the empirical findings within this thesis. Previous research exploring patients preferences has identified that Consultant led care is strongly valued (Kew et al, 2009; Bradley et al, 1999), whilst other studies have identified that Specialist Nurse led care is valued (Lewis et al., 2009b), or has mixed valuations (Lydon et al, 2009). The systematic literature review (Chapter 2) and qualitative study (Chapter 3) also identified that patients have differing valuations for the Health Care Professional that they would like to deliver care. The qualitative study (Chapter 3) identified that some patients had a strong preference to see the 'top dog', whilst others were happy to, or had a preference to receive care delivered by the Specialist Nurse either face-to-face or on the telephone. Within this study it was also identified that patients generally had a preference for care not to be delivered by the General Practitioner, but valued the idea of care delivered within the multidisciplinary team. However, the discrete choice

experiment (Chapter 5) identified that patients have a preference for care to be delivered by the Consultant and do not value care to be delivered by the General Practitioner, whilst care delivered by the Specialist Nurse and care delivered within the multidisciplinary team was not important to patients' decision making for which model of care they preferred. This differential finding could in part be explained by findings of the qualitative study and previous research, that patients with direct experience of specialist nurse led care have a preference for this model of care over standard care (Morrison et al., 2018; Beaver et al., 2017; Cox et al., 2008; Frew et al., 2010; Salkeld et al., 2000), and that patients value consistency of care provider which is not possible when care is delivered by trainee Doctors on rotation (Lewis et al., 2009b). However, could be due to the relatively small sample size of participants in the discrete choice experiment (see Chapter 5). Previous research regarding Health Care Professionals' preference for whom should provide care is also mixed, with multidisciplinary care valued by some (Brennan et al., 2011; Brennan et al., 2010), Specialist Nurse led care also valued by some (Lewis et al., 2009b), but care delivered by the General Practitioner not valued due to the belief that General Practitioner's do not have the required expertise to detect a recurrence, and so provide reassurance (Lewis et al., 2009b). The findings from the systematic literature review (Chapter 2) supports the finding that there is a preference for multidisciplinary care, but with involvement from the Specialist Nurse and/or General Practitioner. The discrete choice experiment (Chapter 5) confirmed the finding that multidisciplinary care was valued, but identified that care led by a General Practitioner was not, whilst care delivered by the Specialist Nurse and care delivered by the Consultant was not important to Health Care Professionals' decision making for which model of care they preferred.

The qualitative study (Chapter 3) identified an emerging relationship between patients' preferences for care and cancer demographics (treatment received, whether the patient was currently experiencing physical treatment effects) and psychological factors (appraisal of the disease, perceived risk of susceptibility and severity of a recurrence, and the perceived benefit of follow-up/outcome expectancies and the value of this, and coping). Whereas in informal caregivers, preferences seemed to be mediated by caregiving factors (caregiving role and caregiving tasks), and psychological factors (illness representations, perceived susceptibility and perceived severity, and the perceived benefit of follow-up/outcome expectancies and the value of this, and coping). These findings offer support to the self-regulation model of illness (Leventhal et al., 1992), health belief model (Becker, 1974; Becker & Rosentock, 1984; Rosentock, 1974), and theory of planned behaviour (Ajzen, 1991; Ajzen, 1985) in understanding factors underlying behavioural intention, and stated preferences. Due to the sample size obtained in the discrete choice experiment (Chapter 5), subgroup analyses to explore whether preferences differed by these individual factors could not be conducted. There is a policy and research focus on survivorship care

plans in cancer care and gynaecology cancer care more specifically, with the aim of recognising individual needs and improving patients outcomes (National Cancer Survivorship Initiative, 2013; Salani, 2013). Previous research highlights that patients and Health Care Professional's value the idea of a survivorship care plans to meet unmet needs (Dahl et al., 2015; Brennan et al., 2011; Brennan et al., 2010; Hewitt, Bamundo, Day & Harvey, 2007). In support, a key theme in the qualitative data (Chapter 3), from both the patients and informal caregivers' perspective, was personalised care, personalised in response to a needs assessment, with onward referrals if needed. At the time of the study patients were not given survivorship care plans as part of their follow-up care. However, a previous cross-sectional study conducted post treatment for gynaecology cancer care identified no effect of the survivorship care plan on service received, overall experience and likelihood to recommend care. Although it must be noted that a lack of effect could be explained by the significantly lower scores that were allocated to one treating physician (Brothers et al., 2013).

There is little previous research identifying the informational preferences of gynaecology cancer follow-up patients, and no research exploring the informational preferences of informal caregivers and Health Care Professionals. The empirical findings regarding informational preferences reported in this thesis are therefore of particular interest. A systematic review of information needs in the follow-up period for cancer generally identified that in the follow-up period, patients have informational needs about the treatment received and rehabilitation (Finney-Rutten, Arora, Bakos, Aziz & Rowland, 2005). More recently, a randomised feasibility trial that delivered personalised information verbally over the telephone by the Specialist Nurse, and comprehensive written information in a personalised booklet, identified an improvement in quality of life (Morrison et al., 2018). However, the qualitative study (Chapter 3) identified that patients had differing preferences for differing amounts of information. Comparatively, informal caregivers had a strong preference for comprehensive person centred information delivered verbally by the Health Care Professional, with information reported to be the reason for attending follow-up appointments, but a preference for brief written information. These results offer support to the finding from the discrete choice experiment (Chapter 5), which identified that the provision of comprehensive over basic information was not important to informal caregivers' decision making for which model of care they preferred. However, the provision of comprehensive over basic information was important to patients and Health Care Professional's decision making for which model of follow-up they preferred, with patients willing to trade 4.3 months to receive comprehensive over basic information.

Previous research demonstrates that overall patients have a preference to receive appointments that are the same frequency to which they are currently receiving them, of which varies between no

scheduled appointments (self-management) and 2 yearly appointments (Nicolaije et al., 2013; Greimel et al., 2011; Kew et al., 2009; Kew et al., 2007). However, a questionnaire study identified that in regard to vulvar cancer follow-up, gynaecologists have a strong preference for frequent appointments (Penney et al., 1995). The empirical findings within the thesis provide partial support to this previous research. The qualitative study (Chapter 3) and discrete choice experiment (Chapter 5) identified that patients and their informal caregivers generally value the frequency in which care was delivered to them, valuing more frequent over less frequent appointments, but valuing appointments of a reducing frequency as it was perceived this was an indicator that they were getting better. Interestingly, the qualitative study (Chapter 3) also identified that patients and informal caregivers felt the frequency of appointments should be decided by Health Care Professionals based on the patients' need. However, the discrete choice experiment identified that frequency of care was not valued by Health Care Professionals.

One of the main findings in this thesis is that preferences for gynaecology cancer follow-up care differ between stakeholder perspectives. Although this is unsurprising and not a new finding as such (see for example Harrison et al., 2017; Shin et al., 2013) this is of key importance when designing and delivering health care services. Preference misdiagnosis has been discussed as being equivalent to a medical misdiagnosis given the effect on the model of care delivered, and so the outcomes of care and efficiency of care (Mulley et al., 2012). If patients are fully informed they can 1) make informed decisions regarding their follow-up care, 2) health care would become closer to the market system advocated by Enthoven, (1985), 3) this would have a huge cost saving to the NHS given that when fully informed, patients consume less health care (Mulley et al., 2012; Wennberg, et al., 2010). Informing patients and their informal caregivers of the efficacy of gynaecology cancer follow-up is a key step to achieving prudent health care (Welsh Government and NHS Wales, 2016). Therefore understanding that preferences for care differ by stakeholder (patient/informal caregiver/Health Care Professional) as reported here, raises important questions for the design and delivery of services where patient preferences are often assumed (Mulley et al., 2012).

Another main finding of the thesis is that preferences for gynaecology cancer follow-up differed within stakeholders' perspectives. Previous studies of patients' preferences for gynaecology cancer follow-up have generally not explored differences in preferences by demographics. However, in a cross-sectional study of 54 patients diagnosed with early stage gynaecological cancer, the authors explored whether patients who had a preference for discharge or continued follow-up had different demographics. It was identified that participants with a preference for discharge were more likely to, and was significantly predicted by car ownership, being unemployed, and have lower levels of anxiety (as measured on the mental adjustment to cancer scale: Bradley et al., 2000). However, Howells et al (1999) reported that by

cancer type, there was no statistically significant differences in the preferences for continued follow-up or discharge. Comparatively, previous research in cancer care generally, has identified that preferences for care differ by psychological factors such as perceived benefits and barriers to care, as well as negative affect (Burns et al., 2005).

This thesis provides the first study of stated preferences to explore preferences for gynaecology cancer follow-up, providing information on relative preferences and the trade-off's that stakeholders are prepared to make between attributes. The rigorous and transparently reported mixed methods used to generate the attributes for the discrete choice experiment, from the perspective of multiple stakeholders, ensured that the attributes and levels used to define follow-up care were valid and plausible (Coast et al., 2012; Coast & Horrocks, 2007).

This thesis contributes to the literature on discrete choice experiment design methodology (Vass et al., 2017; Hetler & Boehler, 2016; Ryan et al., 2014; Coast et al., 2012; Coast & Horrocks, 2007). There are no known previous comparisons of attributes generated from different independent methods, and from the perspective of different stakeholders for the purpose of discrete choice experiment design, despite this recommendation (Ryan et al., 2014; Coast et al., 2012; Coast & Horrocks, 2007). The findings from the methodological comparison (Chapter 4) are therefore novel, and add to the existing literature for attribute generation. The findings support the guideline for attribute generation stated by Coast et al (2012), that there is a need for a clear rationale for the methodological choices for attribute selection, acknowledging the available resources of the researchers' knowledge and skills, as well as time available. One of the main findings within this methodological comparison is that narrative literature reviews, in comparison to systematic literature reviews, are subjective and so should not be used to inform decision making (Munn et al., 2018). This raises important findings for the validity of discrete choice experiments and decision making based on these, particularly with the recommendations for, and how frequently literature reviews are conducted to inform discrete choice experiment design (Abihiro et al., 2014; Bridges et al., 2011; Bessen et al., 2014; Burton et al., 2014; Hall et al., 2006; Howard, et al 2015; Kunaviktikul et al., 2014; Lanscar & Louviere, 2008; Robinson et al., 2015). The transparent reporting of the systematic literature review (Chapter 2) and the qualitative study (Chapter 3), can aid health economists in the conduct of these methods for discrete choice experiment attribute generation, given that the existing literature is dominated by basic reporting (Vass et al., 2017). Overall the findings from the methodological comparison add to the increasing literature for attribute generation (Vass et al, 2017; Hetler & Boehler, 2016; Coast et al., 2012; Coast & Horrocks, 2007).

6.4 Recommendations for future policy and practice

This thesis identified that patients and their informal caregivers are not informed of the evidenced based efficiency of gynaecological cancer follow-up care. As such, their attendance and engagement in follow-up care is based on a false reassurance, with this belief underpinning their preferences for care. Within the need for prudent health care and reducing preference misdiagnosis, the first step is to inform patients and their informal caregivers of the limited efficacy of gynaecology cancer follow-up care. Informing patients and their caregivers of this needs to be done sensitively, because of the investment in, and value placed on follow-up.

Once patients and their informal caregivers are informed, their preferences for care may differ (Mulley et al., 2012; Stacey et al., 2011).

6.4.1 Model of care

Given the finite resources and infinite needs, and need for prudent health care, a risk stratified model of gynaecological cancer follow-up should be developed and tested for its effectiveness and cost-effectiveness in line with policy guidance and the findings from this thesis.

- Patients with, 1) a low risk of disease recurrence, 2) which is likely to present symptomatically, and 3) treatment for a recurrence is not with curative intent, should be assigned to an informed self-management pathway. It is important that patients and informal caregivers are informed of the efficacy of the traditional model of care, and evidence regarding any recurrence of disease. Patients should be informed of potential signs of recurrence, and provided with the contact details for re-access to care if they have any concerns.
- Patients who 1) are at a higher risk of recurrence and 2) which is more likely to present asymptotically, and 3) can be treated with curative intent, should be assigned regular follow-up care with the relevant specialist in secondary care.
- Patients (and their informal caregivers) with salient biopsychosocial needs should be provided, or directed to, services that can meet their holistic needs following identification of these needs in a simple, brief needs assessment.

Such stratification will:

- 1) Place the responsibility back on patients in managing their disease. This may help patients and their informal caregivers 'let go' of the need for regular follow-up appointments, as reassurance will be provided by no presence of symptoms of recurrence, rather than a consultation and physical examination.

- 2) Allow reinvestment of resources from the un-necessary investment of resources which provides no research based evidence of increased survival, to the investment of resources to improve the biopsychosocial outcomes of patients and their informal caregivers.
- 3) Reducing unnecessary burden on clinics and Health Care Professionals.

6.5 Strengths and limitations

The aim of the thesis was to explore patients, their informal caregivers and Health Care Professional's preferences for gynaecology cancer follow-up, and to do so using a range of methods including a systematic literature review (Chapter 2), a qualitative study (Chapter 3) and a quantitative study employing a discrete choice experiment (Chapter 5). Combined with a literature review, policy guidance, current practice, informal discussion with Health Care Professionals and nominal group task, the attributes generated from the systematic literature review and qualitative study were compared, providing a further strength to this thesis (Chapter 4). Within the chapters of this thesis, multiple novel contributions to clinical and theoretical literature have been made, adding to clinical and theoretical knowledge.

In addition to the novel contributions described above, offered by the findings reported in the preceding chapters, there are many other notable strengths to this body of work as a whole. Firstly, the addition of the discipline of health psychology combined with the epistemological standpoint of the researcher, has been an asset to what at the grant proposal phase of this studentship aimed to use a purely health economic approach. Health psychology provides information on determinants of preferences, acknowledging the effect and impact of illness representations, experiences, attitudes, and beliefs on preferences for care, and acknowledges the intention-behaviour gap (Ajzen, 2011; Ajzen, 1991; Ajzen, 1985; Becker, 1974; Becker & Rosentock, 1984; Leventhal et al., 1992; Matarazzo, 1982; Morrison & Bennett, 2016; Rosenstock, 1974). Acknowledging the effect and impact of these factors shaped each element of the thesis, and adds to the significant contribution this thesis makes.

- A configurative and aggregative approach was taken to the design, conduct and analysis of the systematic literature review given the research question, hypothesised heterogeneity of research papers identified, and the benefit of exploring rather than merely summing the findings. This approach to systematic literature reviews conducted in health economics is novel, and was an asset to this review and knowledge gained from it. The systematic literature review explored health psychology papers, with no health economic papers identified by the thorough search strategy. The synthesis aimed to explore why

preferences differed by demographics, but due to the limited number of studies identified and generally poor methodology, this was not possible.

- The qualitative study explored preferences based on experiences of care. The supportive environment created for the interviews and focus groups, acknowledged the effect of, and importance of 'feeling comfortable' on individuals elicitation of preferences and these underlying factors. Exploring why patients and informal caregivers held these preferences, as well as asking them to put themselves in 'someone else's shoes' (through presentation of the case studies) aided reflection and elicitation of preferences and underlying factors governing these, and which were not identified in the nominal group task. Furthermore, patients and informal caregivers struggled with the nominal group task which is more traditionally conducted as 'qualitative research' within health economics (Hilgsmann et al., 2013; Powell et al., 2015). The nominal group task identified invalid attributes and invalid group rankings (see Chapter 2 for further discussion). Despite the importance of a top down approach (to ensure attributes are relevant to policy questions), and bottom up approach (to ensure attributes are relevant to patients) researchers have rarely used the framework approach to analyse qualitative data (Richie & Spencer, 1994; Vass et al., 2017).
- Health psychology seeks to answer why. Understanding of this discipline was central to shaping the development and conduct of the methodological comparison (Chapter 4), rather than merely accepting the attributes generated from the systematic literature review and qualitative study.
- The interdisciplinary approach taken improved the content validity of the discrete choice experiment by ensuring that attributes valid to decision making were identified, their perspective understood and contextual factors identified (Mangham, et al, 2008: Coast & Horrocks, 2007; Hall, et al, 2004; Abihiro et al., 2014) thus ensuring requirements of attribute generation were met: Ryan 1996).
- It was an aim of the discrete choice experiment (Chapter 5) to explore whether preferences differed by demographic factors, including health psychology factors such as quality of life and perceived risk of recurrence. However, due to the relatively small sample size and recruitment resources available this was not possible.

Secondly, each component of study has a strong and transparent methodology, the choice of which was designed in accordance with the aims, research questions and resources available. Each chapter is reported in accordance with the relevant methodological standard. The systematic literature review was

conducted in accordance with the EPPI centre guidance (Gough et al., 2012) and reported using the PRISMA framework (Moher et al., 2009). The qualitative research study was conducted in accordance with the guidance described by Krueger & Casey (2009) and Barbour (2008) and reported in accordance with the COREQ checklist (Tong, et al, 2007). The discrete choice experiment was conducted in accordance with the guidance described by the conduct of discrete choice experiments to value health and health care (Ryan et al., 2014; Ryan et al., 2008), and reported using the IPSOR Good Research Practices for Conjoint Analysis Task Force checklist (Bridges et al., 2011).

Thirdly, this thesis is embedded in clinical knowledge and understanding, through attendance at relevant training courses (Good Clinical Practice; Informed consent; Communicating research), extensive reading and communication with different professionals involved in the design and delivery of the gynaecology cancer pathway at the local oncology and gynaecology centres. Being an active member of the working group for a trial conducted alongside this PhD research further aided this (TOPCAT-G: Morrison et al., 2018). Involvement of patients and informal caregivers in the design of the qualitative study and discrete choice experiment, to ensure readability and understanding of the materials and measures, also supported this.

Fourthly, the thesis have been enriched by the conduct of mixed methods research to answer the research questions of the thesis (O’Cathain, Murphy & Nicholl., 2010; Tariq & Woodman, 2010; Creswell & Plano Clark, 2007). The aim of using a mixed method approach was to develop, using the systematic literature review and qualitative research study to inform the design of the discrete choice experiment. The use of triangulation (using the data generated from each method to support the overall findings) was also invaluable. The qualitative study and discrete choice experiment have equal weighting within the thesis. The systematic literature review has less weighting in the interpretation of the results from the thesis as a whole, given the dearth of evidence identified within the systematic literature review, the relevance of individual studies in answering the questions of the systematic review, and overall quality of individual studies identified (Tariq & Woodman, 2010). Regarding the timeframe of mixed methods it was anticipated that the systematic literature review would be designed and analysed before the qualitative study, which would be designed and analysed before the design and analysis of the discrete choice experiment (Tariq & Woodman, 2010). However, given the resources and timings required for completion for each of the research studies i.e., refining the research questions in the systematic review and extensive scoping searches conducted, the descriptive map for the systematic review was conducted after the qualitative study but before the methodological comparison, and the synthesis for the systematic review was conducted after the discrete choice experiment. The separate conduct and analysis of the systematic literature review, qualitative study and discrete choice experiment, then comparison of findings between

studies ensured that the quality of each study as a stand-alone research study was maintained, whilst enhancing the findings from the thesis as a whole (O’Cathain, et al, 2010). This mixed method approach to answering the aims of the thesis increased understanding of patients, their informal caregivers and Health Care Professional’s preferences for care given the depth of insight this approach provided (Tariq & Woodman, 2011).

There are also however several limitations to this PhD. Chronologically, the aim was for the systematic literature review to be conducted first, in order to directly inform the qualitative study, and subsequently the discrete choice experiment. However, given resource constraints and competing PhD research priorities this was not possible as described above. The timing of the conduct of the systematic literature review may have influenced the design of other components reported within the thesis, for example, based on the findings from the systematic literature review synthesis, the semi structured interview guide may have usefully and explicitly raised the topic of alternative models of care (Specialist Nurse led telephone follow-up/involvement from the Specialist Nurse, General Practitioner led follow-up/involvement from the General Practitioner, self-management). However, as the attribute, expertise of Health Care Professional was identified from all methods of attribute generation, including the descriptive map, expertise of Health Care Professional was an attribute within the discrete choice experiment.

Chapter two (the systematic literature review) explored the preferences of patients, informal caregivers and Health Care Professional’s, chapter three (the qualitative research study) explored the preferences of patients and their informal caregivers, and chapter five (the discrete choice experiment) explored the preferences of patients, their informal caregivers and Health Care Professional’s. The reason for this difference in exploration of stakeholders’ perspectives is very much a reflection on my development as a researcher throughout this thesis. It is stated in the grant proposal that the preferences of patients and their informal caregivers would be explored. As my awareness of the literature and research policy questions pertinent to this thesis developed, so did my understanding of the importance of the need to also collect the preferences of Health Care Professional’s delivering gynaecology cancer follow-up. Therefore whilst this is a limitation of my thesis, it is very much a reflection of my journey and development as a researcher throughout this PhD. Exploring the preferences of Health Care Professionals adds further impact to this PhD.

The grant proposal stated that the discrete choice experiment would be administered in a multi-centre research study across Wales. However, along with the other components of the thesis, applications to multiple NHS R&D centres and management of this was not feasible within the resources available. As such, all the findings were obtained from a sample of patients treated within Betsi Cadwaladr University

Health Board, the largest health board in Wales covering six counties and a population of approximately 678,000 people, their respective informal caregivers and Health Care Professional's (<http://www.wales.nhs.uk/sitesplus/861/page/84723>). Therefore whilst the model of care delivered in North Wales is similar to the model of care generally delivered throughout the United Kingdom this may affect the generalisability of the results (Leeson et al., 2013). Given the health board in which participants were recruited from, patients and their informal caregivers were generally of a white British nationality. It is acknowledged that this may affect the generalisability of the results. Given the heterogeneity of model of gynaecology cancer follow-up delivered across Europe and the World (Vistad et al., 2012; Suprasert & Manopunya (2011; Oshima et al., 2011; Oshima et al., 2013), and preference for the status quo (Salkeld, et al., 2000), whilst the results will be of interest to other countries it cannot be assumed that the same results would have been obtained.

Sampling bias refers to differences between the sample and the population (Henderson & Page, 2007). To reduce sampling bias the population was defined and participants were recruited methodologically from all clinics within recruiting hospitals (including telephone clinics) to ensure that the sample was representative of the population in terms of cancer type. However, participants have their own motivations for taking part in research. For example in qualitative research, motivations for participating may include altruism, to voice praise or concern for care, to have someone to talk to and to have access to support services (Gysels, Shipman & Higginson, 2008). As focus groups were held on the Tenovus Cancer Care mobile unit with the offer of provision of support from Tenovus cancer care once the focus group had finished, the qualitative study may have attracted participants more in need of support. This may reflect some of the difference in results between the qualitative study and discrete choice experiment, for example, the qualitative study (Chapter 3) identified that patients placed a strong value on personalised care, but in the discrete choice experiment (Chapter 5) this was not important to patients' decision making for which model of care they preferred. Demographically, some individuals are more likely to take part in research studies, for example, females are more likely to take part in cross-sectional studies than males which may in part explain the low uptake of informal caregivers to the discrete choice experiment (Henderson & Page, 2007). To prevent participants living rurally or/and with restrictive co-morbidities from being able to take part in the qualitative study Chapter 3), interviews were held in the participants' home or on the telephone if following consent they were unable to attend a focus group. Data was not collected on non-responders, so responders and non-responders characteristics could not be compared. By the nature of exploring preferences for follow-up care in a population of people attending for follow-up appointments, any potential patients in the post treatment phase who opt out of follow-up by not attending their appointment, were not invited to participate in the studies.

Conversations with clinicians ascertained that patients do not typically opt-out of follow-up, and screening data from the studies supports this (the number of people scheduled for and attending the clinic generally corroborates: Personal communication, 2015). However, given the system failures experienced by the sample recruited for the qualitative study, some patients post treatment may not be participating in follow-up, and as such, were not invited to participate in the research studies undertaken as part of this research programme. Therefore the findings will not reflect their perspectives.

It must be noted that it is unknown whether participants in the studies synthesised in the systematic literature review (Chapter 2) were informed as to the evidence and limitations of the current model of care. Analysis of the qualitative study (Chapter 3) identified that bar one patient, patients and their informal caregivers were not informed about the evidence for and limitations for the current model of care. This was not measured in the discrete choice experiment and participants were not informed of the efficacy of the current model of care due to ethical concerns. However, this poses the question as to whether if patients and their informal caregivers were informed, would they identify different preferences (Mulley et al., 2012).

Studies conducted within the PhD were cross-sectional in nature, and all bar one study identified in the systematic literature review was cross-sectional in nature. This poses limitations regarding the generalisability of results. However, patients and so their informal caregivers were eligible to participate if they were at any point of their follow-up journey, so having received only one appointment to having received their last follow-up appointment before being discharged from care, to try and minimise this effect. Although, illness representations, experiences, attitudes and beliefs are individual (Ajzen, 1985; Ajzen, 1991; Becker, 1974; Becker & Rosenstock, 1984; Leventhal et al., 1992).

Whilst efforts were taken to reduce the cognitive burden of the discrete choice experiment (minimal number of items in discrete choice experiment questionnaire, piloting, face-to-face research nurse/research officer support to complete the questionnaire, telephone support from researcher) some participants/potential participants struggled to understand the concept of the discrete choice experiment. As such, it was reported that this cognitive burden prevented some patients/informal caregivers from completing the discrete choice experiment.

Research conducted within the thesis aimed to identify stated preferences. With stated preference methods there is a degree of uncertainty as to whether hypothetical decisions match preferences in real life situations (Quaife, Terris-Prestholt, Di Tanna & Vickermann, 2018). However, the exploration of revealed preferences was not plausible in this context as the models of care explored in the discrete choice experiment are not currently offered to patients.

6.6 Future research

Given the above, future research should address the discrepancies identified between the findings from the systematic literature review (Chapter 2) and qualitative study (Chapter 3), compared to the findings from the discrete choice experiment (Chapter 5). It is unknown why the findings from the systematic literature review and qualitative study highlighted a strong preference for patient led holistic care delivered within the multidisciplinary team, but that when described in the discrete choice experiment, these attributes and levels were not important to patients and their informal caregiver's decision making for which model of care they preferred. Similarly, in the qualitative study, informal caregivers placed a strong value on information received, but in the discrete choice experiment, this attribute was not important to decision making for which model of care informal caregivers preferred.

Once this has been explored, future research should explore preferences for care in a sample of patients and informal caregivers who are informed about the aims, limitations and options for care delivery, so that patients can make informed decisions regarding their care. When patients are informed, they have been shown to make different decisions regarding their preferences for care (Mulley et al., 2012; Stacey et al., 2011). This is important within the strategy of prudent health care in Wales (Welsh Government and NHS Wales, 2016). It is essential that alternative models of care are explored in randomised control trials, so that the impact of a patient and informal caregiver preferred model of care, if plausible, can be evaluated before practice changes. There is a particular time element on this, given the emerging policy guidance for a changing model of care, limitations of the current model of care, patients' unmet needs, finite resources within the National Health Service, and current changing model of care within some health boards (Leeson et al., 2017; Stacey et al., 2011; Revenson et al., 2016; Wales Cancer Network, 2016; National Cancer Survivorship Initiative., 2013; Williams et al., 2013; Morrison et al., 2012; Mulley et al., 2012; van Hezewijk et al., 2011; Wolff & Roter, 2011; Lydon et al., 2009; Buchannan, 1988: see Chapter 1 for an in-depth discussion).

6.7 Reflexivity

"A researcher's background and position will affect what they choose to investigate, the angle of investigation, the methods judged most adequate for this purpose, the findings considered most appropriate, and the framing and communication of conclusions" (Malterud, 2001, p. 483-484).

6.7.1 Prior assumptions and experience

I have an undergraduate degree in Psychology and a master's Degree in Health Psychology. In between undertaking my undergraduate degree and master's degree I gained a wealth of experience working with different vulnerable populations within social care.

My experience and training from working with vulnerable populations was undoubtedly an asset in the qualitative research study, providing me with the interpersonal skills needed to conduct this research. I was confident approaching and communicating with Health Care Professionals, patients and their informal caregivers. Because of my understanding of how experience influences preferences, I spent a day at the cancer treatment centre talking to different Health Care Professionals to try and gain some understanding of the experience patients face, and spent time talking about the patient experience with patient and informal caregiver representatives at the time of the design of the qualitative study. Prior training and experience has given me the skill of being a 'good listener'. This was an asset to the focus group and interview study. Because of this experience and knowledge I was particularly wary of the risk of harm to participants by discussing their cancer experience and health care experience. I put steps in place to minimise harm to participants. My health psychology and social care experience, and resulting holistic and deductive approach to research undoubtedly influenced the content of the semi-structured interview guide, and methods of analysis in the systematic literature review and qualitative study, as well as interpretation of results as a whole. I was very mindful of the need to make patients and informal caregivers voices heard (Krueger & Casey, 2009).

I attended the Good Clinical practice training course, and 'Obtaining Informed Consent' course led by NISCHR CRC whilst designing the research project. Good clinical practice guidelines were adhered to in the conduct of this research.

6.7.2 The role of the research team as collaborators

Multidisciplinary collaborative research is recommended from a policy making perspective, combining the strengths of different research fields to answer the research questions (Bhavsar, 2017). My first supervisor, Professor Rhiannon Tudor Edwards, is a health economist specialising in public health. My second supervisor, Professor Val Morrison, is a health psychologist specialising in cancer and informal caregiving. Seow Tien Yeo, a health economist specialising in discrete choice experiments, supported this element of the thesis. I had regular contact with a gynaecology oncologist (SL), an oncologist (NS) and a specialist nurse (LH).

The knowledge, experience and insight gained from iterative discussions with Val Morrison aided the analysis and refinement of the themes in the systematic literature review and qualitative study. The

experience and knowledge insight from Seow Tien Yeo was beneficial in designing the normative group component of the interview guide, and reducing the number of attributes and levels for the discrete choice experiment. The final list of attributes and levels was discussed within the multidisciplinary team. My experience echoes the complexities of using qualitative research to generate attributes as discussed by Coast and colleagues (2012). I found the reductionist nature of identifying attributes and levels from the mixed method data complex, and at the time felt the attributes and levels over simplified the knowledge gained (Coast & Horrocks, 2007). In hindsight, I feel that the final choice of attributes and levels did the data justice.

The multidisciplinary approach taken ensured understanding of the clinical field and the issues faced clinically, as well as the importance of understanding individuals' experience and preferences at the individual and group level, with the aim of improving patients and their informal caregiver's outcomes and the efficiency of care.

6.7.3 Fair dealing

Fair dealing refers to ensuring that the research incorporates different perspectives, so that the perspective of one group is not presented as the truth, reducing bias (Dingwall, 1992). Participants in the qualitative study were purposefully recruited to ensure that despite the small sample size associated with qualitative research, participants had received differing diagnoses of gynaecology cancer. Participants in the qualitative research study and discrete choice experiment had been diagnosed with different types of gynaecology cancer, received different treatments, and were different durations post treatment. Due to resource restraints, all participants were recruited within Betsi Cadwaladr University Health Board. During analysis, no particular individuals or group views were considered superior to another.

The concept of fair dealing is the opposite to the reductionist approach of the nominal group technique. Researchers with a different epistemological perspective may have found this technique beneficial in generating attributes and levels for the discrete choice experiment.

6.7.4 Awareness of the wider social and political context

At the time of the PhD research there was intermittent media coverage regarding the finite resources of the NHS, infinite demands, the need to make financial cuts and discussion around privatisation of the NHS in England (BBC News, 2014a; BBC News 2014b). I was mindful of the possible impact of this on the findings of the qualitative study and discrete choice experiment. The semi-structured interview guide used case studies to encourage participants to think about their experiences and preferences for care based on their experience. This was successful in breaking down the 'we are happy with the care, everything is

fine' attitude that some participants originally presented with. Some focus group and interview participants refused to talk about willingness to pay values as a monetary figure.

6.8 Conclusion

This thesis valued, explored and identified the preferences of patients, their informal caregivers and Health Care Professionals for the changing model of gynaecology cancer follow-up care. An interdisciplinary approach, using a health economic and health psychology perspective was taken. This ensured that determinants of preferences were explored providing greater benefit to policy makers and health care commissioners in their decision making. A mixed method inductive and deductive approach comprised of a systematic literature review, qualitative study and discrete choice experiment was undertaken. Each element of this thesis, and the findings from the thesis as a whole make multiple novel contributions to both health economic methodology and policy. This thesis provides information to policy makers and health care commissioners within the need for a changing model of care, to improve the efficiency of care within limited resources.

References

- Abihiro, G. A., Leppert, G., Mbera, G. B., Robyn, P. J., & De Allegri, M. (2014). Developing attributes and attribute-levels for a discrete choice experiment on micro health insurance in rural Malawi. *BMC Health Services Research*, 14, 235.
- Ajzen, I. & Fishbein, M. (1970). The prediction of behaviour from attitudinal and normative beliefs. *Journal of Personality and Social Psychology*, 6(4), 466-487.
- Ajzen, I. (1985). From intentions to actions: A theory of planned behaviour. In Kuhl, J. & Beckman, J. (eds). *Action-control: From cognition to behaviour*. Heidelberg: Springer-Verlag.
- Ajzen, I. (1991). The theory of planned behaviour. *Organizational Behaviour and Human Decision Processes*, 50(2), 179-211.
- Allirajah, D. (2010). Demonstrating the economic value of co-ordinated cancer services. An examination of resource utilisation in Manchester.
<http://www.macmillan.org.uk/Documents/AboutUs/Commissioners/MonitorResearchinManchesterbriefing.pdf>
- Aromataris, E. & Pearson, A. (2014). The systematic review: An overview. *The American Journal of Nursing*, 114(3), 55-58.
- Arrow, K. J. (1963). Uncertainty and the welfare economics of medical care. *The American Economic Review*, 5(3), 941-973.
- Aslam, R. W., Pye, K. L., Rai, T. K., Hall, B., Timmis, L. J., Yeo, S. T., & Leeson, S. (2016). Follow-up strategies for women with endometrial cancer after primary treatment (Protocol). *Cochrane Database of Systematic Reviews*, 2016(10), CD012386.
- Barbour, R. (2008). *Doing Focus Groups*. London: Sage.
- BBC News (2014a). Review 2014: NHS turbulence continues in Wales. Retrieved from <http://www.bbc.co.uk/news/uk-wales-30575898>
- BBC News (2014b). The very public NHS privatisation debate. Retrieved from <http://www.bbc.co.uk/news/health-28581878>
- Beaver, K., Williamson, S., Sutton, C., Hollingworth, W., Gardner, A., Allton, B., Abdel-Aty, M., Blackwood, K., Burns, S., Curwen, D., Ghani, R., Keating, P., Murray, S., Tomlinson, A., Walker, B., Willet, M., Wood, N., & Martin-Hirsch, P. (2017). Comparing hospital and telephone follow-up for patients treated for stage-I endometrial cancer (ENDCAT) trial: A randomised, multicentre, non-inferiority trial. *BJOG*, 124, 150-160.
- Becker, M. H. (1974). The health belief model and the personal health behaviour. *Health Education Monographs*, 2, 324-508.

- Becker, M. H. & Maiman, L. A. (1975). Socio-behavioural determinants of compliance with health and medical care recommendations. *Medical care*, 13(1), 10-14.
- Becker, M. H., Haefner, D. P., & Maiman, L. A. (1977). The health belief model in the prediction of dietary compliance: A field experiment. *Journal of Health and Social Behaviour*, London: Academic Press.
- Becker, M. H. & Rosenstock, I. M. (1984). Comparing social learning theory and the health belief model. In Ward, W. B. (ed.), *Advances in Health Education and Promotion*, Greenwich, CT: JAI Press.
- Beesley V., Eakin, E., Steginga, S., Aitken, J., Dunn, J., & Battistutta, D. (2008). Unmet needs of gynaecological cancer survivors: implications for developing community support services. *Psycho-oncology*, 17, 392-400.
- Bessen, T., Chen, G., Street, J., Elliott, J., Karnon, J., Keefe, D., & Ratcliffe, J. (2014). What sort of follow-up services would Australian breast cancer survivors prefer if we could no longer offer long-term specialist-based care? A discrete choice experiment. *British Journal of Cancer*, 110, 859-867.
- Boynton, P. M. & Greenhalgh, T. (2004). Hands-on guide to questionnaire research: selecting, designing, and developing your questionnaire. *BMJ*, 328(7451): 1312-1315.
- Bradley, E. J., Pitts, M. K., Redman, C. W. E., & Calvert, E. (1999) The experience of long-term hospital follow – up for women who have suffered early stage gynecological cancer: a qualitative interview study. *International Journal of Gynecological Cancer*, 9(6), 491-496.
- Bradley, E. J., Pitts, M. K., Redman, C. W. E., Calvert, E., Howells, R., & Wafai, C. (2000). What are the factors associated with the follow-up preferences of women in long-term remission from gynaecological cancer? *Journal of Obstetrics and Gynaecology*, 20(4), 408-411.
- Brandenberg, D., Berendsen, A. J., & G.H. de Bock, G. H. (2017). Patients' expectations and preferences regarding cancer follow-up care. *Maturitas*, 105, 58–63.
- Bray, N. (2015). More than mobility: Applying health economics to wheelchair interventions for disabled children. Thesis, School of Healthcare Sciences, Bangor University.
- Brazier, J., Ratcliffe, J., Salomon, J. A. and Tsuchiya, A. (2007). *Measuring and valuing health benefits for economic evaluation*. Oxford: Oxford University Press.
- Brennan, M. E., Butow, P., Marven, M., Spillane, A. J., & Boyle, F. M. (2011). Survivorship care after breast cancer treatment-experiences and preferences of Australian women. *Breast*, 20(3), 271–277.

- Brennan, M. E., Butow, P., Spillane, A. J., & Boyle, F. M. (2010) Survivorship care after breast cancer: Follow-up practices of Australian health professionals and attitudes to a survivorship care plan. *Asian Pacific Journal of Clinical Oncology*, 6(2), 116–125.
- Bridges, J. F. P., Hauber, A. B., Marshall, D., Lloyd, A., Prosser, L. A., Regier, D. A., Johnson, F. R., & Mauskopf, J. (2011). Conjoint Analysis Applications in Health – a checklist: A report of the ISPOR good research practices for conjoint analysis task force. *Value in Health*, 14(4), 403 – 413.
- British Gynaecological Cancer Society and Royal College of Obstetrics and Gynaecologists, (2014). *Guidelines for the Diagnosis and Management of Vulval Carcinoma*. Retrieved from <https://bgcs.org.uk/Vulval%20Cancer%20booklet%2020052014.pdf>
- Brodaty, H., & Donkin, M. (2009). Family caregivers of people with dementia. *Dialogues in clinical neuroscience*, 11(2), 217-228.
- Brothers, B. M., Easley, A., Salani, R., & Andersen, B. L. (2013). Do survivorship care plans impact patients' evaluations of care? A randomized evaluation with gynecologic oncology patients. *Gynecologic Oncology*, 129(3), 554-558.
- Buchanan, A. (1988). Principal/agent theory and decision making in health care. *Bioethics*, 2(4), 317-333.
- Budd, L., Morrison, V., Seale, T., & Edwards, R. T. (2015). Patients, carers and health professionals' preferences for gynaecological cancer follow-up care: a mixed method systematic review (Protocol). Centre for reviews and dissemination. Retrieved from http://www.crd.york.ac.uk/PROSPERO_REBRANDING/display_record.asp?ID=CRD42015016230
- Burns, D. S., Sledge, R. B., Fuller, L. A., Daggy, J. K., & Monahan, P. O. (2005). Cancer patients' interest and preferences for music therapy. *Journal of Music Therapy*, 42(3), 185-199.
- Burton, C. R., Fargher, E., Plumpton, C., Roberts, G., Owen, H., & Roberts, E. (2014). Investigating preferences for support with life after stroke: a discrete choice experiment. *BMC Health Services Research*, 14(63), 1-13.
- Cardella, J., Coburn, N. G., Gagliardi, A., Maier, B.-A., Greco, E., Last, L., Smith, A. J., Law, C., Wright, F. (2008). Compliance, attitudes and barriers to post-operative colorectal cancer follow-up. *Journal of evaluation in clinical practice*, 14, 407-415.
- Carr, E.C., & Worth, A. (2001). The use of the telephone interview for research. *Journal of Research in Nursing*, 6, 511–524.
- Carroll, F. E., Owen-Smith, A., Shaw, A., & Montgomery, A. A. (2012). A qualitative investigation of the decision-making process of couples considering prenatal screening for Down syndrome. *Prenatal diagnosis*, 32(1), 57–63.

- Center for the Evaluative Clinical Sciences. (2007). *Preference-Sensitive Care*. A Dartmouth Atlas Project Topic Brief.
- Chalmers, I., Atkinson, P., Fenton, M., Firkins, L., Crowe, S., & Cowan, K. (2013). Tackling treatment uncertainties together: The evolution of the James Lind Initiative, 2003-2013. *Journal of the Royal Society of Medicine*, 106(12), 482-491.
- Chan, Y. M., Ngan, H. Y. S., Li, B. Y. G., Yip, A. M. W., Ng, T. Y., Lee, P. W. H., Yip, P. S., & Wong, L. C. (2001). Longitudinal study on quality of life after gynecologic cancer treatment. *Gynecologic Oncology*, 83(1), 10–9.
- Clarke, T., Galaal, K., Bryant, A., & Naik, R. (2014). Evaluation of follow-up strategies for patients with epithelial ovarian cancer following completion of primary treatment. *Cochrane Database of Systematic Reviews*, 2014(9). CD006119.
- Coalition for Collaborative Care. (2014). *Our vision for the future*. Retrieved from http://coalitionforcollaborativecare.org.uk/wpcontent/uploads/2014/11/C4CC_210x297_final_pages.pdf
- Coast, J., & Horrocks, S. A. (2007). Developing attributes and levels for discrete choice experiments using qualitative methods. *Journal of Health Services Research & Policy*, 12(1), 25–30.
- Coast, J., Al-Janabi, H., Sutton, E. J., Horrocks S. A., Vosper, J. A., Swancutt, D. R., & Flynn, T. N. (2012). Using qualitative methods for attribute development for discrete choice experiments: issues and recommendations. *Health Economics*, 21(6) 730-741.
- Cooke, R., & French, D. P. (2008). How well do the theory of reasoned action and theory of planned behaviour predict intentions and attendance at screening programmes? A meta-analysis, *Psychology and Health*, 23(7), 745-765.
- Coulter, J., & Gleeson, N. (2003). Local and regional recurrence of vulval cancer: Management dilemmas. *Best Practice & Research Clinical Obstetrics & Gynaecology*, 17(4), 663-681.
- Courtens, A.M., Stevens, F.C.J., Crebolder, H., & Philipsen, H. (1996). Longitudinal study on quality of life and social support in cancer patients. *Cancer nursing*, 19(3), 162-169.
- Cox, A., Bull, E., Cockle-Hearne, J., Knibb, W., Potter, C., & Faithfull, S. (2008). Nurse led telephone follow-up in ovarian cancer: A psychosocial perspective. *European Journal of Oncology Nursing*, 12(5), 412-417.
- Crabtree, B. F., Yanoshik, M. K., Miller, M. L., & O'Connor, P. J. (1993). Selecting individual or group interviews. In: Morgan DL (Ed) *Successful Focus Groups: Advancing the State of the Art*. (pp 137-149). Newbury Park. CA: Sage.

- Creswell, J. W., & Plano Clark V. L. (2007) *Designing and Conducting Mixed Methods Research*. London. Sage.
- Culyer, A. J. (1971). The nature of the commodity 'Health care' and its efficient allocation. *Oxford Economic Papers*, 23(2), 189-211.
- Dahl, L., Wittrup, I., Væggemose, U., Petersen, L. K., & Blaakaer, J. (2013) Life after gynecologic cancer — a review of patients quality of life, needs, and preferences in regard to follow-up. *International Journal of Gynecological Cancer*, 23(2), 227–234.
- Dahl, L., Wittrup, I., Petersen, L. K., Blaakaer, J., & Væggemose, U. (2015). Paradoxes of follow-up – health professionals' views on follow-up after surgical treatment in gynecological cancer. *Acta Oncologica*, 54(2), 194-199.
- De Bekker-Grob, E. W., Donkers, B., Jonker, M. F., & Stolk, E. A. (2015). Sample size requirements for discrete-choice experiments in healthcare: a practical guide. *Patient*, 8, 373-384.
- De Bekker-Grob, E. W., Ryan, M., & Gerard, K. (2012). Discrete choice experiments in health economics: A review of the literature. *Health Economics*, 21, 145-172.
- de Bock, G. H., Bonnema, J., Zwaan, R. E., van de Velde, C. J. H., Kievit, J., & Stiggelbout, A. M. (2004). Patient's needs and preferences in routine follow-up after treatment for breast cancer. *British Journal of Cancer*, 90(6), 1144–1150.
- De Brun, A., Flynn, D., Ternent, L., Price, C. I., Rodgers, H., Ford, G. A., Rudd, M., Lanscar, E., Simpson, S., Teah, J., & Thomson, R. G. (2018). A novel design process for selection of attributes for inclusion in discrete choice experiments: Case study exploring variation in clinical decision-making about thrombolysis in the treatment of acute ischaemic stroke. *BMC Health Services Research*. 18, 483.
- Department of Health (2012). Liberating the NHS: No decision about me, without me. (2012). Retrieved from https://consultations.dh.gov.uk/choice/choice-future-proposals/supporting_documents/Choice%20consultation%20%20No%20decison%20about%20me%20without%20me.pdf
- DeShazo, J. R., & Fermo, G. (2002). Designing choice sets for stated preference methods: the effects of complexity on choice consistency. *Journal of Environmental Economics and Management*, 44(1), 123-143.
- Deshields. T. L., Rihanek, A., Potter, P., Zhang, Q., Kuhrik, M., Kuhrik, N., O'Neill, J. (2012). Psychosocial aspects of caregiving: Perceptions of cancer patients and family caregivers. *Support Care Cancer*, 20, 349-356.
- Dingwall, R. (1992). Qualitative methods in health studies. In J. Daly, I. McDonald, & E. Willis (Eds.), *Researching health care* (pp. 161–175). London, England: Tavistock/Routledge.

- Dixon, A. (2012). Are we wasting money on care that patients don't want? Retrieved from <https://www.kingsfund.org.uk/blog/2012/05/are-we-wasting-money-care-patients-don%E2%80%99t-want>.
- Dixon, S., Nancarrow, S. A., Enderby, P. M., Moran, A. M., & Parker, S. G. (2015). Assessing patient preferences for the delivery of different community-based models of care using a discrete choice experiment. *Health Expectations*, 18(5), 1204-1214.
- Dixon, P., Beaver, K., Williamson, S., Sutton, C., Martin-Hirsch, P., & Hollingworth, W. (2018). Cost-consequence analysis alongside a randomised controlled trial of hospital versus telephone follow-up after treatment of endometrial cancer. *Applied Health Economics and Health Policy*, 16(3), 415-427.
- Edwards, W. (1954). The theory of decision making. *Psychological Bulletin*, 51(4), 380-417.
- Elit, L., Fyles, A. W., Devries, M. C., Oliver, T. K., & Fung-Kee-Fung, M. (2009). Follow-up for women after treatment for cervical cancer: a systematic review. *Gynecologic Oncology*, 114(3), 528–535.
- Engel, G. L. (1980). The clinical application of the bio-psychosocial model. *American Journal of Psychiatry*, 137(5), 535-544.
- Engel, G. L. (1977). The need for a new medical model: A challenge for biomedicine. *Science*. 196(4286), 129-136.
- Enthoven, A. (1985). *Reflections on the Management of the National Health Service: An American looks at incentives to efficiency in health services management in the UK*. London: Nuffield Provincial Hospitals Trust.
- EPPI-Centre (2010). *EPPI-Centre Methods for Conducting Systematic Reviews*. Retrieved from <http://eppi.ioe.ac.uk/cms/LinkClick.aspx?fileticket=hQBu8y4uVwI%3D&tabid=88>
- Figueredo, A., Rumble, R.B., Maroun, J., Earle, C.C., Cummings, B., McLeod, R., Zuraw, L., Zwaal, C. & Gastrointestinal Cancer Disease Site Group of Cancer Care Ontario's Program in Evidence-based Care. (2003). Follow-up of patients with curatively resected colorectal cancer: a practice guideline. *BMC Cancer*, 3, 26.
- Finch, J (1984). "It's great to have someone to talk to": the ethics and politics of interviewing women', in C. Bell & H. Roberts (eds), *Social Reasoning: Politics, Problems & Practice*. (pp 70-87). London: Routledge.
- Finney Rutten, L. J., Arora, N. K., Bakos, A. D., Aziz, N., & Rowland, J. (2005). Information needs and sources of information among cancer patients: A systematic review of research (1980-2003). *Patient Education and Counseling*, 57, 250-261.

- Flynn, T. N., Louviere, J. J., Marley, A. A., Coast, J., & Peters, T. J. (2008). Rescaling quality of life tariffs from discrete choice experiments for use as QALYs: a cautionary tale. *Population Health Metrics*, 6, 6.
- Fotopoulou, C., Hall, M., Cruickshank, D., Gabra, H., Ganesan, R., Hughes, C., Kehoe, S., Ledermann, J., Morrison, J., Naik, R., Rolland, P., & Sundar, S. (2017). *British Gynaecological Cancer Society (BGCS) Epithelial Ovarian / Fallopian Tube / Primary Peritoneal Cancer Guidelines: Recommendations for Practice*. Retrieved from <https://bgcs.org.uk/BGCS%20Guidelines%20Ovarian%20Guidelines%202017.pdf>.
- Frew, G., Smith, A., Zutshi, B., Young, N., Aggarwal, A., Jones, P., Kockelbergh, R., Richards, M., & Maher, E. J. (2010). Results of a Quantitative Survey to Explore Both Perceptions of the Purposes of Follow-up and Preferences for Methods of Follow-up Delivery Among Service Users, Primary Care Practitioners and Specialist Clinicians after Cancer Treatment. *Clinical Oncology*, 22(10), 874-884.
- Fung-Kee-Fung, M., Dodge, J., Elit L, Lukka, H., Chambers, A., & Oliver, T. (2006). Follow-up after primary therapy for endometrial cancer: A systematic review. *Gynecologic Oncology*, 101(3), 520-529.
- Gerard, K., Tinelli, M., Latter, S., Smith A., & Blenkinsopp, A. (2014). Patients' valuation of the prescribing nurse in primary care: a discrete choice experiment. *Health Expectations*, 18(6), 2223-2235.
- Giuffrida, A. and Torgerson, D. J. (1997). Should We Pay the Patient? Review of Financial Incentives to Enhance Patient Compliance. *BMJ*, 315(7110), 703-707.
- Gough, D. (2007). Weight of evidence: a framework for the appraisal of the quality and relevance of evidence. *Research Papers in Education*, 22(2), 213-228.
- Gough, D., Oliver, S., & Thomas, J. (2012). *An introduction to systematic reviews* (1st ed.). London: Sage.
- Greenhalgh, T., Robert, G., MacFarlane, F., Bate, P., & Kyriakidou, O. (2005). Diffusion of innovations in service organizations: Systematic review and recommendations. *The Milbank Quarterly*, 82(4), 581-629.
- Greimel, E., Lahousen, M., Dorfer, M., Lambauer, M., & Lang U (2011). Patients' view of routine follow-up after gynecological cancer treatment. *European Journal of Obstetrics & Gynecology and Reproductive Biology*, 159(1), 180-183.
- Grossman, M. (1972). On the concept of health capital and the demand for health. *The Journal of Political Economy*, 80(2), 223-255.
- Guerts, S. M. E., de Vegt, F., van Altena, A., Tjan-Heijnen, V. C. G., Massuger, L. F. A. G., Adang, E. M., van Dijck, J. A. A. M., & Verbeek, A. L. M. (2012). Impact of routine follow-up examinations on life

- expectancy in ovarian cancer patients: A simulation study. *International Journal of Gynecological Cancer*, 22(7), 1150-1157.
- Gysels, M., Shipman, C., & Higginson, I. J. (2008). "I will do it if it will help others:" Motivations Among Patients Taking Part in Qualitative Studies in Palliative Care. *Journal of Pain and Symptom Management*, 35(4), 347-355.
- Hahn, G. J & Shapiro, S. S. (1966). *A catalogue and computer program of the design and analysis of orthogonal symmetric and asymmetric fractional factorial experiments. Report 66-C-165*. General Electric Research and Development Centre, Schenectady, New York.
- Hall, J., Fiebig, D. G., King, M. T., Hossain, I., & Louviere, J. J. (2006). What influences participation in genetic carrier testing? Results from a discrete choice experiment. *Journal of Health Economics*, 25(3), 520-537.
- Hall, J., Viney, R., Haas, M., & Louviere, J. J. (2004). Using stated preference discrete choice modelling to evaluate health care programs. *Journals of Business Research*. 57(9), 1026-1032.
- Hall, M., & Rustin, G. (2011). Recurrent Ovarian Cancer: When and How to Treat. *Current Oncology Reports*, 13(6), 459-471.
- Hanoch, Y., & Gummerum, M. (2008). What can health psychologists learn from health economics: From monetary incentives to policy programmes. *Health Psychology Review*, 2(1), 2-19.
- Harden, A. (2010). Mixed-Methods systematic reviews: Integrating Quantitative and Qualitative. *Findings. Focus. Technical Brief Number 25*, 1-8.
- Harrison, M., Milbers, K., Hudson, M., & Bansback, N. (2017). Do patients and health care providers have discordant preferences about which aspects of treatments matter most? Evidence from a systematic review of discrete choice experiments. *BMJ Open*, 7(5), e014719
- Hauber, A. B., González, J. M., Groothuis-Oudshoorn, C. G., Prior, T., Marshall, D. A., Cunningham, C., IJzerman, M. J., & Bridges, J. F. (2016). Statistical Methods for the Analysis of Discrete Choice Experiments: A Report of the ISPOR Conjoint Analysis Good Research Practices Task Force. *Value in Health*, 19(4), 300-315.
- Helter, T. M., & Boehler, C. E. (2016). Developing attributes for discrete choice experiments in health: a systematic literature review and case study of alcohol misuse Interventions. *Journal of Substance Misuse*, 21(6), 662-668.
- Henderson, M. & Page, L. (2007). Appraising the evidence: what is selection bias? *Evidence Based Mental Health*, 10(3), 67-68.

- Hensher, D. A., Rose, J. M., & Greene, W. H. (2005). *Applied Choice Analysis: A Primer*. Cambridge: University Press.
- Hewitt, M. E., Bamundo, A., Day, R., & Harvey, C. (2007). Perspectives on post- treatment cancer care: qualitative research with survivors, nurses, and physicians. *Journal of Clinical Oncology*, 25(16), 2270–2273.
- Higgins, J. & Green, S. (2011). *Cochrane Handbook for Systematic Review of interventions*. Version 5.1.0. The Cochrane collaboration.
- Hiligsmann, M., van Durme, C., Geusens, P., Dellaert, B. G. C., Dirksen, C. D., van der Weijden, T., Reginster, J. -Y., & Boonen, A. (2013). Nominal group technique to select attributes for discrete choice experiments: An example for drug treatment choice in osteoporosis. *Patient Preference and Adherence*, 7, 133-139.
- Hochschild, A. R. (1983). *The managed heart: The commercialization of human feeling*. Berkeley, CA: University of California Press.
- Hodgen, A., Greenfield, D., Nugus, P. & Kierman, M. C. (2013). What are the roles of carers in decision-making for amyotrophic lateral sclerosis multidisciplinary care? *Patients Preference and Adherence*, 7, 171-181.
- Hodgkinson, K., Butow, P., Fuchs, A., Hunt, G. E., Stenlake, A., Hobbs, K. M., Brand, A., & Wain, G. (2007). Long-term survival from gynaecologic cancer: psychosocial outcomes, supportive care needs and positive outcomes. *Gynecologic oncology*, 104(2), 381-389.
- Hole, A. R (2006). *A comparison of approaches to estimating confidence intervals for willingness to pay measures*. University of York, Centre for Health Economics, research paper 8.
- Howard, K., Salkeld, G. P., Patel, M. I., Mann, G. J., & Pignone, M. P. (2015). Men’s preferences and trade-offs for prostate cancer screening: a discrete choice experiment. *Health Expectations*, 18(6), 3123-3135.
- Howell, D, Hack, T. F., Oliver, T. K., Chulak, T., Mayo, S., Aubin, M., Chasen, M., Earle, C. C., Friedman, A. J., Green, E., Jones, G. W., Jones, J. M., Parkinson, M., Payeur, N., Sabiston, C. M., & Sinclair, S. (2012). Models of care for post-treatment follow-up of adult cancer survivors: A systematic review and quality appraisal of the evidence. *Journal of Cancer Survival*, 6(4), 359-371.
- Howells, R. E. J., Bradley, E., Pitts, M., Calvert, E., & Redman, C. W. E. (1999). Audit of the views of gynaecological cancer patients regarding follow-up. *Journal of Obstetrics and Gynaecology*, 19(2), 167-168.
- INVOLVE. (2012). *Briefing notes for researchers: involving the public in NHS, public health and social care research*. INVOLVE, Eastleigh.

- Janda, M., Steginga, S., Dunn, J., Langbecker, D., Walker, D., & Eakin, E. (2008). Unmet supportive care needs and interest in services among patients with a brain tumour and their carers. *Patient Education and Counseling*, 71(2), 251-258.
- Jeffery, M., Hickey, B. E., Hider, P. N., & See, A. M. (2002). Follow-up strategies for patients treated for non-metastatic colorectal cancer. *Cochrane Database System Review*, 2016(11), CD002200.
- Jefford, M., Rowland, J., Grunfeld, E., Richards, M., Maher, J., & Glaser, A. (2013). Implementing improved post-treatment care for cancer survivors in England, with reflections from Australia, Canada and the USA. *British Journal of Cancer*, 108(1), 14–20.
- Jeppesen, M. M., Mogensen, O., Hansen, D. G., Iachina, M., Korsholm, M., & Jensen, P. T. (2017). Detection of recurrence in early stage endometrial cancer – the role of symptoms and routine follow-up. *Acta Oncologica*. 56(2), 262-269.
- Johnson, R., & Orme, B. (2003). Getting the most from CBC. Sawtooth Software Research Paper Series.
- Johnson, T. P., & Wislar, J. S. (2012). Response rates and nonresponse rates in surveys. *JAMA*, 307(17), 1805-1806.
- Kateeb, E. T., Warren, J., Gaeth, G., Damiano, P., Momany, E., Kanellis, M. J., Weber-Gasparoni, K., & Ansley, T. (2014). The willingness of US pediatric dentists to use atraumatic restorative treatment (ART) with their patients: a conjoint analysis. *Journal of Public Health Dentistry*, 74(3), 234-240.
- Kernick, D. P. (2003). Introduction to health economics for the medical practitioner. *Postgraduate Medical Journal*, 79(929), 147-150.
- Kerr-Wilson, R. H., & McCrum, A. (1995). Follow-up of patients with gynaecological cancer. *Australia and New Zealand Journal of Obstetrics and Gynaecology*, 35, 298–299.
- Kew, F. M., Galaal, K. & Manderville, H. (2009). Patients' views of follow-up after treatment for gynaecological cancer. *Journal of Obstetrics and Gynaecology*, 29(2), 135-142.
- Kew, F. M., Galaal, K., Manderville, H., & Verleye, L. (2007). Professionals' and patients' views of routine follow-up: a questionnaire survey. *International Journal of Gynecological Cancer*, 17(3), 557-560.
- Kim, Y., Spillers, R. L., & Hall, D. L. (2012). Quality of life of family caregivers 5 years after a relative's cancer diagnosis: Follow-up of the national quality of life survey for caregivers. *Psycho-oncology*, 21(3), 273-281.
- Kim, K. C. K., Howells, R. E. J., & Evans, A. S. (2004). The role of clinical follow-up in early stage cervical cancer in South Wales. *BJOG: An International Journal of Obstetrics and Gynaecology*, 111(12), 1444-1448.

- Kitzinger, J. & Barbour, R. S. (1999). 'Introduction: The Challenge and promise of focus groups' in, RS Barbour & J Kitzinger (eds). *Developing Focus Group Research: Politics, Theory and Practice*. (pp 1-20). London: Sage.
- Kløjgaard, M. E., Bech, M., & Sjøgaard, R. (2012). Designing a stated choice experiment: the value of a qualitative process. *Journal of Choice Modelling*, 5(2), 1–18.
- Kolutek, R., Avci, I. A., & Sevig, U. (2016). Effect of planned follow-up on married women's health beliefs and behaviours concerning breast and cervical cancer. *Journal of Cancer Education*, 33(2), 375-382.
- Krueger, R. A., & Casey, M. A. (2009) *Focus Groups: A practical guide for applied research*. London: Sage.
- Kunaviktikul, W., Chitpakdee, B., Srisuphan, W., & Bossert, T. (2014). Preferred choice of work setting among nurses in Thailand: A discrete choice experiment. *Nursing and Health Sciences*, 147(1), 126-133.
- Kuzel, A. J. (1992). 'Sampling in qualitative theory' in BF Crabtree & WI Miller (eds). *Doing Qualitative Research*. (pp 311-344) Newbury Park, CA: Sage.
- Lajer, H., Jensen, M. B., Kilsmark, J., Albæk, J., Svane, D., Mirza, M. R., Geertsen, P. F., Reerman, D., Hansen, K., Milter, K., & Mogensen, O. (2010). The value of gynecologic cancer follow-up: evidence based ignorance? *International Journal of Gynecologic Cancer*, 20(8), 1307–1320.
- Lancaster, K. (1966). A new approach to consumer theory. *Journal of Political Economy*, 74, 132-157.
- Lanceley, A., Fiander, A., McCormack, M., & Bryan, A. (2013). Follow-up protocols for women with cervical cancer after primary treatment (Review). *Cochrane Database of Systematic Reviews*, 2013(11), CD008767.
- Lanceley, A., Berzuini, C., Burnell, M., Gessler, S., Morris, S., Ryan, A., Ledermann, J. A., & Jacobs, I. (2017). Ovarian cancer follow-up: A preliminary comparison of 2 approaches. *International Journal of Gynecological Cancer*, 27(1), 59-68.
- Langer, S. L., Brown, J. D., & Syrjala, K. L. (2009). Intrapersonal and interpersonal consequences of protective buffering among cancer patients and caregivers. *Cancer*, 115(18), 4311-4325.
- Lanscar, E. & Louviere, J. (2008) Conducting discrete choice experiments to inform healthcare decision making: A user's guide. *Pharmacoeconomics*, 26(8), 661-677.
- Leeson, S. C., Beaver, K., Ezendam, N. P. M., Maćuks, R., Martin-Hirsch, P. L., Miles, T., Jeppesen, M. M., Jensen, P. T., & Zola P (2017). The future for follow-up of gynaecological cancer in Europe. Summary of available data and overview of ongoing trials. *European Journal of Obstetrics & Gynecology and Reproductive Biology*, 210, 376-380.

- Leeson, S., Stuart, N., Sylvestre, Y., Hall, L., & Whitaker R. (2013). Gynaecological cancer follow-up audit of current practice: national survey of current practice in the UK. *British Medical Journal Open*, 3(7), e002859.
- Leung, J., Pachana, N. A., & McLaughlin, D. P. (2014). Social support and health-related quality of life in woman with breast cancer: A longitudinal study. *Psycho-oncology*, 23(9), 1014-1020.
- Leventhal, H., Diefenbach, M., & Leventhal, E. A. (1992). Illness cognition: Using common sense to understand treatment adherence and affect cognition interactions. *Cognitive Therapy and Research*, 16(2), 143-163.
- Lewis, R. A., Neal, R. D., Hendry, M., France, B., Williams, N. H., Russell, D., Hughes, D. A., Russell, I., Stuart, N. S., Weller, D., & Wilkinson, C. (2009a). Follow-up of cancer in primary care versus secondary care: systematic review. *British Journal of General Practice*. 59(564), e234–e247.
- Lewis, R. A., Neal, R. D., Hendry, M., France, B., Williams, N. H., Russell, D., Hughes, D. A., Russell, I., Stuart, N. S., Weller, D., & Wilkinson, C. (2009b). Patients' and healthcare professionals' views of cancer follow-up: systematic review. *British Journal of General Practice*, 59(564):e248–e259.
- Lewis, R., Neal, R. D., Williams, N. H., France, B., Wilkinson, C., Hendry, M., Russell, D., Russell, I., Hughes, D. A., Stuart, N. S., & Wilkinson, C. (2009c). Nurse-led vs conventional physician-led follow-up for patients with cancer: systematic review. *Journal of Advanced Nursing*, 65(4), 706 – 723.
- Liberati, A., Altman, D. G., Tetzlaff, J., Mulrow, C., Gøtzsche, P. C., Ioannidis, J. P. A. Clarke, M., Devereaux, P. J., Kleijnen, J., & Moher, D. (2009). The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate health care interventions: Explanation and elaboration. *Guidelines and Guidance*, 6(7), e1000100.
- Lim, K. C., Howells, R. E., & Evans, A.S. (2004). The role of clinical follow-up in early stage cervical cancer in South Wales. *BJOG: An International Journal of Obstetrics and Gynaecology*. 111(12), 1444-1448.
- Linley, W. G. (2013). Reimbursement decisions for new medicines: Exploration of the preferences of decision-makers and the public. Institute of Medical and Social Care Research, Bangor University.
- Louviere, J. J., Hensher, D. A., & Swait, J. D. (2000). *Stated Choice Methods: Analysis and Application*. Cambridge: Cambridge University Press.
- Luszczynska, A., Pawlowska, I., Cieslak, R., Knoll, N. & Scholz, U. (2013). Social support and quality of life among lung cancer patients: A systematic review. *Psycho-oncology*, 22(10), 2160-2168.

- Lydon, A., Beaver, K., Newbury, C., & Wray, J. (2009). Routine follow-up after treatment for ovarian cancer in the United Kingdom (UK): Patient and health professional views. *European Journal of Oncology Nursing*, 13(5), 336-343.
- Lynn, F. A., Crealey, G. E., Alderdice, F. A., & McElnay, J. C. (2015). Preferences for a third-trimester ultrasound scan in a low-risk obstetric population: a discrete choice experiment. *Health Expectations*, 18(5), 892-903.
- Mangham, L. J., Hanson, K., & McPake, B. (2009). How to do (or not to do)... Designing a discrete choice experiment for application in a low-income country. *Health Policy and Planning*, 24(2), 151-158.
- Manski, C. F. (1977). The structure of random utility models. *Theory and decision*, 8(3), 229-254.
- Matarazzo, J. D. (1982). Behavioral health's challenge to academic, scientific and professional psychology. *American Psychologist*, 37(1), 1-14.
- McElduff P, Boyes A, Zucca A, Girgis A. (2004). Supportive Care Needs Survey: A Guide to Administration, Scoring and Analysis. The University of Newcastle: Newcastle, Australia.
- Mcfadden, D. (1974). Conditional logit analysis of qualitative choice behavior. In Zarembka, P. (Eds.). *Frontiers in econometrics*. New York: Academic Press.
- Miller, B. E., Pittman B., & Strong, C. (2003). Gynecologic cancer patients' psychosocial needs and their views on the physician's role in meeting those needs. *International Journal of Gynecological Cancer*, 13(2), 111-119.
- Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G for the PRISMA group (2009). Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *British Medical Journal*, 339, b2535.
- Morgan, D. L. (1988). *Focus Groups as Qualitative Research*. London: SAGE.
- Morris, S., Devlin, N., & Parkin, D. (2007). *Economic analysis in health care*. Chichester: Wiley.
- Morrison, V., & Bennett, P. (2016). *Introduction to Health Psychology*. United Kingdom: Pearson.
- Morrison, V., Spencer, L. H., Totton, N., Pye, K., Yeo, S. T., Butterworth, C., Hall, L., Whitaker, R., Edwards, R. T., Timmis, L. J., Hoare, Z., Neal, R. D., Wilkinson, C., & Leeson, S. (2018) Trial of Optimal Personalised Care After Treatment - Gynaecological Cancer (TOPCAT-G) A Randomized Feasibility Trial. *International Journal of Gynecological Cancer*, 28(2) 401-411.
- Morrison, V., Henderson, B. J., Zinovieff, F., Davies, G., Cartmell, R., Hall, A., & Gollins, S. (2012). Common, important, and unmet needs of cancer outpatients. *European Journal of Oncology Nursing*, 16(2), 115-123.

- Mulley A, Trimble C, & Elwyn G. (2012). *The problem of silent misdiagnosis*. Retrieved from http://www.kingsfund.org.uk/sites/files/kf/field/field_publication_file/patients-preferences-matter-may-2012.pdf
- Munn, Z., Peters, M. D. J., Stern, C., Tufanaru, C., McArthur, A., & Aromataris, E. (2018). Systematic review or scoping review? Guidance for authors when choosing between a systematic or scoping review approach. *BMC Medical Research Methodology*, 18(1), 143.
- Nama, V., Nordin, A., & Bryant, A. (2013). Patient-reported outcome measures for follow-up after gynaecological cancer treatment. *Cochrane Database of Systematic Reviews*, 2013(11).
- National Alliance for caregiving and AARP. (2015). *Caregiving in the U.S. 2015*. Bethesda, MD: National Alliance for Caregiving.
- National Cancer Survivorship Initiative (2013). *Living With and Beyond Cancer: Taking Action to Improve Outcomes*. Retrieved from <http://www.ncsi.org.uk/wpcontent/uploads/howtoguide.pdf>
- National Institute for Health Research (2013). Introduction to Good Clinical Practice (GCP): A practical guide to ethical and scientific quality standards in clinical research. Consolidated version 2.2 May 2013.
- Ngune, I., McManus, A., & Parsons, R. (2015). Predicting general practice attendance for follow-up cancer care. *American Journal of Health Behaviour*, 39(2), 167-174.
- NHS improvement. (2018). Reference costs 2017/18: National Schedule of Reference Costs. Retrieved from <https://improvement.nhs.uk/resources/reference-costs/#rc1718>
- Nicolaije, K. A. H., Ezendam, N. P. M., Vos, M. C., Boll, D., Pijnenborg, J. M. A., Kruitwagen, R. F. P. M., Lybeert, M. L. M., & van de Poll-Franse, L. V. (2013). Follow-up practice in endometrial cancer and the association with patient and hospital characteristics: A study from the population-based PROFILES registry. *Gynecologic Oncology*, 129(2), 324-331.
- Nicolaije, K. A. H., Husson, O., Ezendam, N. P. M., Vos, M. C., Kruitwagen, R. F. P. M., Lybeert, M. L. M., & van de Poll-Franse, L. V. (2012). Endometrial cancer survivors are unsatisfied with received information about diagnosis, treatment and follow-up: A study from the population-based PROFILES registry. *Patient Education and Counselling*, 88(3), 427-435.
- Novick, G. (2008). Is There a Bias Against Telephone Interviews in Qualitative Research? *Research in Nursing & Health*, 31, 391-398
- Noyes, J. & Lewin, S. (2011). Chapter 5: Extracting qualitative evidence. In J. Noyes, A. Booth, K. Hannes, A. Harden, J. Harris, S. Lewin, C. Lockwood (Eds), *Supplementary Guidance for Inclusion of Qualitative Research in Cochrane Systematic Reviews of Interventions*.

Version 1 (updated). London: Cochrane Collaboration Qualitative Methods Group.

O’Cathain, A., Murphy, E., & Nicholl, J. (2010). Three techniques for integrating data in mixed methods studies. *British Medical Journal*, 341, c4587.

Office for National Statistics (2013). *Cancer Survival in England: patients diagnosed 2007 – 2011 and followed up to 2012*. Retrieved from <http://www.ons.gov.uk/ons/rel/cancer-unit/cancer-survival/cancer-survival-in-england-patients-diagnosed-2007-2011-and-followed-up-to-2012/stb-cancer-survival-in-england-patients-diagnosed-2007-2011-and-followed-up-to-2012.html>

Olaitan, A., Murdoch, J., Anderson, R., James, J., Graham, J., & Barley, V. (2001). A critical evaluation of current protocols for the follow-up of women treated for gynecological malignancies: A pilot study. *International Journal of Gynecological Cancer*, 11(5), 349-353.

Onwumere, J., Kuipers, E., Bebbington, P., Dunn, G., Fowler, D., Freeman, D., Watson, P., & Garety, P. (2016). Caregiving and illness beliefs in the course of psychotic illness. *The Canadian Journal of Psychiatry*, 53(7), 460-468.

Oonk, M. H. M., de Hullu, J. A., Hollema, H., Mourits, M. J. E., Pras, E., Wymenga, A. N. M., & van der Zee, A. G. J. (2003). The value of routine follow-up in patients treated for carcinoma of the vulva. *American Cancer Society*, 98(12), 2624-2629.

Orme, B. (1998). *Sample size issues for conjoint analysis studies*. Sequim: Sawtooth Technical Paper.

Oshima, S., Kisa, K., Terashita, T., Habara, M., Kawabata, H., & Maezawa, M. (2011). A qualitative study of Japanese patients' perspectives on post-treatment care for gynecological cancer. *Asian Pacific Journal of Cancer Prevention*, 12, 2255-2261.

Oshima, S., Kisa, K., Terashita, T., Kawabata, H., & Maezawa, M. (2013). Care-seeking behavior of Japanese gynaecological cancer survivors suffering from adverse effects. *BMC Women’s health*, 13, 1.

Palmer, C., Pratt, J., Basu, B., & Earl H (2006). A study to evaluate the use of CA125 in ovarian cancer follow-up: a change in practice led by patient preference. *Gynecologic Oncology*, 101(1), 4-11.

Pearlin, L. I., Mullon, J. T., Semple, S. J., & Skaff, M. M. (1990). Caregiving and the stress process: an overview of concepts and their measures. *The Gerontologist*, 30(5), 583-594.

Penney, G. C., Kitchener, H. C., & Templeton, A. (1995). The management of carcinoma of the vulva: current opinion and current practice among consultant gynaecologists in Scotland. *Health Bulletin*, 53(1), 47-65.

Pearson, A. (2004). Balancing the evidence: Incorporating the synthesis of qualitative data into systematic reviews. *JBIM Reports*, 2(2), 45-64.

- Pearson, A., Jordan, Z., & Munn, Z. (2012) Translational science and evidence-based healthcare: A clarification and reconceptualization of how knowledge is generated and used in healthcare. *Nursing Research and Practice*, 2012, 792519.
- Powell, G., Holmes, E. A. F., Plumpton, C. O., Ring, A., Baker, G. A., Jacoby, A., Pirmohamed, M., Marson, A. G., & Hughes, D. A. (2015). Pharmacogenetic testing prior to carbamazepine treatment of epilepsy: Patients' and physicians' preferences for testing and service delivery. *British Journal of Clinical Pharmacology*, 90(5), 1149-1159.
- Quaife, M., Terris-Prestholt, F., Di Tanna, G. L., & Vickermann, P. (2018). How well do discrete choice experiments predict health choices? A systematic review and meta-analysis of external validity. *The European Journal of Health Economics*, 19(8), 1053–1066.
- Rabiee, F. (2004). Focus-group interview and data analysis. *Proceedings of the Nutrition Society*, 63(4), 655-660.
- Rae, H. M. (1998). Managing feelings caregiving as emotion work. *Research on Aging*, 20(1), 137-160.
- Råheim, M., Magnussen, L. H., Sekse, R. J. T., Lunde, A., Jacobsen, T., & Blystad, A. (2016). Researcher – researched relationship in qualitative research: shifts in positions and researcher vulnerability. *International Journal of Qualitative Studies on Health and Wellbeing*, 11, 30996.
- Reamy, A. M., Kim, K., Zarit, S. H., & Whitlatch, C. J. (2011). Understanding discrepancy in perceptions of value: Individuals with mild to moderate dementia and their family caregivers. *The Gerontologist*, 51(4), 473-483.
- Renehan, A. G., Egger, M., Saunders, M. P., & O'Dwyer, S. T. (2002). Impact on survival of intensive follow-up after curative resection for colorectal cancer: Systematic review and meta-analysis of randomised trials. *British Medical Journal*, 324(7341), 813.
- Revenson, T. A., Griva, K., Luszczynska, A., Morrison, V., Panagopoulou, E., Vilchinsky, N., & Hagedoorn, M. (2016). *Caregiving in the illness context*. Hampshire, UK: Palgrave Macmillan.
- Richardson, E. M., Schüz, N., Sanderson, K., Scott, J. L & Schüz, B. (2017). Illness representations, coping, and illness outcomes in people with cancer: A systematic review and meta-analysis. *Psycho-oncology*, 26(6), 724-737.
- Richie, J., & Spencer, L. (1994) Qualitative data analysis for applied policy research. In: Bryman A, Burgess RG (eds) *Analysing Qualitative Data*. (pp 173-194). London: Routledge.
- Robinson, S. M., Ní Bhuachalla, B., Ní Mhaille, B., Cotter, P. E., O'Connor, M., & O'Keeffe, S. T. (2015). Home, please: A conjoint analysis of patient preferences after a bad hip fracture. *Geriatrics and Gerontology international*, 15(10), 1165-1170.

- Robinson, P. & Lowe, J. (2015). Literature review vs systematic review. *Australian and New Zealand Journal of Public Health*, 39(2), 103.
- Romito, F., Goldzweig, G., Cormio, C., Hagedoorn, M., & Andersen, B. L. (2013). Informal caregiving for cancer patients. *Cancer*, 119(S11), 2160-2169.
- Rosenstock, I. M. (1974). The health belief model and preventative health behaviour. *Health Education Monographs*, 2(4), 354-386.
- Rustin, G. J. S., van der Burg, M. E. L., Griffin, C. L., Guthrie, D., Lamont, A., Jayson, G. C., Kristensen, G., Mediola, C., Coens, C., Qian, W., Parmar, M. K. B., Swart, A. M., MRC OV05, & EORTC 55955 investigators (2010). Early versus delayed treatment of relapsed ovarian cancer (MRC OV05/EORTC 55955): a randomised trial. *Lancet*, 376(9747): 1155–1163.
- Ryan, M. & Gerrard, K. (2003). Using discrete choice experiments to value health care programmes: Current practice and future research reflections. *Applied Health Economics and Health Policy*, 2(1), 55-64.
- Ryan, M. (1996). Using consumer preferences in health care decision making. The application of conjoint analysis. London: Office of Health Economics.
- Ryan, M. (2004). Discrete choice experiments in health care: NICE should consider using them for patient centred evaluations of technologies. *BMJ*, 328, 360-361.
- Ryan, M., & Farrar, S. (2000). Eliciting preference for healthcare using conjoint analysis. *BMJ*, 320, 1530-1535.
- Ryan, M., Bate, A., Eastmond, C. J., & Ludbrook, A. (2001). Use of discrete choice experiments to elicit preferences. *Quality in Health Care*, 10(Suppl I), i55–i60.
- Ryan, M., Gerard, K., & Amaya-Amaya, M. (2008). *Using discrete choice experiments to value health and health care*. Dordrecht: Springer.
- Ryan, M., Watson, V., Krucien, N., & Heidenreich, S. (2014). *Using discrete choice experiments in health economics: theoretical and practical issues (comp.)*. Aberdeen: Health Economics Research Unit, University of Aberdeen.
- Salani, R. (2013). Survivorship planning in gynecologic cancer patients. *Gynecologic Oncology*, 130(2), 389–397.
- Salkeld, G., Ryan, M., & Short, L. (2000). The veil of experience: Do consumers prefer what they know best. *Health Economics*, 9(3), 267-270.
- Sandsund, C., Towers, R., Thomas, K., Tigue, R., Lalji, A., Fernandes, A., Doyle, N., Jordan, J., Gage, H., & Shaw, C. (2017). Holistic needs assessment and care plans for women with gynaecological

- cancer: Do they improve cancer-specific health-related quality of life? A randomised controlled trial using mixed methods. *BMJ Supportive & Palliative Care*, 0, 1-14.
- Sekse, R. J. T., Raaheim, M., Blaaka, G., & Gjengedal, E. (2010). Life beyond cancer: women's experiences 5 years after treatment for gynaecological cancer. *Scandinavian Journal of Caring Sciences*, 24(4):799–807.
- Sheeran, P., & Orbell, S. (2000). Using implementation intentions to increase attendance for cervical cancer screening. *Health Psychology*, 19(3), 283-289.
- Sheldon, H., Davis, A., & Parsons, S. (2008). *Cancer follow-up care: The views of patients and carers expressed in a range of focus groups*. Oxford: The Picker Institute.
- Shuy, R.W. (2002). In-person versus telephone interviewing. In J.G. Gubrium & J.A. Holstein (Eds.), *Handbook of interview research: Context and method* (pp. 537–555). Thousand Oaks, CA: Sage.
- Shin, D. W., Cho, J., Roter, D. L., Kim, S. Y., Sohn, S. K, Yoon, M.-S., Kim, Y.-W., Cho, B., & Park, J.-H. (2013). Preferences for and experiences of family involvement in cancer treatment decision-making: Patient-caregiver dyads study. *Psycho-Oncology*, 22, 2624-2631.
- Smith, I. E., & Schiavon, G. (2013). Follow-up tests to detect recurrent disease: Patient's reassurance or medical need? *The Breast*, 22(2), S156-S160.
- Song, L., Rini, C., Ellis, K. R., & Northouse, L. L. (2016). Appraisals, perceived dyadic communication, and quality of life over time among couples coping with prostate cancer. *Support Care Cancer*, 24(9), 3757-3765.
- Srivastava, A. & Thomson, S. B. (2009). Framework Analysis: A Qualitative Methodology for Applied Policy Research. *Journal of Administration and Governance*, 4(2), 72-79.
- Stacey, D., Legare, F., Lewis, K., Barry, M. J., Bennett, C. L., Eden, K. B., Holmes-Rovner, M., Llewellyn-Thomas, H., Lyddiatt, A., Thomson, R., & Trevena, L. (2011). Decision aids for people facing health treatment or screening decisions. *Cochrane Database of Systematic Reviews*, 2017(4), CD001431.
- Stafford, L. & Judd, F. (2010). Partners of long-term gynaecological cancer survivors: Psychiatric morbidity, psychosexual outcomes and supportive care needs. *Gynecologic Oncology*, 118, 268-273.
- Steinberg, E., Greenfield, S., Mancher, M., Wolman, D. M., & Graham, R. (2011). *Clinical practice guidelines we can trust*. Institute of Medicine. Washington, DC: National Academies Press.

- Stewart, D. E., Wong, F., Cheung, A. M., Dancey, J., Meana, M., Cameron, J. I., McAndrews, M. P., Bunston, T., Murphy, J., & Rosen, B. (2000). Information needs and decisional preferences among women with ovarian cancer. *Gynecologic Oncology*, 77(3), 357-361.
- Sturges, J.E., & Hanrahan, K.J. (2004). Comparing telephone and face-to-face qualitative interviewing: A research note. *Qualitative Research*, 4, 107–118.
- Sundar, S., Balega, J., Crosbie, E., Drake, A., Edmondson, R., Fotopoulou, C., Gallos, I., Ganesan, R., Gupta, J., Johnson, N., Kitson, S., Mackintosh, M., Martin-Hirsch, P., Miles, T., Rafii, S., Reed, N., Rolland, P., Singh, K., Sivalingam, V., & Walther, A. (2017). *BGCS uterine cancer guidelines: recommendations for practice*. Retrieved from <https://bgcs.org.uk/BGCS%20Endometrial%20Guidelines%202017.pdf>
- Suprasert, P., & Manopunya, M. (2011). Financial burden of gynecologic-cancer survivors associated with attendance in a surveillance program at a tertiary care hospital in Thailand. *Asian Pacific Journal of Cancer Prevention*, 12(7), 1761-1763.
- Sweet, L. (2002). Telephone interviewing: Is it compatible with interpretive phenomenological research? *Contemporary Nurse*, 12, 58–63.
- Tanner-Smith, E., & Brown, T. N. (2010). Evaluating the health belief model: A critical review of studies predicting mammographic and pap screening. *Social Theory & Health*, 8(1), 95-125.
- Tariq, S. & Woodman, J (2010). Using mixed methods in health Research. *JRSM short reports*, 4(6), 2042533313479197.
- The Joanna Briggs Institute (2014). Joanna Briggs Institute Reviewers' Manual: 2014 edition / supplement. The Joanna Briggs Institute: Adelaide, Australia. Retrieved from http://joannabriggs.org/assets/docs/sumari/ReviewersManual_2014-The-Systematic-Review-of-Prevalence-and-Incidence-Data_v2.pdf
- The Kings Fund (2010). How to deliver high-quality, patient-centred, cost-effective care. Consensus solutions from the voluntary sector. Retrieved from <https://www.kingsfund.org.uk/publications/articles/how-deliver-high-quality-patient-centred-cost-effective-care>.
- Thomas, J., Harden, A., Oakley, A., Oliver, S., Sutcliffe, K., Rees, R., Brunton, G., & Kavanagh F. (2004). Integrating Qualitative Research with trials in systematic reviews: an example review from public health shows how integration is possible and some potential benefits. *BMJ*, 328, 1010–1012.
- Thompson, T., Barbour, R. S., & Schwartz, L. (2003a). Advance directives in critical care decision making: a vignette study. *BMJ*, 327, 1011-1015.

- Thompson, T., Barbour, R. S., & Schwartz, L. (2003b). 'Health Professionals' views on advance directives – a qualitative interdisciplinary study. *Palliative Medicine*, 17, 403-409.
- Tong, A., Sainsbury, P., & Craig, J. (2007). Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*, 19(6), 349-357.
- Torgeson, C. J., Gorard, S., Low, G., Aimsword, H., See, B. H., & Wright, K. (2008). *What are the factors that promote high post-16 participation of many minority ethnic groups? A focused review of the UK-based aspirations literature*. London: EPPI-Centre, Social Science Research Unit, Institute of Education, University of London.
- Treweek, S., Lockhart, P., Pitkethly, M., Cook, J. A., Kjeldstrøm, M., Johansen, M., Taskila, T. K., Sullivan, F. M., Wilson, S., Jackson, C., Jones, R., & Mitchell, B. D. (2013). Methods to improve recruitment to randomised controlled trials: Cochrane systematic review and meta-analysis. *BMJ Open*, 3(2), e002360.
- University Hospitals of Leicester NHS trust. (2016). *Patient initiated follow-up of gynaecological cancers*. Leicester.
- University Hospital Southampton NHS foundation trust. (2014). *Your guide to self-managed follow-up after cancer of the womb*. Southampton.
- van Hezewijk, M., Hille, E., Scholten, A., Marijnen, C., Stiggelbout, A., & van de Velde, C. (2011). Professionals' opinion on follow-up in breast cancer patients: perceived purpose and influence of patient' risk factors. *European Journal of Surgical Oncology*, 37(3), 217 – 224.
- Vass, C., Rigby, D., & Payne, K (2017). The Role of qualitative research methods in discrete choice experiments: a systematic review and survey of authors. *Medical decision making*, 37, 298-313.
- Vella, K., Goldfrad, C., Rowan, K., Bion, J., & Black, N. (2000). Use of consensus development to establish national research priorities in critical care. *BMJ*, 320(7240), 976-980.
- Viney, R., Norman, R., Brazier, J., Cronin, P., King, M., Ratcliffe, J., & Street, D. (2014). An Australian discrete choice experiment to value EQ-5D Health States. *Health Economics*, 23(6), 729-742.
- Vistad, I., Cvancarova, M., & Salvesen, H. B. (2012). Follow-up of gynecological cancer patients after treatment- the views of European experts in gynecologic oncology. *Acta Obstetricia et Gynecologica Scandinavica*, 91(11), 1286-1292
- Wales Cancer Network (2016). *Cancer Delivery Plan for Wales 2016-2020: The highest standard of care for everyone with cancer*. Retrieved from www.walescanet.wales.nhs.uk/sitesplus/documents/1113/161114cancerplanen.pdf

- Walker, R. C., Hanson, C. S., Palmer, S. C., Howard, K., Morton, R. L., Marshall, M. R., & Tong, A. (2015). Patient and caregiver perspectives on home hemodialysis: A systematic review. *American Journal of Kidney Diseases*, 65(3), 451-463.
- Wanless D (2002). *Securing our Future Health: Taking a long-term view. Final report*. London: Department of Health.
- Welsh Government and NHS Wales. (2016). Prudent Healthcare Securing Health and Well-being for Future Generations. Retrieved from <http://www.prudenthealthcare.org.uk/wp-content/uploads/2016/02/Securing-Health-and-Wellbeing-for-Future-Generations1.pdf>
- Wennberg, D. E, Marr, A., Lang, L., O'Malley, S., & Bennett, G. (2010). A randomised trial of a helpline care management strategy. *The New England Journal of Medicine*, 363(13), 1245-1255.
- Wenzel, L. B., Donnelly, J. P., Fowler, J. M., Habbal, R., Taylor, T. H., Aziz, N., & Cella, D. (2002). Resilience, reflection, and residual stress in ovarian cancer survivorship: a gynecologic oncology group study. *Psycho-oncology*, 11(2), 142–153.
- Williams, A. (1988). Ethics and efficiency in the provision of health care. *Royal Institute of Philosophy Supplements*, 23, 111-126.
- Williams, A. L., Tisch, A. J. H., Dixon, J., & McCorckle, R. (2013). Factors associated with depressive symptoms in cancer family caregivers of patients receiving chemotherapy. *Supportive Care in Cancer*, 21(9), 2387-2394.
- Wolff, J. L., & Roter, D. L. (2011). Family presence in routine medical visits: A meta-analytical review. *Social science & Medicine*, 72(6), 823-831.
- Wu, L. M., Mohammed, N. E., Winkel, G. & Diefenbach, M. A. (2013). Patient and spouse illness beliefs and quality of life in prostate cancer patients. *Psychology & Health*, 28(4), 355-368.
- Yeo, S. T., Edwards, R. T., Fargher, E. A., Luzio, S. D., Thomas, R. L., & Owens, D. R. (2012). Preferences of people with diabetes for diabetic retinopathy screening: a discrete choice experiment. *Diabetic Medicine*, 29(7), 869–877.
- Zola, P., Fuso, L., Mazzola, S., Piovano, E., Perotto, S., Gadducci, A., Galletto, L., Landoni, F., Maggino, F., Maggino, T., Raspagliesi, F., Satori, E., & Scambia, G. (2007). Could follow-up different modalities play a role in asymptomatic cervical cancer relapses diagnosis? An Italian multicentre retrospective analysis. *Gynecologic Oncology*, 107(1), S150-S154.

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Appendices relating to Thesis as a whole

Appendix 0.1 PhD Studentship grant protocol

Removed for copyright

Appendices relating to Chapter 2, Systematic Literature Review

Appendix 1.1 PROSPERO systematic review protocol

Removed for copyright

Appendix 1.2 Example of search terms (PsycInfo)

	Search Terms (limited to human, include duplicates. No search term for VULVAL, ENDOMETRIAL, ENDOMETRIUM) Conducted on 18.12.14
1	Uterus.su
2	Ovaries.su
3	Cervix.su
4	Gynecology.su
5	(Cervi* OR ovar* OR endometri* OR Uter* OR Gyn*e* OR Vulva*)ti, ab
6	Or/ 1-5
7	Neoplasms.su
8	Oncology.su
9	(Cancer* OR neoplasm* OR malignan* OR tumo*r* OR oncology OR carcinom* OR adenocarcinom*).ti, ab
10	Or/ 7 – 9
11	6 and 10
12	"posttreatment followup".su
13	"continuum of care" .su
14	Aftercare.su
15	recurrence.su
16	Recovery (disorders) .su
17	("follow*up" OR "post*treatment" OR "post*therapy" OR "post*operative" OR "After*care" OR "after*treatment" OR Surveillance OR Survivor* OR "multi*disciplinary" OR "needs assessment" OR "Self*management" OR "remote monitoring" OR "patient*led" OR "risk*stratif*" OR *phone OR internet OR "e*mail" OR Consultant OR Nurse OR GP OR "General practitioner" OR "family doctor" OR "primary care" OR "second*ry care") NOT (Screen* OR HPV)
18	Or/ 12 – 17
19	11 and 18
20	Attitudes.su
21	preferences.su
22	"Consumer satisfaction".su
23	"Patient satisfaction".su
24	"choice behaviour".su
25	"qualitative research".su
26	interviews.su (conjoint* OR "Part-worth utilit*" OR "functional measurement" OR "paired comparison" OR "Pairwise Choice" OR "Binary choice" OR "Stated Preference*" OR "Discrete choice*" OR "Choice experiment*" OR "Decision analysis" OR utilit* OR preference*) (interview* or discussion* or questionnaire* or focus group* or qualitative or ethnograph* or fieldwork or "field work" or "key informant") .ti,ab
29	Or/ 21-28
30	20 and 29

Appendices relating to Chapter 3, Qualitative study

Gynaecological cancer follow-up after treatment care: What attributes do patients and their informal caregivers value

Protocol

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Abstract

There are respectively 8378, 6692, 2659, new cases of endometrial, ovarian and cervical cancer each year in the United Kingdom. Currently there are no NICE guidelines and no consensus regarding the most effective and cost effective follow up after treatment care for endometrial, cervical, and ovarian cancer. The NHS is striving to improve cancer outcomes, ensuring that outcomes are based on patients' and carers' needs. The aim of this research is to identify the factors (described in Health Economics as attributes and attribute levels) that gynaecological cancer follow up patients and their carers value. Findings from this PhD project will influence the design of a discrete choice experiment (a validated preference elicitation method), and survey, with the overarching aim of understanding patients' and their informal caregivers' perspectives and preferences for different models of gynaecological cancer follow-up after treatment care.

Introduction

Endometrial, ovarian and cervical cancer

In the UK, gynaecological cancers such as endometrial (uterus), ovarian and cervical cancer are respectively the fourth, sixth, and tenth most common cancer sites affecting women. There are 8378, 6692, 2659, new cases of endometrial, ovarian, and cervical cancer each year, and the incidence of endometrial cancer is increasing (<http://eco.iarc.fr/>). Of these, endometrial cancer has the highest prevalence, with approximately 38,700 women alive up to ten years after being diagnosed with uterine cancer as compared to ovarian (around 25,100 women), and cervical cancer (around 19,000 women: National Cancer Intelligence Network, 2006). The five year survival rate for patients diagnosed in England and Wales with cervical cancer, ovarian cancer, and endometrial cancer between the year 1971 - 1975 was 52%, 21% and 61% respectively (www.cancerresearchuk.org). This is compared to a 5-year survival rate of 67.4%, 46.3%, and 77.4% in patients diagnosed in England for the period 2007 - 2011 (Office for National Statistics, 2013). This data is not available for the UK as a whole. The World Health Organisation reported that in the UK in 2012, ovarian cancer accounted for more deaths (4,400 deaths) than all the other gynaecological cancers combined (approximately 3,200 deaths), with the mortality rate for endometrial cancer 1,711 deaths per year, and cervical

cancer 979 deaths per year (<http://eco.iarc.fr>). Hospitals in North Wales (Ysbyty Gwynedd, Ysbyty Glan Clwyd, and Ysbyty Maelor) serve a population of 740,000 people. Within these hospitals, there are approximately 70 diagnoses of endometrial cancer per annum, 80 diagnoses of ovarian cancer per annum, and 35 diagnoses of cervical cancer per annum (Leeson et al, 2012).

Cancer follow up needs

A health care need is defined as an ability to benefit from the provision of health care (Morris et al, 2007). Traditionally in the NHS it is considered that whether or not an individual has a health care need should be assessed by a health professional who is considered an 'expert' in need identification. However, experts opinions of what defines a health need may differ (Morris et al, 2007). Additionally, there is a growing emphasis that patients should be involved in the identification and discussion of their holistic needs (NICE, 2005). Therefore there is a shift in health care with the dominant view that the identification of needs are patient led and hence subjective and variable (Tamburini et al, 2000; Boberg et al., 2003). Unmet needs may arise when patients express that they have a need relating to their cancer, but do not feel this need is met by the health care services they are provided with.

There are some cancer follow up needs that are specific to the cancer type and treatment type whilst others are generic. Research shows that at follow up, across cancer sites and treatments, approximately 500,000 cancer follow up care patients in the UK are experiencing disability or poor health as a result of their cancer or cancer treatment (Macmillan Cancer Care, 2013). Examples of the biopsychosocial long term consequences of cancer and cancer treatment faced by patients include lymphedema (fluid retention which causes persistent swelling of tissue), pain, chronic fatigue, urinary and gastrointestinal problems, anxiety, depression and sexual problems (Adams et al, 2014; Andreyev, Davidson, Gillespie, Allu & Swarbrick, 2012; Prue, Rankin, Allen, Gracey & Cramp, 2006; van den Beuken-van Everdingen et al, 2007; Macmillan Cancer Care, 2013). Greer et al (2011) noted that individuals with a cancer history were more likely in the last twelve months than healthy controls, to be diagnosed with an anxiety disorder and have taken medication prescribed by a psychiatrist. Cancer and cancer treatment effects can last over a decade post treatment (Adams et al.,

2014), and may have a wider impact on an individual's life, for example, financially through an inability to work (Macmillan Cancer Care, 2013).

Radiotherapy and surgery in the pelvic area may result in parts of the bowel sticking together. For 12% of women who receive radiotherapy, this results in bowel incontinence, and for 38% of women results in urinary incontinence lasting up to 11 years post treatment (Adams et al, 2014). However, a local survey by Hull Gynaecological cancer care services highlighted that the prevalence of bowel and bladder changes is significantly higher, and that in the three months following treatment, 84% of patients suffered from bowel and/or bladder changes (Holmes, 2012). [Adams et al \(2014\) reported that](#) 24% of women treated by radiotherapy to the pelvic area say that they have sexual difficulties for at least 11 years post treatment. Treatment for gynaecological cancer may also result in loss of fertility, hot flushes, difficulties with body image and secondary cancer (Ng & Travis, 2008; Maddams, Parkin & Darby, 2011). Therefore, gynaecological cancer and its treatment may have a large impact on quality of life (QoL: Nama et al, 2013), and place a range of challenges in front of patients and the health and social care services they have access to, in order to meet any perceived unmet need.

Follow up care

The aim of follow up care is to enable the management of patients' physical and psychological morbidity which may allow detection of possible recurrent disease (The Royal Society of Medicine, 1999). Whilst follow-up after treatment for women with gynaecological cancer is routine, it is not evidence based. To date, there are no NICE recommendations as to what form or frequency of follow up after care is appropriate for survivors with gynaecological cancer, nor is the cost to the NHS weighed up against the benefits of other follow up care approaches. The Royal Society of Medicine (1999), claimed that there is no consensus as to what form or frequency of follow-up is appropriate or effective. A UK study found that 85% of the consultant gynaecologists surveyed (584/684) used 106 different follow-up protocols and the remaining 15% reported no routine follow-up (Kerr-Wilson and McCrum, 1995). Leeson et al (2013), conducted a UK nationwide survey of current practice in gynaecological cancer follow-up and reported similar findings. It is therefore unknown what follow up after care provision is most effective, good value for money to the NHS, and provides equitable

access. Furthermore it is unknown whether follow up care meets the needs of gynaecology cancer patients themselves.

Traditional models of follow up

The traditional model of follow up care is led by the consultant and requires multiple visits to hospital and biological tests to identify possible cancer reoccurrence. The frequency of follow up appointments under this model depends on the hospital protocol (Leeson et al, 2013).

Reoccurrence is often symptomatic and patients often present with symptoms between scheduled follow up appointments (Vistad et al, 2011). This would trigger a referral for further investigation (Leeson et al, 2012). Additionally, it is unlikely that a patient will be diagnosed with reoccurrence based on test results alone (Nama, Nordin & Bryant, 2013). The efficiency of this model of after care is therefore questioned. This model of gynaecological cancer follow up care has an average cost of £127 per unit of follow up care, with 86,072 units of follow up care provided each year in England. However, the cost varies greatly with a minimum cost of £48 per unit compared to a maximum cost of £130 per unit. The reasons for the difference in unit cost are unknown (Department of Health, 2013). It was estimated based on the 2010 – 2011 NHS reference costs, current survival and current follow up protocols, that the cost of gynaecological cancer follow up care in Wales exceeds one million pounds per annum (Leeson et al, 2012). However, the average cost of follow up care has increased from £118 per unit to £127 per unit in the period 2012 - 2013, and the rate of survival is steadily increasing so this figure is likely to be higher in 2014 (Department of Health, 2013).

The traditional model of follow up after treatment care can lead to high levels of anxiety amongst patients (Kew, Galaal, & Maderville, 2009) and patients have asked for alternative models of follow up after treatment care (Lydon, Beavery, Newbury, & Wray, 2009).

Alternative models of follow up care

There are various alternative forms of follow up after care including specialist nurse led follow up, telephone follow up, GP led follow up, and self-management. The direct cost of follow up provided by a consultant or other health professional is provided by the Department of Health (2013), in the document, 'NHS reference costs 2012 - 2013' (please see table 1). However, whilst these costing's are provided, an explanation of why follow up care delivered by a health

care professional other than the consultant is more costly to the NHS is not given. Additionally, the publication of NHS reference costs for 2010 – 2011, reported that non-consultant led, non face to face follow up cost £10 per unit, with non-consultant led, not face to face, multi-professional follow up costing £93 a unit (Department of Health, 2011). Why these costs have increased is unknown. The exact costs and benefits of each of the GP led and self-management models of follow up care are unknown.

Table 1. NHS reference costs 2012 - 2013 (Department of Health, 2013).

	Consultant led (per unit)	Non consultant led (per unit)
Face to face Not admitted	£127	£162
Face to face Not admitted Delivered as part of multi- disciplinary team	£132	£997
Not face to face Not admitted	£31	£94
Not face to face Not admitted Delivered as part of a multi- disciplinary team	£80	unknown

The National Cancer Survivorship Initiative (2013a) recommends a stratified approach to cancer follow up care (see Figure 1), as opposed to the current traditional pathway which adopts a one size fits all approach (Macmillan Cancer Support, 2013). It is argued that by using a stratified patient centred approach, resources can be released and reinvested back into ‘survivorship pathways’ in order to improve patients’ outcomes and reduce the cancer burden on patients, carers, and their families (National Health Service Improvement, 2013). It has been suggested that patients with no cancer or cancer related morbidities could be assigned a self-management follow up pathway, whilst patients with non-cancer related morbidities could be assigned to a GP led follow up pathway, and those with cancer related morbidities could receive increased support from the cancer team (National Health Service Improvement, 2013).

A self-management approach to follow up care is based on 'the recovery package'. The recovery package is comprised of a holistic needs assessment to be completed with patients and members of the multi-disciplinary team, with a care plan and onward referrals to meet the identified needs; a treatment summary; a six week care plan review with the individual's GP to discuss long term effects; a health and well-being clinic to increase patients' and carers' confidence and self-efficacy in self-management. Self-efficacy refers to the individuals' belief in their ability to carry out the behaviour: Bandura, 1977). The health and wellbeing clinic should include education about signs and symptoms, resources available to the individual and family, lifestyle advice, work rehabilitation, and support in accessing appropriate programmes (Macmillan Cancer Support, 2013; The National Cancer Survivorship Initiative, 2013). It is reported using a sample of breast and lung cancer patients, that using a stratified approach for the whole cancer journey could, once all inpatient, outpatient, and emergency costs are considered, improve the efficiency, care, and management of co-morbidities. This would release a saving of 10% of the current NHS costs for patients suitable for a less intensive pathway of care, as well making a saving to the wider economy through increased tax contributions and saved benefit payments (Allirajah, 2010; National Cancer Survivorship Initiative, 2013). Even so, there is no current research into the suitability, effectiveness and cost effectiveness of the stratified model in gynaecological cancer care. Therefore the suitability of this model for gynaecological cancer patients would have to be modelled, as has been done with lung and breast cancer patients (Allirajah, 2010).

Hull Gynaecological cancer care services modified their follow up after treatment pathway after results from a survey indicated that 84% of participants experienced significant bowel or bladder changes in the three months following treatment. Patients in Hull are now given a holistic needs assessment of their biopsychosocial needs (for example, assessing the impact of the cancer and any late treatment effects on psychological wellbeing or social functioning). Pathways have been developed with other services to help meet the needs identified. Patients are also educated about possible signs of cancer reoccurrence. Whilst this model has been discussed there is no evidence as yet regarding its effectiveness and cost effectiveness (Holmes, 2012).

The NHS is prioritising the improvement of outcomes from chronic diseases, including cancer (Department of Health, 2012). This priority is reiterated by Macmillan Cancer Support

(2013), who argue that the development of new pathways and follow up after care models to address the consequences of cancer and cancer treatment, and undertaking cost analyses of these models is a priority, particularly in pelvic cancers (Macmillan Cancer Care, 2013).

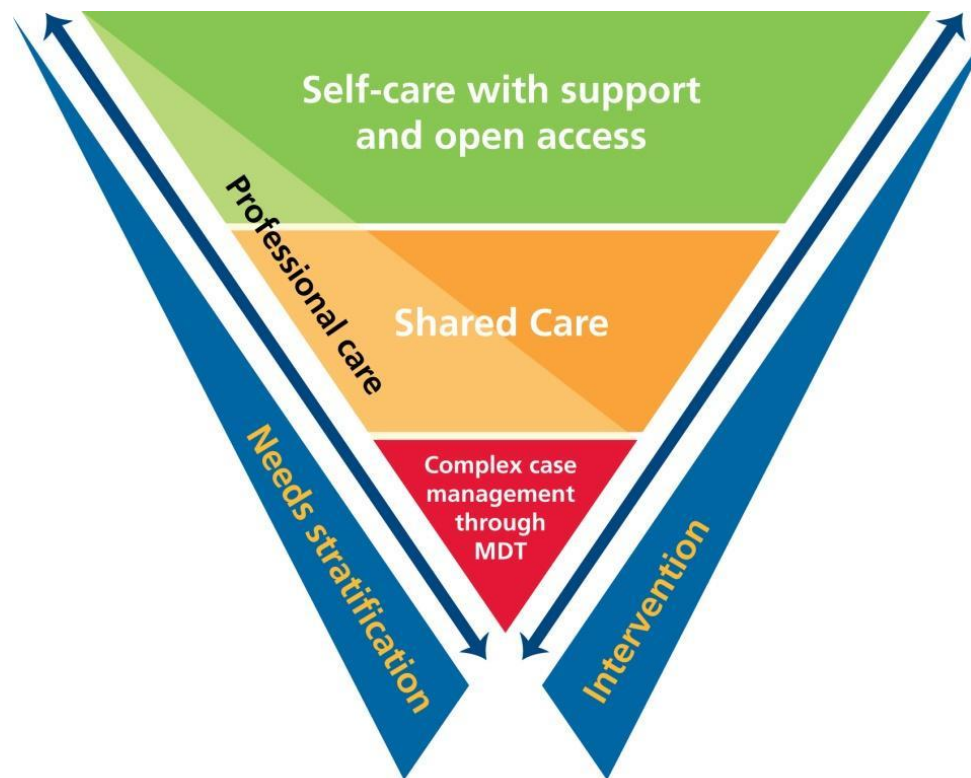


Figure 1. Stratified approach to follow up care (National Cancer Survivorship Initiative, 2013b)

Patient Centred Services

The NHS's modernisation plan (Department of Health, 1997) and the new Welsh cancer plan 2012 (Welsh Government, 2012) highlight the importance of designing a patient-centred service around the needs and aspirations of service users. Additionally, the International Society for Pharmacoeconomics and Outcome research (2011) states that:

"The perspective on outcomes should be all direct health effects, whether for patients or, when relevant, other people (principally carers)"

However, there is no published information relating to the needs and aspirations for a preferred model of follow up care in women with gynaecological cancer and their informal carers. It can therefore be seen that in order for the NHS to meet patients' follow up needs,

the biopsychosocial needs and aspirations of endometrial, ovarian and cervical cancer patients need to be better understood, from a patient not the health professional perspective. Identifying patients' and their informal caregivers' perspectives and preferences for patient identified models (attributes and attribute levels) of gynaecological cancer follow-up services would undoubtedly facilitate future health service reconfiguration, whilst perhaps reducing inefficiencies in the provision of healthcare services within limited NHS resources, and improving equity of access. Following identification of attributes of follow up care that patients have a preference and high utility for, a multi-centred randomised controlled trial and economic evaluation can be conducted, comparing the clinical effectiveness and cost-effectiveness of the traditional model of gynaecological cancer follow up care to the patient preferred model. However, it is acknowledged that this would require significant powering, therefore other research designs would also need to be explored.

Health Economics

Health Economics is the study of how scarce resources can be allocated when needs are infinite (Morris, Delvin, & Parkin, 2007). The aim of health economics is to increase the efficiency and equity of health care resource provision, whilst acknowledging that this is a trade-off. Therefore adopting a health economics perspective in this research can aid understanding of how to improve the efficiency of gynaecological cancer follow up care, whilst exploring equity applications and remaining patient centred, focused on the needs of patients as valued by patients and caregivers themselves.

It has been recommended that NICE use discrete choice experiments as a way of identifying patient preferences (Ryan, 2004). Discrete choice experiments are embedded in welfare economics. In this stated preference elicitation method it is assumed that individuals are rational decision makers who make decisions based on, and aiming to maximise their innate stable preferences (Consumer Choice Theory). In accordance with neoclassical consumer theory, individuals will have a preference and hence make a choice for the bundle off attributes that has the maximum benefit for their utility, taking into consideration their budget. Individuals derive their utility, and hence preference, from the collection of attributes opposed to the overall commodity (Lancaster's Economic Theory of Value: Lancaster, 1966). Therefore, individuals will make a preference and choice for the bundle of attributes that has

the highest overall utility (benefit) for them. If any of the attribute levels that make up this bundle changes, it may cause a discrete shift in a preference from one set of attributes to another set of attributes. By asking participants to make a choice between a set of attributes it provides information on individuals' relative preferences for each attribute (Ryan et al, 2008). Whilst discrete choice experiments have not been previously used to identify patients' and carers' preferences for gynaecological cancer follow up care, they have been used to identify the preferences for attributes of follow up care in breast cancer patients (Kinman, Dellaert, Boersma, Lambin & Dirksen, 2010; Simba, 2008). Results showed that health care provider, mode of contact, frequency of visits and waiting time were all important attributes of breast cancer follow up care as valued by patients. However, provision of an educational program compared to no educational program was not significant. Follow up care delivered by a consultant was preferred to follow up care delivered by a specialist nurse or GP, but follow up care delivered alternatively by a consultant and specialist nurse was also acceptable to the participants. Participants had a strong preference for face to face opposed to telephone appointments, and preferred a reduced waiting time and an increased frequency of appointments (Kinman et al, 2010). However, Simba (2008) found that patients had a preference for follow up care delivered twice a year over five years by a surgeon in a face to face format, however, follow up over 3 years, face to face by a specialist nurse was also acceptable to patients.

Aims and Objectives

In light of the above, the aim of this PhD (3 phases of research) is to identify gynaecological cancer survivors' and their informal caregivers' perspectives and preferences for different models of follow up care. The aim of the focus group (phase one) is to identify the attributes of gynaecological cancer follow up care that patients and their informal caregivers value most. The attributes identified will, together with the systematic literature review form the design of the discrete choice experiment (phase two), and the design of the survey (phase three). The specific aims of the focus group are to:

7. Develop an understanding of the attributes of gynaecological cancer follow up care that are considered important/not important by patients and their informal caregivers.

8. Develop an understanding of why these attributes are valued as they are.
9. Identify attribute levels for the identified attributes. For example, if the attribute 'health care professional that delivers follow up care' is valued, the attribute levels that might be identified are: the consultant, specialist nurse, and GP.
10. Identify and understand hypothetical willingness to pay thresholds for alternative models of cancer follow up care.
11. Identify and understand how long patients and carers would be willing to wait between follow up care appointments.
12. Develop an understanding of how patients and informal caregivers feel that follow up after treatment care could be improved.

Ethical approval is being sought for phase one of the research (the focus groups). Ethical approval will be sought for phase two and phase three of the research (the discrete choice experiment and survey) at a later date.

Method

Patient involvement

Patient representatives will be identified through follow up clinics within Betsi Cadwaladr University Health Board. Patient representatives will not be a participant in the research study. The aim of patient involvement is to ensure that the research in phase one, phase two, and phase three is relevant to the patient population, and not based on the researchers' conceptualisation of what is important to patients (INVOLVE, 2012). It is intended that there will be at least two patient representatives for this research study.

Design

The three phase body of research is a mixed method study, whereby results from the cross-sectional qualitative focus group will together with findings from the systematic review and in collaboration with health professionals, influence the design of the discrete choice experiment (phase two) and mixed method survey (phase three). Focus groups are becoming increasingly used to identify attributes and attribute levels for the design of a discrete choice

experiment (Ryan, Gerard & Amaya-Amaya, 2008). Ethical approval is being sought for phase one of the research (the focus groups). Ethical approval will be sought for phase two and phase three of the research (the discrete choice experiment and survey) at a later date.

As a guide there will be four focus groups, two for patients and two for their carers, in accordance with focus group sample size guidance. In accordance with this guidance, if data saturation is not reached further focus groups will be conducted (Straus & Corbin, 1990).

Participants

Sixteen women (and their informal caregivers) who are currently undergoing follow up after treatment care for endometrial, ovarian or cervical cancer within Betsi Cadwaladr University Health Board will be invited to take part in four focus groups. Patients will not be excluded if they do not provide contact details for a carer. Health care professionals in the respective outpatient wards will identify, screen, and give the recruitment pack (patient and carer **invite letter, information sheet, consent form, demographic questionnaire and request for more information**), to patients at or after their follow up appointment.

Inclusion criteria

- Patients undergoing follow up after treatment care for endometrial, ovarian, or cervical cancer, or an informal caregiver for this patient.
- The patient (or patient for whom the carer is a caregiver) has had at least one follow up after care appointment.
- The patient (or patient for whom the carer is a caregiver) was free from disease at their last follow up after care appointment.
- The patient (or patient for whom the carer is a caregiver) is receiving follow up after treatment care with Betsi Cadwaladr University Health Board.
- The participant is aged 18 years or over.
- The participant can speak English.
- The participant has the capacity to consent.

Exclusion criteria

- Patients and carers will be excluded if the patient is part of a clinical trial
- Patients who are cognitively impaired (for example, dementia), have a serious psychiatric condition or a serious developmental or learning difficulty, as advised by the health care professional
- Carers who are cognitively impaired (for example, dementia), have a serious psychiatric condition or a serious developmental or learning difficulty, as advised by the carer themselves
- To ensure that views of patients with different tumour types and stages are represented in the sample, the researcher will monitor recruitment. For example, if when 50% of participants have been recruited, the sample is made up of over 75% of a specific disease type and stage, subsequent recruitment will focus on other tumour types and stages.

Measures

Participants will be asked to complete a **demographic questionnaire** which includes questions about the individual's cancer diagnosis, treatment, and follow up care. The focus group will follow the structure of the **focus group schedule**, which consists of three different focused tasks. First, participants will be asked to list all aspects of follow up care that are important to them on the '**discussion group questions**', and then rank these in order of importance. Following this, as a group, participants will rank the perceived order of importance of these attributes. Participants will be asked to do this individually to ensure data saturation, and as a group to explore participants thought processes and reasoning behind the ranking. Sub questions will be used to elicit explanation of why the attributes are ranked as they are, and to identify suitable levels for each of these attributes. Participants will then be asked (1) How often would you like between follow up appointments? (2) What was good about the follow up care you received in North Wales? (3) Which of your needs are not met by follow up care in North Wales (4) What aspects of follow up care do you think could be improved, explaining why (5) Hypothetically, if you had to pay for follow up after treatment care, how much would you be willing to pay? Based on participants' responses, sub questions will be asked to further understand participants' valuations for different attributes.

Materials

All materials will be translated to Welsh following ethical approval.

For Participants

Participants will be given a **participant information sheet** which outlines the aim of the study, what is required of their participation, any foreseeable risks to taking part, as well as outlining their right to withdraw, the use and storage of their data and contact details for the researcher (see *ethical considerations*). Participants will be asked to complete a **consent form**, where they will be asked to sign that they consent to take part in the study and acknowledge the ethical considerations discussed (see *ethical considerations*). If patients and/or carers would like more information before consenting to take part in the study, they are asked to telephone the researcher or complete and return the **request for more information**. The **debrief form** will again explain the aim of the study, how the participants' data will be used, contact details for the chief investigator, and contact details for the Tenovus Cancer Support line and North Wales Cancer Network Patient Forum, in case the individual would like to discuss anything regarding their cancer needs following involvement in the study. An audio recording device will be used to record the focus groups.

For Health Care Professionals

Health Care Professional's will record the names of patients identified, screened, given recruitment documents to, and consented (see **Eligibility Screening**). Only health care professional's recruiting participants within the outpatient ward will view this document. Patient's names will be recorded as a way of ensuring that a patient is not screened twice. Only the numbers of patients screened, eligible to take part, and given the recruitment pack will be given to the Chief Investigator (see **screening, eligibility and recruitment pack record**).

For Chief Investigator

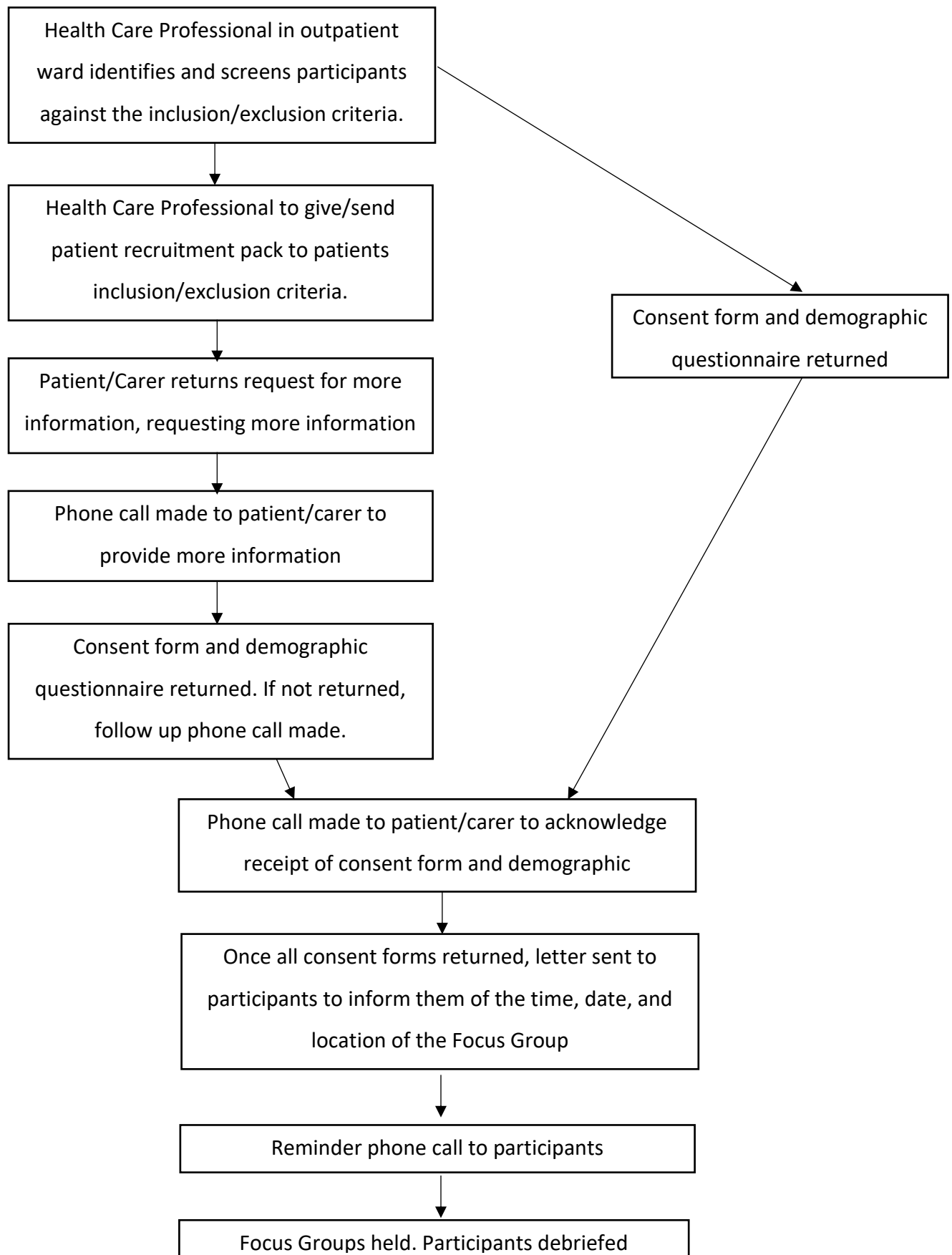
When the researcher has contact with the potential participant or participant, they will complete the **contact form**. Each individual who makes contact with the Chief Investigator will have their own **case report form**. This is to be completed following every contact, using the information from the **contact form**.

Procedure

Once ethical and governance approval is granted from the School of Psychology, Bangor University, and the National Health Service research ethics committee and research and development forum, recruitment will commence (May 2014). There will be a phased approach to recruitment (see Figure 2). In the first phase Health Care Professionals in the respective outpatient wards, at/after the patients follow up appointment, will identify and screen participants against the eligibility criteria (see **Screening, Eligibility and Recruitment Record**). The **recruitment pack** will then be given to patients in person or sent to them in the post. The recruitment pack contains the **invite letter, participant information sheet, consent form, demographic questionnaire, request for more information** and FREEPOST envelope for patients and their carer. The carer recruitment pack will be in a separate envelope, and each of the documents will be on a different coloured piece of paper to avoid over burdening patients and carers. Patients are prompted to contact the researcher if they have any questions before consenting, and can do this by telephoning the researcher on the contact details provided, or by returning the **request for more information**. If the patient/carer returns the **request for more information**, the researcher will make a phone call to the patient/carer to explain the research study further and to answer any questions the individual may have. If the patient/carer has expressed an interest in the research study but does not return a **consent form** and **demographic questionnaire**, a follow up telephone call will be made a couple of weeks later if there is an insufficient sample size. Once the documents have been returned the researcher will make a telephone call to the participant to acknowledge receipt of the documents, build up a rapport with the participant, and answer any questions they may have. Once sufficient consent forms have been returned the researcher will send an **arrangement letter** to participants, with the time, date and location of the focus group. A phone call will be made to participants a couple of days before the focus group as a reminder (see Figure 2).

The optimum number of participants in each focus group is between five and eight (Krueger & Casey, 2009). To allow for participant drop out, eight patients will be recruited to each patient focus group, and their respective informal caregivers will be recruited to the respective caregivers' focus group. Good practice guidelines for the conducting of focus groups will be followed (Krueger & Casey, 2009).

Figure 2. Procedure



Focus groups will be held on the Tenovus mobile unit, which will be parked in a suitable location. The Tenovus mobile unit is a large mobile van with comfortable seating, creating a relaxed atmosphere. Refreshments will be provided in the focus groups. Separate focus groups will be held for patients and informal caregivers as they will have different experiences of follow up after care. In addition, the dynamics of interpersonal relationships can mean that discussion is dominated by one person in the relationship whilst the other remains silent, even if the participant who is silent disagrees with what is said. There is also an effect of gender (the 'peacock effect') in mixed gender focus groups (Krueger & Casey, 2009).

Upon arrival at the focus group participants will be welcomed and introduced to each other, before refreshments are provided and ground rules discussed. The **consent form** will then be completed again before participants are given the opportunity to ask further questions. The focus group will then commence. An observational researcher will be present at the focus group to support the researcher. Following completion of the focus group participants will be thanked for their time, given a verbal and **written debrief**, and given the opportunity to ask any questions.

Ethical considerations

At the beginning of the focus group the researcher will discuss the right for everyone to be treated with respect, with differences between individuals respected. Participants will be informed that if they find anything distressing they can have a break if they wish, and will be asked before returning to the group if they wish to continue. Two researchers and another health professional will conduct the focus groups together to manage this potential risk. As stated in the inclusion/exclusion criteria, potential participants will be excluded if they do not have the capacity to consent. The focus groups will be held in the Tenovus mobile unit with Tenovus staff members present. The researcher will ensure that they do not give advice to participants, but will direct any requests for advice to the Tenovus staff member (to be discussed after the focus group). The Tenovus member of staff will be able to make a referral for further Tenovus support if it is felt this is needed (for example, counselling or welfare benefits advice). Whilst a Tenovus member of staff will be on hand to deal with any distress caused by participating and to answer questions more broadly, they will not have access to the data. The participants' GP will be informed of their participation in the research study.

The patient and carer **information sheet** contains detailed information about the research study; its aims; methods; confidentiality; anonymity; the right to withdraw without adverse consequences; the right to decline answering questions; the right to have data supplied withdrawn up to the point of analysis; data storage; the duration of participant involvement; details of perceived risks and benefits; the name and contact details of the principal investigator; the investigator's supervisors and heads of department in the case that the participant has concerns or complaints; and information about how the data will be used; and how the research study findings will be made available to participants. Participants are given the time to consider the information discussed, prompted to discuss participation in the research study with others if they wish, and prompted to contact the researcher to ask questions before consenting if they wish.

The **consent form** contains the contact details for the researcher and a series of explicit statements requiring the participants to indicate by writing their initials in the appropriate box, if they consent to each statement. Participants are asked to confirm that they have had read and understood the participant information sheet, and have had the time to ask any questions they may have. Participants are also asked to indicate that they agree to the focus group being audio recorded; that they understand that their participation is voluntary; understand their right to withdraw without adverse consequences; that they consent to their GP being informed of their participation; that they consent to Tenovus being given their contact details so that Tenovus can get in contact to see if they can offer them any support services to help them; that participation is confidential unless they disclose harm to an individual; they understand data is anonymous and retained securely in accordance with the Data Protection Act; understand that data may be audited by the NHS or Bangor University; agree to the researcher using direct anonymous quotes; and consent to taking part in the study. There will be two copies of each consent form, one to be kept by the participant and one to be stored by the researcher. The consent form will be completed again before the focus group commences, to ensure that participants are still willing and able to consent.

Analysis

Demographic data will be analysed to explore the characteristics of the sample. Focus group recordings will be transcribed then analysed, to identify the attributes of gynaecological cancer follow up care that participant's value, and identify suitable levels for these attributes. Transcripts will be verified by a second member of the research team. A framework approach will be used as the objectives of the research are pre-set (to inform the design of the discrete choice experiment, which aims to address health policy questions). Therefore, whilst the analysis will be inductive (key themes will be identified from participants' data), the analysis will begin deductively, based on the aims and objectives of the research. There are five stages of data analysis using the framework approach: familiarisation; identifying a thematic framework; indexing; charting; mapping and interpretation (Kruger, 1994; Richie & Spencer, 1994; Rabiee, 2004). The final list of attributes and attribute levels identified from each focus group will be sent to two members of the respective focus groups for verification.

Attributes and attribute levels identified will, together with findings from the systematic review and discussion with health professionals and the patient representatives, influence the design of the discrete choice experiment (phase two) and survey (phase three). It is likely that the following attributes will be identified from the focus group study: professional who delivers the follow up care; location of follow up care; frequency of follow up appointments; content of follow up care. The discrete choice experiment will aim to compare the relative importance of the identified attributes and attribute levels.

Confidentiality and data security

Once patients and carers have consented to participate in the focus group they will be assigned a unique study identification number. This number will be used to identify the participant throughout the study. In the focus group participants will be known by their first name only.

Non-anonymous data will be stored in a locked filing cabinet within the Centre for Health Economics and Medicines Evaluation at Bangor University and on an encrypted password protected laptop that is property of Bangor University. The Chief Investigator will hold the key to the filing cabinet. The chief investigator, Laura Budd, and her supervisors Professor Rhiannon Tudor Edwards and Dr Valerie Morrison are the only researchers who will

have access to the non-anonymous data. Anonymous data will be stored in a separate locked filing cabinet within the Centre for Health Economics and Medicines Evaluation at Bangor University, and on an encrypted password protected laptop that is property of Bangor University. The Chief Investigator will keep the key to this filing cabinet. The chief investigators and her supervisory team will have access to this data. Whilst a Tenovus staff member will be present during the focus groups to give broad advice, the Tenovus staff member will not have access to the data provided. With participants' permission, the researcher will provide Tenovus with the participants contact details, so that Tenovus can make contact with the participant to see if they can provide them with any support. Study documentation following completion of the research and PhD will be archived within the Centre for Health Economics and Medicines Evaluation, Bangor University, in accordance with the school's policy.

Dissemination

The study and its results will be published in a peer reviewed academic journal, and be presented at the Tenovus conference and the Welsh Health Economic Group Conference.

References

Adams, E., Boulton, M., Horne, A., Rose, P. W., Durrant, L., Collingwood, M., Ozkrochi, R., Davidson, S. E., & Watson, E. (2014). The effects of pelvic radiotherapy on cancer survivors: Symptom profile, psychological morbidity and quality of life. *Clinical Oncology*, 26, 10 – 17. doi: <http://dx.doi.org/10.1016/j.clon.2013.08.003>

Allirajah, D. (2010). *Demonstrating the Economic Value of Co-ordinated Cancer Services*. Retrieved from <http://www.macmillan.org.uk/Documents/AboutUs/Commissioners/MonitorResearchinManchesterbriefing.pdf>

Andreyev, H. J. N., Davidson, S. E., Gillespie, C., Allum, W. H., & Swarbrick, E. (2012). Practice guidance on the management of acute and chronic gastrointestinal problems arising as a result of treatment for cancer. *Gut*, 61, 179 – 192.

Bandura, A. (1977). Self-efficacy: Toward a unifying theory of behavioral change. *Psychological Review*, 84, 191-215. doi: [10.1037/0033-295X.84.2.191](https://doi.org/10.1037/0033-295X.84.2.191)

Boberg, E. W., Gustafon, D. H., Hawkins, R. P., Offord, K. P., Koch, C., Wen, K.-Y., Kreutz, K., & Salner, A. (2003). Assessing the unmet information, support and care delivery needs of men with prostate cancer. *Patient Education and Counselling*, 49, 233 - 242.

Department of Health. (1997). *The new NHS: Modern, Dependable*. Retrieved from, <http://www.archive.official-documents.co.uk/document/doh/newnhs/contents.htm>.

Department of Health. (2011). *NHS reference costs 2010 to 2011*. Retrieved from, <https://www.gov.uk/government/publications/nhs-reference-costs-2012-to-2013>

Department of Health (2012). The NHS Outcomes Framework 2013-2014. https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/213055/121109-NHS-Outcomes-Framework-2013-14.pdf

Department of Health (2013). *NHS reference Costs 2012 to 2013*. Retrieved from <https://www.gov.uk/government/publications/nhs-reference-costs-2012-to-2013>

Greer, J. A., Solis, J. M., Temel, J. S., Lennes, I. T., Prigerson, H. G., Maciejewski, P. K., Pirl, W. F. (2011). Anxiety disorders in long-term survivors of adult cancers. *Psychosomatics*, 52, 417-423.

Holmes, L. (2012) *Addressing the consequences of treatment in Gynaecological cancer patients. Care Integration Awards*. Retrieved from <http://www.ncsi.org.uk/wp-content/uploads/Article1-doc-final-version-Gynae-oncology-CNS-Lynn-Holmes.pdf>

International Society for Pharmacoeconomics and Outcome research. (2011). *Pharmacoeconomic Guidelines around the World*. Retrieved from <http://www.ispor.org/peguidelines/countrydet.asp?c=23&t=2>

INVOLVE. (2012). *Briefing notes for researchers: involving the public in NHS, public health and social care research*. INVOLVE, Eastleigh.

Kerr-Wilson, R. H., & McCrum, A. (1995). Follow-up of patients with gynaecological cancer. *Australian and New Zealand Journal of Obstetrics and Gynaecology*, 35, 298-299.

Kew, F. M., Galaal, K., Maderville, H. (2009). Patients' views of follow-up after treatment for gynaecological cancer. *Journal of Obstetrics and Gynaecology*, 29, 135 – 142.

Kimman, M. L., Dellaert, B., Boersma, L. J., Lambin, P., & Dirksen, C. D. (2010). Follow-up after treatment for breast cancer: one strategy fits all? An investigation of patient preferences using a discrete choice experiment. *Acta Oncologica*, 49, 328 – 337.

Krueger, R. A., & Casey, M. A. (2009). *Focus Groups: A practical guide for applied research* (4th ed.). Sage: London

Lancaster, K. (1996). A new approach to consumer theory. *Journal of Political Economy*, 74, 132 – 157.

Leeson, S., Jones, J., Stuart, N. A., Whitaker, R., Edwards, R. T., Hall, L., Neal, R., Sylvestre, Y., Yeo, S. T., Morrison, V. (2012). Research for Patient and Public Benefit Wales: A trial of optimal personalised care after treatment for gynaecological cancer (TOPCAT –G). NISCHR, Welsh Government.

Leeson, S., Stuart, N., Sylvestre, Y., Hall, L., & Whitaker, R. (2013). Gynaecological cancer follow-up audit of current practice: national survey of current practice in the UK. *British Medical Journal open*, 3, e002859. doi:10.1136/bmjopen-2013-002859.

Lydon, A., Beavery, K., Newbury, C., Wray, J. (2009). Routine follow-up after treatment for ovarian

cancer in the United Kingdom (UK): patient and health professional views. *European Journal of Oncology Nursing*, 13, 336 – 343. doi: 10.1016/j.ejon.2009.04.007

Macmillan Cancer Support (2013b). Throwing light at the consequences of cancer and its treatment. Retrieved from <http://www.macmillan.org.uk/Documents/AboutUs/Research/Researchandevaluationreports/Throwinglightontheconsequencesofcanceranditstreatment.pdf>

Maddams, J., Parkin, D. M., & Darby, S. (2011). The cancer burden in the United Kingdom in 2007 due to radiotherapy. *International Journal of Cancer*, 129, 2885-2893.

Morris, S., Delvin, N., & Parkin, D. (2007). *Economic analysis in Health Care*. John Wiley & Sons: Chichester, West Sussex.

Nama, V., Nordin, A., Bryant, A. (2013). Patient related outcome measures for follow-up after gynaecological cancer treatment (Protocol). *The Cochrane collaboration*. Doi: 10.1002/14651858.CD010299.

National Cancer Intelligence Network. (2006). *One, five, and ten year cancer prevalence by cancer network, UK, 2006*. Retrieved from www.ncin.org.uk/view?rid=76

National Cancer Survivorship Initiative (2013a). *Living With and Beyond Cancer: Taking Action to Improve Outcomes*. Retrieved from <http://www.ncsi.org.uk/wp-content/uploads/howtoguide.pdf>

National Cancer Survivorship Initiative (2013b). Stratification process. Retrieved from <http://www.ncsi.org.uk/what-we-are-doing/risk-stratified-pathways-of-care/risk-stratification/>

National Health Service Improvement (2013). *Innovation to implementation: Stratified pathways of care for people living with or beyond cancer, a 'how to guide'*. Retrieved from <http://www.ncsi.org.uk/wp-content/uploads/howtoguide.pdf>

National Institute of Clinical Excellence (2005). *Supportive and Palliative Care: The Manual*. Retrieved from, <http://www.nice.org.uk/nicemedia/pdf/csgspmanual.pdf>. Ng, A. K., & Travis, L. B. (2008). Subsequent neoplasms in cancer survivors. *The Cancer Journal*, 14, 429 – 434.

Office for National Statistics (2013). *Cancer Survival in England: patients diagnosed 2007 – 2011 and followed up to 2012*. Retrieved from, <http://www.ons.gov.uk/ons/rel/cancer-unit/cancer-survival/cancer-survival-in-england--patients-diagnosed-2007-2011-and-followed-up-to-2012/stb-cancer-survival-in-england--patients-diagnosed-2007-2011-and-followed-up-to-2012.html>

Pope, C., Ziebland, S., & Mays, N. (2000). Qualitative research in health care: Analysing qualitative data. *British Medical Journal*, 320, 114-116.

Prue, G., Rankin, J., Allen, J., Gracey, J., Cramp, F. (2006). Cancer-related fatigue: A critical appraisal. *European Journal of Cancer*, 42, 846 – 863.

Rabiee, F. (2004). Focus Group Interview and Data Analysis. *Proceedings of the Nutrition Society*, 63, 655 – 660. doi:10.1079/PNS2004399.

Ryan, M. (2004). Discrete Choice Experiments in Health Care. *British Medical Journal*, 328, 360-361.

Ryan, M., Gerard, K., Amaya – Amaya, M. (2008). *Using Discrete Choice Experiments to Value Health and Health Care*. Springer: Dordrecht, The Netherlands.

Richie, J., & Spencer, L. (1994). *Qualitative data analysis for applied policy research*. In A. Bryman & R. G. Burgess (Eds.). *Analysing Qualitative Data* pp. 173 -194. London: Routledge.

Simba, T. (2008). *Patient preferences for breast cancer follow up: A discrete choice experiment*. Unpublished Masters Dissertation, University of Twente, The Netherlands.

Tamburini, M., Gangeri, L., Brunelli, C., Beltrami, E., Boeri, P., Borreani, C., Fusco Karmann, C., Greco, M., Miccinesi, G., Murru, L., Trimigno, P. (2000). Assessment of hospitalised cancer patients' needs by the needs evaluation questionnaire. *Annals of Oncology*, 11, 31 – 37.

The Royal Society of Medicine. (1999). Management of Gynaecological Cancer. *Effective Health Care*, 5(3), ISSN: 0965-0288.

Van den Beuken – van Everdingen, M. H. J., de Rijke, J. M., Kessels, A. G., Schouten, H. C., van Kleef, M., & Patijn, J. (2007). Prevalence of pain in patients with cancer: a systematic review of the past 40 years. *Annals of Oncology*, 18, 1437 – 1449.

Vistad, I., Moy, B. W., Salvesen, H. B., & Livaag, A. H. (2011). Follow-up routines in gynaecological cancer-time for a change? *Acta Obstet Gynecol Scand*, 90, 707 – 718. doi: 10.1111/j.1600-0412.2011.01123.x

Welsh Government (2012). *Together for Health-Cancer Delivery Plan: A delivery plan up to 2016 for NHS Wales and its Partners – The highest standard of care for everyone with cancer*. Retrieved from <http://wales.gov.uk/docs/dhss/publications/120613cancerplaneh.p>

**Gynaecological cancer follow-up after treatment care:
What attributes do patients and their informal caregivers value?
Patient information sheet**

What is the purpose of this study?

The NHS wants to improve services so that they give patients and their carers the support they need. Therefore, we want to identify what aspects of follow up care you and your carers think are most important.

Why have I been chosen?

You have been chosen because you are currently receiving follow up care after being treated for cancer of the womb, cervix or ovary. It is hoped that 18 patients will take part in the research.

What is involved?

If you decide to take part you would be asked to take part in a discussion group (conducted in English) to discuss which aspects of follow up care are important to you. There would be approximately eight people in the group (all who will be receiving follow up care for gynaecological cancer). The group would last about an hour, and would be held in the Tenovus mobile unit (a large mobile van). The discussion group would be audiotaped to help us explore the discussion after the group had finished. Your informal caregiver would also be asked to take part in a discussion group but this would be a different group to the one you take part in.

Do I have to take part?

No. Participation is completely voluntary. If you decide to take part you can choose not to answer any questions you are asked, and you can withdraw at any time without giving a reason. If you withdraw you have the right to ask for the data you have supplied to be withdrawn and destroyed, up to the point of the data analysis.

Will my information be kept confidential?

- Everything you say to us is treated in the strictest confidence (we won't tell anyone else). However, if you disclose that harm has been or might be caused to an individual, we are required by law to inform the appropriate authority.
- If it is okay with you we will write to your GP to let them know you are taking part in the study.
- If it is okay with you, we will give Tenovus your name and contact details, so that they can tell you about any services that might be of help to you.
- All data will be kept securely and will be stored for 15 years before being destroyed (in accordance with the relevant procedures).
- Direct quotes from the discussion might be used in a report or research publication, but these will be made anonymous.
- Data may be looked at by the NHS and Bangor University for the purpose of audit and monitoring.

Are there any benefits or risks from taking part?

- In the unlikely event that anything in the discussion group causes you distress, we will encourage you to have a break before if you wish, returning to the group.
- Talking to others who have been through a similar experience can be very supportive, helping people to realise that they are not alone.
- Taking part is likely to help others in the future, as it improves our understanding of how we can improve follow up care for patients.
- As the discussion group is held on the Tenovus mobile unit, there will be Tenovus staff members available to speak to after the group has finished, about the work that they do and how they can help you. They will be able to make referrals to services that they think could help you, for example counselling or financial advice.

Who is organising this research study?

This research is being organised by Bangor University, at the Centre for Health Economics and Medicines Evaluation (CHEME), in collaboration with the School of Psychology. This research study is being organised and carried out by Laura Budd as part of her PhD research, under the supervision of Professor Rhiannon Tudor Edwards (Professor of Health Economics), and Dr Val Morrison (Reader in Health Psychology and chartered Health Psychologist).

Who is funding this research?

The research is funded by Tenovus, a leading Wales Cancer Charity (registered charity number 1054015) through a PhD studentship.

Will I get paid?

Unfortunately there are no resources to be able to pay you for participating. However, we are able to reimburse you for your travel costs.

Who has reviewed the ethics of this research study?

The ethics of this research study have been reviewed and approved by the School of Psychology Bangor University, and the National Health Service Research Ethics Committee, and National Health Service Research and Development Forum.

What will happen after this study?

You will be sent a summary of the research findings in the post.

Who do I contact if I want further information?

If you would like any more information about the research study please contact the researcher. If you would like to seek advice about taking part in research generally, please speak to someone at the Macmillan Information and Support Centre in the hospital.

Concerns and Complaints

If you have a concern or complaint about your follow up care please use the normal NHS complaint system. In the event that you have any complaints or concerns about the research please contact Professor Robert T. Woods, Acting Director of the Institute of Medical and Social Care Research, Ardudwy, Normal Site, Bangor University, Bangor, Gwynedd, LL57 2PZ, or email b.woods@bangor.ac.uk. You may also contact Mr Hefin Francis, School Manager, School of Psychology, Bangor University, Adeilad Brigantia, Penrallt Road, Bangor, Gwynedd, Wales, UK LL57 2AS, or email h.franis@bangor.ac.uk.

Thank you very much for taking the time to read this information sheet. The results will improve our understanding of patients and carers follow-up needs and how the NHS could best meet them.

If you would like to take part, please sign the consent form and complete the brief questionnaire, returning both of these documents in the FREEPOST envelope provided. Please then give the 'Carer Envelope' to the person who you would like to take part as your informal carer.

(Sign)

Laura Budd

Appendix 2.3 Example of consent form (patients)

Gynaecological cancer follow-up after treatment care: What attributes do patients and their informal caregivers value? Consent form

	Please initial the box
I confirm that I have read and understand the consent form dated (version) for the above study. I have had the opportunity to consider the information, ask questions, and have had any questions answered satisfactorily.	<input type="checkbox"/>
I consent to the discussion group being audio recorded.	<input type="checkbox"/>
I understand that my participation is voluntary and that I can withdraw at any time without providing a reason, and without my medical care or legal rights being affected.	<input type="checkbox"/>
I consent to my GP being informed that I am taking part in the study	<input type="checkbox"/>
I understand that my participation and the data I provide is strictly confidential. However, I acknowledge that if I disclose harm to an individual that by law, this information has to be reported to the appropriate authority.	<input type="checkbox"/>
I understand that my data will be stored securely, in accordance with the Data Protection Act (1998). I understand that my personal data will be identified using only a number code and will be destroyed after 15 years.	<input type="checkbox"/>
I understand that relevant data collected during the study may be looked at by individuals from the research team, and for the purpose of audit and monitoring, by members of the auditing and monitoring team at Bangor University and the NHS Trust. I give permission for these individuals to have access my data for this purpose.	<input type="checkbox"/>
I agree to the use of direct quotes from the data I provide, and understand that if used, these will be anonymous.	<input type="checkbox"/>
I consent to taking part in the above study	<input type="checkbox"/>
I consent to Tenovus being given my contact details so that they can contact me, to see if they can offer me any support (if you do not agree to this, you are still able to take part in the study)	<input type="checkbox"/>

Name of Patient

Date

Signature

Name of Researcher

Date

Signature

**Gynaecological cancer follow-up after treatment care: What attributes do patients
and their informal caregivers value?**

Patient demographic questionnaire

Contact details

1. What is your name?

2. What is your address?

.....
.....
.....
.....

Is it ok if we contact you via post?

Yes ☐ No ☐

3. What is your email address?

Is it ok if we contact you via email?

Yes ☐ No ☐

4. What is your phone number?

Is it ok if we contact you via phone?

Yes

☐

No

☐

5. Who have you nominated as your informal carer for this research? An informal caregiver is a family member or friend who provides additional support or care for someone, due to an illness or disability they are experiencing.

6. What is their relationship to you?

7. How long do you feel this person has been providing care beyond that of 'wife' 'daughter/son' 'friend' etc?

Demographic questions

8. What is your age?

9. What is your ethnic group?
Choose one option that best describes your ethnic background

White

Welsh

☐

English

☐

Scottish

☐

Northern Irish

☐

British

☐

Irish

☐

Gypsy or Irish Traveller

☐

Any other White background, *please describe*

Mixed / Multiple ethnic groups

☐

White and Black Caribbean

White and Black African

White and Asian

Any other Mixed / Multiple ethnic background, *please describe*.....

Asian / Asian British

Indian

Pakistani

Bangladeshi

Chinese

Any other Asian background, *please*

describe.....

....

Black / African / Caribbean / Black British

African

Caribbean

Any other Black / African / Caribbean background,
please describe.....

Other ethnic group

Arab

Any other ethnic group, *please*

describe.....

10. What is your employment
status? Are you currently....

Employed for wages

A volunteer

Self-employed

Out of work and looking for work

Out of work but not currently looking for work

	A homemaker	
	A full time Carer	<input type="checkbox"/>
	A student	<input type="checkbox"/>
	Military	<input type="checkbox"/>
	Retired	<input type="checkbox"/>
	Unable to work	<input type="checkbox"/>
11. Who do you live with?	Alone	<input type="checkbox"/>
	My partner	<input type="checkbox"/>
	Family	<input type="checkbox"/>
	Friends	<input type="checkbox"/>
	Other	<input type="checkbox"/>
	<i>Please describe.....</i>	

Cancer care questions

12. What form of gynaecological cancer have you been treated for?	Endometrial (uterus or womb) cancer	<input type="checkbox"/>
	Cervical cancer	<input type="checkbox"/>
	Ovarian cancer	<input type="checkbox"/>
	Other	<input type="checkbox"/>
	<i>Please state.....</i>	

13. What stage of cancer did you have?

14. How long ago were you diagnosed with this form of gynaecological cancer?

15. What treatment did you receive? (Please tick all that apply)

Surgery ☐

Chemotherapy ☐

Radiotherapy ☐

Chemoradiation ☐

Other ☐

16. Which hospital were you treated at?

Ysbyty Gwynedd ☐

Ysbyty Glan Clywyd ☐

Ysbyty Maelor ☐

17. When did you receive your last treatment?

.....

18. When did you have your first follow up appointment?

.....

19. How many gynaecological cancer follow up appointments have you had?

.....

20. Where do you receive your follow up appointments?

At the hospital I was treated at ☐

At a local, community hospital ☐

At my GP surgery ☐

Over the phone ☐

I don't have routine follow up appointments,
I just call a nurse or consultant if I am worried
about anything ☐

21. Who delivers your follow up
appointment?

The consultant ☐

A specialist nurse ☐

My GP ☐

Other ☐

22. How often do you have follow
up appointments?

Every 3 months ☐

Every 6 months ☐

Every 12 months ☐

As and when I request one ☐

23. What happens in your follow
up appointment? (tick all that
apply)

I have medical tests ☐

Which medical tests do you have?

.....

I am asked how I am feeling ☐

I am given a needs assessment to look at my
physical, psychological (mental), social and
financial needs ☐

I am referred to other services that can help me ☐

Thank you very much for taking the time to complete this questionnaire. The researcher will be in contact with you in the next few weeks to inform you of the date, time and venue of your focus group.

In the meantime, if you have any questions please contact the Researcher on the contact details below. Thank you very much.

Laura Budd

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Appendix 2.4 Theme tables (patients)

Removed for copyright

Appendix 2.5 Theme tables (informal caregivers)

Removed for copyright

Appendices relating to Chapter 5, Discrete Choice Experiment

Gynaecological cancer patients, their informal caregivers, and health care providers preferences for Gynaecological cancer follow-up care: A discrete choice experiment

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Abstract

The number of gynaecological cancer survivors is increasing. Follow up care is given to patients following treatment, with the aim of enabling the management of patients' physical and psychological morbidity which may allow detection of possible recurrent disease. However, there

are no NICE guidelines regarding the provision of gynaecological cancer follow up after treatment care. The aim of this study is to explore the aspects of gynaecological cancer follow up care that are important to patients, carers and health care professionals, and identify whether 'one size of care fits all'. A discrete choice experiment, a method of eliciting the preferred attributes (aspects of care), will be completed by gynaecological cancer patients, their carers and health care professionals. This method has been previously used within BCUHB to elicit preferences for services. It is hoped that the findings will be used to inform the development of NICE guidelines and improve follow up care for gynaecological cancer follow up care patients across Wales.

Introduction

Endometrial, ovarian and cervical cancer

In the UK each year, there are approximately 8500 diagnoses of endometrial cancer, (c. per year), approximately 7000 diagnoses of ovarian cancer, approximately 3000 diagnoses of cervical cancer, and approximately 1200 diagnoses of vulvar cancer. Respectively, these gynaecological cancers are the fourth, fifth, twelfth and twentieth most common cancer sites in women (www.cancerresearchuk.org). The five year survival rate for patients diagnosed in England and Wales

with cervical cancer, ovarian cancer, and endometrial cancer between the year 1971 - 1975 was 52%, 21% and 61% respectively. This is compared to a 5-year survival rate of 67.4%, 46.3%, and 77.4% in patients diagnosed in England for the period 2007 - 2011 (Office for National Statistics, 2013).

Betsi Cadwaladr University Health Board (Ysbyty Gwynedd, Ysbyty Glan Clwyd, and Ysbyty Maelor) serves a population of approximately 740,000 people. Within Betsi Cadwaladr University Health Board in 2013, 91 women were diagnosed with ovarian cancer, 113 women were diagnosed with endometrial cancer, and 30 women were diagnosed with cervical cancer (Welsh Cancer Intelligence and Survival Unit, 2015).

Follow up care

The aim of follow up care is to enable the management of patients' physical and psychological morbidity which may allow detection of possible recurrent disease (The Royal Society of Medicine, 1999). Whilst follow-up after treatment for women with gynaecological cancer is routine, it is not evidence based. To date, there are no NICE recommendations as to what form or frequency of follow up after care is appropriate for survivors with gynaecological cancer, nor is the cost to the NHS weighed up against the benefits of other follow up care approaches. The Royal Society of Medicine (1999), claimed that there is no consensus as to what form or frequency of follow-up is appropriate or effective. A UK study found that 85% of the consultant gynaecologists surveyed (584/684) used 106 different follow-up protocols and the remaining 15% reported no routine follow-up (Kerr-Wilson and McCrum, 1995). Leeson et al (2013), conducted a UK nationwide survey of current practice in gynaecological cancer follow-up and reported similar findings. It is therefore unknown what follow up after care provision is most effective, good value for money to the NHS, and provides equitable access. Furthermore it is unknown what model of follow up care patients value.

Traditional models of follow up

The traditional model of follow up care is led by the consultant and requires multiple visits to hospital and biological tests to identify possible cancer reoccurrence. The frequency of follow up appointments under this model depends on the hospital protocol (Leeson et al, 2013). Reoccurrence is often symptomatic and patients often present with symptoms between scheduled follow up appointments (Vistad et al, 2011). This would trigger a referral for further investigation (Leeson et al, 2012). Additionally, it is unlikely that a patient will be diagnosed with reoccurrence based on test results alone (Nama, Nordin & Bryant, 2013). The traditional model of follow up after treatment care can lead

to high levels of anxiety amongst patients (Kew, Galaal, & Maderville, 2009) and patients have asked for alternative models of follow up after treatment care (Lydon, Beavery, Newbury, & Wray, 2009).

This traditional model of gynaecological cancer follow up care has an average cost of £127 per unit of follow up care, with 86,072 units of follow up care provided each year in England. However, the cost varies greatly with a minimum cost of £48 per unit compared to a maximum cost of £130 per unit (Department of Health, 2013). It was estimated based on the 2010 – 2011 NHS reference costs, current survival and current follow up protocols, that the cost of gynaecological cancer follow up care in Wales exceeds one million pounds per annum (Leeson et al, 2012). However, the average cost of follow up care has increased from £118 per unit to £127 per unit in the period 2012 - 2013, and the rate of survival is steadily increasing so this figure is likely to be higher in 2015 (Department of Health, 2013). Given the above, the efficiency of this traditional model of after care is questioned.

Alternative models of follow up care

There are various alternative forms of follow up after care including specialist nurse led follow up, telephone follow up, GP led follow up, and self-management.

The National Cancer Survivorship Initiative (2013a) recommends a stratified approach to cancer follow up care (see Figure 1), as opposed to the current traditional pathway which adopts a one size fits all approach (Macmillan Cancer Support, 2013). It is argued that by using a stratified patient centred approach, resources can be released and reinvested back into ‘survivorship pathways’ in order to improve patients’ outcomes and reduce the cancer burden on patients, carers, and their families (National Health Service Improvement, 2013). It has been suggested that patients with no cancer or cancer related morbidities could be assigned a self-management follow up pathway, whilst patients with non-cancer related morbidities could be assigned to a GP led follow up pathway, and those with cancer related morbidities could receive increased support from the cancer team (National Health Service Improvement, 2013).

It is reported using a sample of breast and lung cancer patients, that using a stratified approach for the whole cancer journey could, once all inpatient, outpatient, and emergency costs are considered, improve the efficiency, care, and management of co-morbidities. This would release a saving of 10% of the current NHS costs for patients suitable for a less intensive pathway of care, as well making a

saving to the wider economy through increased tax contributions and saved benefit payments (Allirajah, 2010; National Cancer Survivorship Initiative, 2013).

The NHS is prioritising the improvement of outcomes from chronic diseases, including cancer (Department of Health, 2012). This priority is reiterated by Macmillan Cancer Support (2013), who argue that the development of new pathways and follow up after care models to address the consequences of cancer and cancer treatment, and undertaking cost analyses of these models is a priority, particularly in pelvic cancers (Macmillan Cancer Care, 2013).

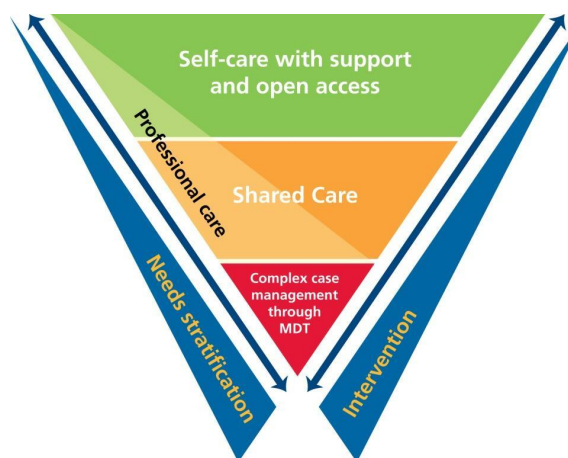


Figure 1. Stratified approach to follow up care (National Cancer Survivorship Initiative, 2013b)

Patient Centred Services

The NHS's modernisation plan (Department of Health, 1997) and the new Welsh cancer plan 2012 (Welsh Government, 2012) highlight the importance of designing a patient-centred service around the needs and aspirations of service users. Additionally, the International Society for Pharmacoeconomics and Outcome research (2011) states that:

"The perspective on outcomes should be all direct health effects, whether for patients or, when relevant, other people (principally carers)"

Carers, or informal caregivers in this population, are family members or friends whom provide a bit of extra support for the patient, in their gynaecological cancer experience. This could be supporting them to attend follow up care appointments and/or providing emotional support.

However, there is limited published information relating to the preferred model of follow up care for women with gynaecological cancer and their informal carers. It can therefore be seen that in order for the NHS to meet patients' follow up needs, patient preferences need to be better understood. Identifying patients' and their informal caregivers' preferences for the model (attributes and attribute levels) of gynaecological cancer follow-up services would undoubtedly facilitate future health service reconfiguration, whilst perhaps reducing inefficiencies in the provision of healthcare services within limited NHS resources, and improving equity of access.

Health Economics

Health Economics is the study of how scarce resources can be allocated when needs are infinite (Morris, Delvin, & Parkin, 2007). The aim of health economics is to increase the efficiency and equity of health care resource provision, whilst acknowledging that this is a trade-off. Therefore adopting a health economics perspective in this research can aid understanding of how to improve the efficiency of gynaecological cancer follow up care, whilst exploring equity applications and remaining patient centred, focused on the needs of patients as valued by patients, informal caregivers and health care professionals.

It has been recommended that NICE use discrete choice experiments as a way of identifying patient preferences (Ryan, 2004). Discrete choice experiments are embedded in welfare economics. In this stated preference elicitation method it is assumed that individuals are rational decision makers who make decisions based on, and aiming to maximise their innate stable preferences (Consumer Choice Theory). In accordance with neoclassical consumer theory, individuals will have a preference and hence make a choice for the bundle of attributes that has the maximum benefit for their utility, taking into consideration their budget. Individuals derive their utility, and hence preference, from the collection of attributes opposed to the overall commodity (Lancaster's Economic Theory of Value: Lancaster, 1966). Therefore, individuals will make a preference and choice for the bundle of attributes that has the highest overall utility (benefit) for them. If any of the attribute levels that make up this bundle changes, it may cause a discrete shift in a preference from one set of attributes to another set of attributes. By asking participants to make a choice between a set of attributes it provides information on individuals' relative preferences for each attribute (Ryan et al, 2008). Whilst discrete choice experiments have not been previously used to identify patients, informal caregivers and health care professionals' preferences for gynaecological cancer follow up care, they have been used to identify the preferences for attributes of follow up care in breast cancer patients (Kinman, Dellaert, Boersma, Lambin & Dirksen, 2010; Simba, 2008). Results showed that health care provider, mode of

contact, frequency of visits and waiting time were all important attributes of breast cancer follow up care as valued by patients. However, provision of an educational program compared to no educational program was not significant. Follow up care delivered by a consultant was preferred to follow up care delivered by a specialist nurse or GP, but follow up care delivered alternatively by a consultant and specialist nurse was also acceptable to the participants. Participants had a strong preference for face to face opposed to telephone appointments, and preferred a reduced waiting time and an increased frequency of appointments (Kinman et al, 2010). However, Simba (2008) found that patients had a preference for follow up care delivered twice a year over five years by a surgeon in a face to face format, however, follow up over 3 years, face to face by a specialist nurse was also acceptable to patients.

Aims and Objectives

In light of the above, the aim of this phase of the PhD research (3rd phase of the PhD research project) is to identify gynaecological cancer survivors', their informal caregivers', and health care professionals' preferences for the different models and respective aspects/attributes (for example the professional that provides the care), and attribute levels of follow up care (for example consultant, GP, or specialist nurse). Using the attributes and respective levels identified from the systematic literature review (phase one), and qualitative research focus group (phase two, IRAS number: 149889) the aim of the discrete choice experiment (phase three) is to:

1. Identify the model and respective attributes of follow up care preferred by gynaecological cancer patients (as measured by the discrete choice experiment in accordance with health economic theory).
2. Identify the model and respective attributes of follow up care preferred by the informal caregivers of gynaecological cancer patients (as measured by the discrete choice experiment in accordance with health economic theory).
3. Identify the model and respective attributes of follow of follow up care preferred by the health care professionals delivering follow up care (as measured by the discrete choice experiment in accordance with health economic theory).
4. Draw comparisons between the preferred model of care and respective attributes for patients, carers and health care professionals (as measured by the discrete choice experiment in accordance with health economic theory).
5. Identify whether participants with different characteristics have different preferences (as measured by the socio demographic questionnaire; quality of life, health related quality of life,

perceived risk of recurrence measures; and the discrete choice experiment in accordance with health economic theory).

6. Explore associations between patient and carer characteristics (for example, stage of disease and fear of recurrence, in accordance with health psychology theory)

Methods

Design

This is the third phase of a mixed method study, whereby results from the systematic literature review (phase one), and cross-sectional qualitative study (phase two: IRAS number 149889), have influenced the design of the discrete choice experiment (phase three). The discrete choice experiment is a cross sectional questionnaire study. This research study meets the inclusion criteria for adoption by the NISCHR CRC portfolio, and so will be registered on this database following ethical and R&D approval.

Participants

Women (and their informal caregivers) who have completed primary treatment for endometrial, ovarian, cervical or vulvar cancer and are currently undergoing follow up after treatment within Wales will be invited to take part in the current study. Patients will not be excluded if their informal caregiver does not consent and complete the questionnaire booklet and carers will not be excluded if the person they provide informal care for does not consent and complete the questionnaire booklet. Health care professionals and research nurses and research officers in the respective outpatient wards will identify, screen, and give the recruitment pack (envelope containing the research invite letter, participant information sheet, consent form, request for more information, questionnaire booklet and a smaller envelope containing the informal caregiver versions of these documents) to eligible patients in the waiting room before their follow up care appointment, or after their follow up appointment.

The PhD is funded by Tenovus Cancer Care. Tenovus Cancer Care will send a research study advertisement poster and the recruitment documents to individuals on their mailing list (this mailing list has been set up to contain the information of people who would like to take part in research). One of Tenovus Cancer Care's research advisory group members is active in an ovarian cancer support group and so may be able to forward any request for involvement to that group.

Secondary health care professionals who work in the area or have an interest in gynaecological oncology in Wales will be invited to take part in the current study. Health care professionals will be recruited at the South Wales Gynaecology Oncology Network Meeting, North Wales Gynaecological Cancer Educational Evening in Winter 2015 and other relevant training days/presentations/conferences via an interactive presentation using Microsoft PowerPoint and participation handsets. It is important to understand secondary Health Care Professionals views, because as the people providing the care, their preferences influence service delivery.

Primary health care professionals working in Betsi Cadwalder University Health Board will be invited to take part in the study. NISCHR CRC Primary Care workers will send an expression of interest and information sheet to various primary care practices, and a couple of weeks following will send the various practices a copy of the health care professional questionnaire. Macmillan Cancer Care discuss the importance of primary care having increased involvement in cancer follow up, and there is a shift to more involvement of GPs in cancer follow up. Therefore it is important to understand Primary health care providers preferences for the delivery of gynaecological cancer follow up.

Sample size

In accordance with DCE theory and guidance, given the number of attributes and attribute levels in the discrete choice experiment, a minimum sample size of 30 is required (Ryan et al, 2008).

Based on North Wales diagnosis and five year survival rates (Public Health Wales, 2015; Leeson et al, 2012), there are approximately 575 patients undergoing gynaecological cancer follow up care each year in BCUHB. This equates to approximately 191 patients being seen for a follow up appointment in a four month period. Assuming a 50% recruitment period it is anticipated that 96 patients will be recruited. Based on the recruitment of carers in the qualitative study, it is anticipated that 50 carers will be recruited.

It is unknown how many patients and carers will be recruited via online methods, as it is unknown how many people will view the research study advert, how many of these people will be eligible to take part and how many will want to take part.

Based on the number of health care professionals who attend the at the North Wales Gynaecology Cancer Education Evening and South Wales Gynaecology Oncology meeting, and assuming a 50% response rate it is anticipated that at least 50 secondary health care professionals will be recruited. Based on communication with Melissa Van Der Bijl (NISCHR CRC Primary Care Team Leader North

Wales), if the study is adopted by the NISCHR CRC portfolio, it is envisaged that 50 primary health care professionals (GP's and practice nurses) will be recruited.

Inclusion criteria for patients and carers

- Patients undergoing follow up after treatment for gynaecological cancer following treatment with curative intent, or an informal caregiver for this patient.
- The patient (or patient for whom the informal caregiver provides care) has had at least one follow up after treatment appointment.
- The patient (or patient for whom the informal caregiver provides care) had no sign of active disease at their last follow up appointment.
- The patient (or patient for whom the informal caregiver provides care) is receiving follow up after treatment within Wales
- The participant is aged 18 years or over.
- The participant can speak English.
- The participant has the capacity to consent.

Exclusion criteria for patients and carers

- Patients and carers who are cognitively impaired (for example, dementia), have a serious psychiatric condition or a serious developmental or learning difficulty.

Inclusion criteria for health care professionals

- Health care professionals who have an interest in gynaecology oncology or are involved in the delivery of gynaecological cancer follow up (under the holistic definition of follow up care).
- Health care professionals who practice in Wales.

Measures

Patients, carers and health care professionals will complete measures specific to the population, as well as generic measures. These measures were chosen because of the issues highlighted in the systematic literature review (phase one) and the qualitative study (phase two). Participants' responses to these measures will be imputed into the DCE model, to identify whether preferences for gynaecological cancer follow up differ by participant characteristics.

Socio-demographic questionnaire

The patient socio-demographic questionnaire includes questions regarding the patients' cancer diagnosis, treatment and follow up care, as well as questions about their age, employment status and household income.

The informal caregiver socio-demographic questionnaire includes questions about the relationship of the informal caregiver to the patient, the informal caregiver's role as well as questions about their age, employment status and household income.

The Health Care Professionals socio-demographic questionnaire includes questions about the occupation and role of the health care professional.

Patient and informal caregiver specific measures

Health Outcomes

The EQ-5D-5L is a validated and generic measure of health outcomes widely used in clinical studies. It is comprised of five questions relating to the domains: mobility; usual activities; self-care; anxiety and depression; pain and discomfort. Under each of these domains participants are required to indicate whether they have: no problems; slight problems; moderate problems; severe problems; or are unable to carry out this task. Following the questions participants are asked to complete a visual analogue scale (VAS) requiring participants to on a 10cm line how their health is today.

Quality of Life

EORTC QLQ C30 is a 30 item validated measure of quality of life. Participants are required to respond to five items relating to current health, 22 items relating to health in the past week requiring participants to indicate their response on a four point numerical scale 'not at all' (one), 'a little' (two), 'quite a bit' (three), 'very much' (four). Two final items require participants to rate their health and quality of life overall in the past week on a scale of one to seven with one being 'very poor' and seven being 'excellent'.

Fear of cancer recurrence

Analysis of the data from phase two of the research (IRAS number: 149889) highlighted the importance of patients and carers perceived risk of recurrence in the valuation of their follow up care. There are many measures used to measure fear of cancer recurrence in different cancer populations (Thewes et al, 2012; Sinard et al., 2013; Cancer Australia 2014). However, few have been validated (Thewes et al, 2012; Simard et al, 2013). The Assessment of Cancer Survivor Concerns

(Gotay & Pagano, 2007) has shown high levels of internal consistency ($\alpha = 0.93$ for cancer worry, $\alpha = 0.72$ for health worry) and internal and external validity in a sample of over 700 general cancer survivors, and in a systematic review of fear of cancer recurrence was assessed as having the highest total quality rating of brief measures when assessed against the medical outcomes trust criteria (Scientific Advisory Committee of the Medical Outcomes Trust, 2002; Thewes et al, 2012). The measure consists of five questions regarding fear of recurrence, fear of a new diagnosis, fear of future tests, fear of health and fear of general health. Participants are required to respond to each of these questions on a likert scale 'not at all' (1) to 'very much' (4). Higher scores are indicative of a higher perceived fear of cancer recurrence (Gotay & Pagano, 2007, Thewes et al, 2012).

Generic measures (for patients, carers and health care professionals)

Discrete choice experiment (primary outcome measure)

The discrete choice experiment will contain eight questions, where the patient is asked to choose which hypothetical model of care they prefer between 'Follow up Care A' or 'Follow up Care B' (a pairwise choice DCE). Each of these hypothetical models will be described by the aspects (referred to in health economics as attributes and attribute levels) that comprise it. Qualitative research, a systematic review, and discussions with health care professionals were used to identify the attributes and attribute levels to be used in the discrete choice experiment. This pilot work resulted in the identification of five attributes. Four attributes have two levels, and one attribute has four levels, resulting in a full factorial model with 32 possible scenarios (see Table 1). To reduce burden a fractional factorial design (FrFD) was therefore designed (Kocur et al, 1982). The minimum number of choice sets (questions) for this number of attributes and respective levels was eight. Using this catalogue (Kocur et al, 1982) ensured that there was orthogonality (that the occurrence of one attribute was not dependant on the occurrence of another attribute), and level balance (that each attribute level is presented the same number of times). The DCE will be piloted and an analysis conducted, and if necessary, the DCE will be amended accordingly. The wording of the DCE has been amended to suit the perspective of different populations (carers and health care professionals). For example, "I have the same medical care as everyone else who has had gynaecological cancer" has been changed to "I would like the person I care for to have the same medical care as everyone else who has had gynaecological cancer" for caregivers, and to "Patients have the same medical care as everyone else who has had gynaecological cancer" for health care professionals.

Where available and validated, Welsh versions of the measures will be included in the Welsh language pack.

Table 1. Attributes and attribute levels for the discrete choice experiment

Attributes	Levels (questionnaire design code)
Personalised care	I have the same medical care as everyone else who has had gynaecological cancer (0) The care I receive is specific to my physical and mental needs (1).
Information received	I receive basic information (0), I receive comprehensive information (1)
Medical examination	I have a medical examination if evidence shows that it cannot detect whether the cancer has come back if I have no symptoms (0) I have a medical examination if there is evidence to show that the examination can detect recurrence (1)
Health care professional seen	I see the GP for my appointments (0), I see the specialist nurse for my appointments(1), I see the consultant for my appointments (2), I see different people in the multidisciplinary team for my appointments depending on my needs (3)
Frequency of appointments	I have an appointment every 12 months (0) I have an appointment every 6 months (1)

Materials

All materials will be translated into the Welsh language following ethical approval. Materials will be printed onto different coloured paper to aid understanding of what documents need to be returned to the researcher, thus reducing participant burden. These materials will be sent via email to potential participants approached using online methods (apart from the 'research invite letter' and 'request for more information').

For Patient and Informal caregiver Participants

Participants will be given a **participant information sheet** which outlines the aim of the study, what is required of their participation, confidentiality and contact details for the researcher (see *ethical considerations*).

Participants will be asked to complete a **consent form**, where they will be asked to sign that they have read and understood the participant information sheet and had the opportunity to have any questions answered; understand that participation is voluntary; that participation and the data they provide is strictly confidential. However, if they disclose harm to an individual that by law, this information has to be reported to the appropriate authority; that data will be stored securely, in accordance with the Data Protection Act (1998); that data may be looked at for the purpose of audit and monitoring by members of the auditing and monitoring team at Bangor University and the NHS Trust; that they consent to taking part in the study; that they consent to their GP being informed that they are taking part in the study; that they consent to Tenovus Cancer Care being given their contact details so that they can contact them, to see if they can offer the participant any support (if participants do not agree to this, they are still able to take part in the study) (see *ethical considerations*).

If patients and/or carers would like more information before consenting to take part in the study, or would like some help in completing the questionnaire, they are asked to telephone the researcher or complete and return the **request for more information**. The **debrief form** will again explain the aim of the study, how the participants' data will be used, contact details for the chief investigator, and contact details for the Tenovus Cancer Care Support line in case the individual would like to discuss anything regarding their cancer needs following involvement in the study.

For patients and informal caregivers recruited online, these materials will be returned to the researcher via email or post using the FREEPOST address.

For Health Care Professional Participants identified at the North or South Wales Gynaecology Oncology educational afternoon/evening

A paper copy of the **participant information sheet** will be given to potential participants and presented to them on a PowerPoint presentation (as part of the **Health care professional presentation**). The participant information sheet outlines the aim of the study, what is required of their participation, consent, and confidentiality. Participants are asked to respond by pressing a button on the participation handsets if they consent to taking part in the study, before the questionnaire begins. The use of a presentation and participation handsets (see Figure 2) in this way ensures participants understand the questions asked of them, as well as aiding a timely response to the questions. The participation handsets are owned by Bangor University and supplied by Turning Technologies (<http://www.turningtechnologies.co.uk/>). Participants' responses are anonymous and will be saved as a data set for later statistical analysis. These handsets are therefore suited to be used to collect questionnaire data (<http://www.sussex.ac.uk/tel/learningtechnologies/prs/tp>). The **debrief form** (on paper and as part of the **Health care professional presentation**) will again explain the aim of the study, how the participants' data will be used, and contact details for the chief investigator. If technology problems occur, participants will be given a paper copy of the presentation and asked to respond to the questions on the paper version.

For Health Care Professional Participants identified through GP practices

Following portfolio adoption of the research study with NISCHR CRC, NISCHR CRC Primary Care workers will send an expression of interest and information sheet to PiCRIS and other GP practices in BCUHB. The participant information sheet outlines the aim of the study, what is required of GP and practice nurse participation and confidentiality. Consent will be assumed by completion of the questionnaire. The debrief form will be viewed via email (and can be printed) following completion of the questionnaire.

For Recruiting Health Care Professionals, Research Nurses and Research Officers

Health Care Professional's and Research Nurses and research officers will complete the **eligibility screening form** for every patient screened. This document will then be put in the patients file as a record and to prevent them from being screened twice. An **eligibility screening sticker** will be put on the patients file so research nurses, research officers and health care professionals can easily identify whether the patient has been given the recruitment pack. Only health care professionals, research

nurses and research officers recruiting participants within the respective outpatient unit will view this document. The relevant information on the eligibility screening form (eligibility, if applicable reason not eligible, whether recruitment pack was given) will then be copied onto the **screening, eligibility and recruitment pack record** and will be given or emailed to the Chief Investigator after each clinic.

Figure 2. A picture of the participation handset



For the Chief Investigator

When the researcher has contact with the potential participant or participants, they will complete the contact form. Each individual who makes contact with the Chief Investigator will have their own case report form. This is to be completed following every contact, using the information from the contact form.

Procedure

Procedure for recruitment of patients and carers within Betsi Cadwaladr University Health Board

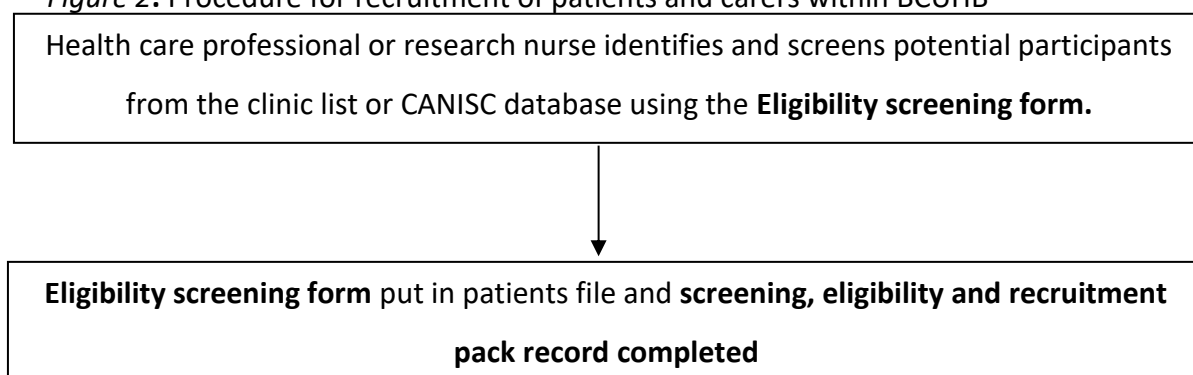
Once ethical and governance approval is granted from the School of Psychology, Bangor University, and the National Health Service Research Ethics Committee and Research and Development Forum, recruitment will commence (Autumn 2015). There will be a phased approach to recruitment of patients and carers (see Figure 2). In the first phase health care professionals and/or research nurses and/or research nurses in the relevant outpatient clinics will identify and screen participants against the eligibility criteria from the clinic list (see **eligibility screening form**). Research nurses and/or research officers and/or health care professionals may also screen patients for eligibility using the Cancer Network Information System Cymru Database CANISC . The CaNISC database holds data on an

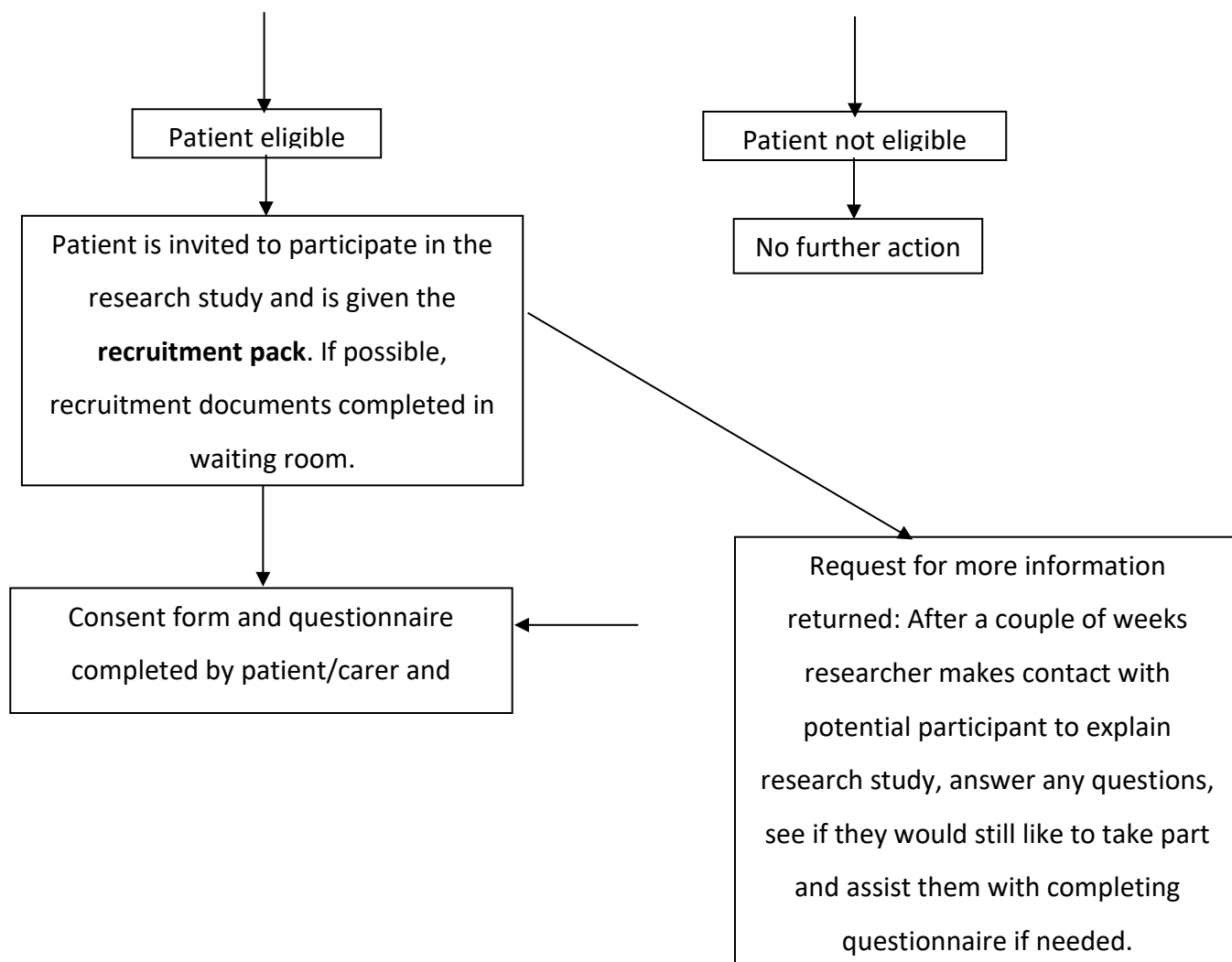
individual level for cancer patients, the data is held at a single location but is able to be accessed across Wales by health care professionals. The research nurses and/or research officers will confirm with the treating clinician the eligibility of the patient, before they invite the patient to take part in the study and provide the recruitment pack.

Where possible, research nurses or research officers or health care professionals will give eligible participants the **recruitment pack** to complete in the waiting room before their appointment, with the research nurse/research officer nearby to assist in any clarification. When this is not possible, the **recruitment pack** will be given to patients in person at or after their appointment or sent to them in the post. On the front of the recruitment pack is the **request for more information**, which patients can complete and return to the research nurse, research officer or clinician in their appointment if they would like more information or for someone to contact them regarding the study. The recruitment pack contains the **research invite letter, participant information sheet, consent form, questionnaire booklet** (containing the relevant measures as described above), **debrief form** and two FREEPOST envelopes for patients and their informal caregiver. The recruitment pack for the informal caregiver will be in a separate envelope. Each of the documents will be on different coloured paper as described above.

Potential participants are prompted to contact the researcher if they have any questions before consenting, and can do this by telephoning the researcher on the contact details provided, or by returning the **request for more information**. If the patient/informal caregiver returns the **request for more information**, the researcher will make contact with the patient/informal caregiver to explain the research study further and to answer any questions the individual may have, and if applicable, provide more detailed instructions to aid completion of the questionnaire (if needed, filling this in on the phone with them). For research documents completed in the clinic, the research nurse/research officer will return the documents to the researcher via post, e-mail or face to face.

Figure 2. Procedure for recruitment of patients and carers within BCUHB





If the patient/informal caregiver has expressed an interest in the research study but does not return their **consent form** and **questionnaire booklet**, follow up telephone call(s)/email(s)/letter(s) will be made a couple of weeks later if there is an insufficient sample size.

Following return of the **consent form** and **questionnaire booklet**, if there is any missing data in any of the measures, the researcher will make contact with the participant to retrieve this data, supporting the participant on the phone to answer the questions relating to the missing data if required. If the participant provided the relevant consent, the researcher will provide a named contact at Tenovus Cancer Care with their contact details so Tenovus Cancer Care can offer the participant any follow up support (*see ethical considerations*).

Procedure for Recruitment via Online methods

The PhD is funded by Tenovus Cancer Care. Tenovus Cancer Care will send a research study advertisement poster and the recruitment documents to individuals on their mailing list (this mailing list has been set up to contain the information of people who would like to take part in research).

Patient and carer participants recruited within BCUHB or online and who complete the recruitment questionnaire will be entered into a prize draw with the winner chosen at random, to receive one of two £50 or £25 vouchers. In a systematic literature review the use of financial incentives have been shown to increase recruitment (Giuffrida & Torgerson, 1997).

Procedure for Recruitment of Health Care Professional Participants identified at the North or South Wales Gynaecology Oncology educational afternoon/evening

At the North Wales Gynaecology Cancer Education Evening on Thursday 19th November the Researcher is providing a brief overview of the results from her focus group and interview study (phase two, IRAS number: 149889). During this presentation the researcher will present the study to attendees using Microsoft PowerPoint and Participation Handsets (**health care professional presentation**). The **health care professional presentation** contains the **participant information sheet**, **consent form**, brief **socio-demographic questionnaire**, the **discrete choice experiment** and the **debrief form**. A paper copy of the health care professionals version of the **participant information sheet** and **debrief form** will be given to participants.

At the South Wales Gynaecology Oncology Meeting in Winter 2015 the researcher will present the research study to attendees using Microsoft PowerPoint and Participation Handsets (**health care professional presentation**). The **health care professional presentation** contains the **participant information sheet**, **consent form**, brief **socio-demographic questionnaire**, the **discrete choice experiment** and the **debrief form**. A paper copy of the health care professionals version of the **participant information sheet** and **debrief form** will be given to participants.

The researcher will use this method described to recruit other health care professionals with an interest in gynaecology oncology at other relevant training days/presentations/conferences.

Use of Microsoft PowerPoint and Participation Handsets in this way will ensure that all participants understand the task, as well as ensuring a high response rate of anonymous data. Following completion of the **socio-demographic** data and the **discrete choice experiment**, participant

responses will be saved and later inputted into the data sheet for analysis. The presentation has been piloted and has a duration of ten minutes. In technology problems occur, participants will be given a paper copy of the presentation and asked to respond to the questions on the paper version.

For Health Care Professional Participants identified through GP practices

Following portfolio adoption of the research study with NISCHR CRC, NISCHR CRC Primary Care workers will send out an expression of interest and information sheet to PiCRIS and other GP practices within BCUHB. In the couple of months following, NISCHR CRC Primary Care workers will send the same various practices a copy of the health care professional questionnaire for completion. Primary care practices can return the questionnaire via email or post. Reminders will be sent as and when required. Completion of the questionnaire will count towards the surgeries PiCRIS award.

PiCRIS is the primary care research incentive scheme, and is part of NISCHR CRC. PiCRIS supports and funds general practices across Wales as an incentive to become actively involved in research. PiCRIS practices receive different funding amounts per year depending on the number of studies they engage in.

Patient Involvement

Patient and caregiver representatives have been consulted regarding the readability and understanding of the recruitment documents and recruitment processes.

Statistical Analysis

The data generated by the study will be analysed by the researcher with the support of her supervisors, and statistical advisors if required. The statistical aspects of the research have been reviewed by a statistician within the research team (Seow Tien Yeo).

The primary outcome is patients, carers and health care professionals' preferences for gynaecological cancer follow up care as measured by the discrete choice experiment.

Method of Analysis

The data will be inputted into STATA and analysed in accordance with economic theory (Ryan et al, 2008). The internal and external validity of the DCE model will be explored, before the main effects and interactions are identified. The analysis will increase understanding of the model of follow up care which is most valued by patients, carers and health care professionals, and provide an

understanding of the trade-offs participants are prepared to make between the different attributes and levels (this analysis will meet objective one, two and three). Patients, carers and health care professionals preferences will then be compared (This analysis will meet objective four).

The secondary outcome seeks to answer the questions:

- 1) Do patients, carers and health care professionals with different characteristics have different preferences for care

Method of Analysis

Sub group analyses will be conducted on the DCE model (the primary outcome analysis described above) to identify whether participants with different characteristics have different preferences for gynaecological cancer follow up after care in accordance with health economic theory (This analysis will meet objective five).

The third outcome is:

Any further analysis that are deemed of interest depending on the findings from the first and second analysis as described above, for example, the association between patients fear of cancer recurrence and carers fear of cancer recurrence in accordance with health psychology theory as this PhD is a collaboration of health economic and health psychology theory (This analysis will meet objective six). It is important to be explicit that such analyses will be conducted without being explicit as to the exact hypotheses and subsequent analysis. As part of the PhD process students are required to look at the data generated and generate further hypotheses of interest based on these findings. This is not a fishing exercise and all analyses' conducted will be included in the Chief Investigators doctoral thesis.

Method of Analysis

The data will be inputted into SPSS and analysed to explore associations and/or significant differences between the above variables of interest as identified in the analysis of outcome one and two. The data will be checked for normality and homogeneity of variance, before being analysed using the appropriate parametric or non-parametric test.

Professor Valerie Morrison's undergraduate project students will be given a sample of up to 30 **anonymous** health care professional's data sets, for comparison with a similar study they have conducted in a student population.

Ethical considerations

Capacity to consent

Health care professionals and research nurses and research officers within Betsi Cadwaladr University Health Board will screen patients based on the inclusion criteria, of which one criterion is patient's capacity to consent. For informal caregiver's recruited via the patient, and for patients and carers recruited by online methods, capacity to consent will be indicated by the individual's ability to complete the consent form and measures. Health care professional's capacity to consent will be assumed by their presence at the gynaecology education afternoon/evening.

Informed consent

The patient and informal caregiver **information sheet** contains detailed information about the research study; its aims; methods; confidentiality; the name and contact details of the principal investigator; information about how the data will be used; and how the research study findings will be made available to participants. Because it is a questionnaire study, patients and carers who do not wish to participate will not return the consent form and questionnaire booklet. Participants are prompted to contact the researcher to ask any questions before consenting if they wish. Participants are then asked to complete the **consent form** if they would like to take part in the study. Participants are asked to sign that they have read and understood the participant information sheet and had the opportunity to have any questions answered; understand that participation is voluntary; agree that participation and the data they provide is strictly confidential, however, if they disclose harm to an individual that by law, this information has to be reported to the appropriate authority; consent for data to be stored securely in accordance with the Data Protection Act (1998); consent for their data to be looked at for the purpose of audit and monitoring by members of the auditing and monitoring team at Bangor University and the NHS Trust; that they consent to taking part in the study; that they consent for their GP to be informed that they are taking part in the study; that they consent to Tenovus Cancer Care being given their contact details so that they can contact them to see if they can offer any support (if participants do not agree to this, they are still able to take part in the study) (*see ethical considerations, potential distress*). There will be two copies of each consent form, one to be kept by

the participant and one to be stored by the researcher. The researcher will sign both copies before it is included in the recruitment pack.

Potential Benefit

By taking part in the research study participants have the opportunity to hopefully influence the re-design of follow up care in the future. Participants who complete the recruitment pack will be entered into a prize draw with the winner chosen at random, to receive one of two £50 or £25 vouchers.

Potential Distress

There is a small risk that patients and carers may find completing the questionnaires upsetting as they encourage participants to think about their cancer pathway and how they are feeling. The participant information sheet and consent form clearly explains the potential risks. In the previous research study (Phase two: IRAS number 149889), it was clear that some participants were distressed as a result of their cancer pathway. Providing participants with the option for their contact details to be passed to Tenovus Cancer Care provided an opportunity for patient's and carer's needs to be addressed, and reduced the emotional burden placed on the researcher. Participants' GP's will be informed that they are taking part in the study, so that if participants speak to their GP about the research study they are fully aware of the study and able to offer appropriate support.

In the socio demographic questionnaire participants are asked about their income and employment status. It is acknowledged that these questions are of a sensitive nature, however, are relevant to the study of health economics and will be used to describe the population. Additionally, the income question relates to the final question in the DCE regarding participants' willingness to pay for follow up care. Responses to these questions will be used to identify the value participants place on their/their informal caregivers follow up care appointments.

It is not envisaged that any harm will be caused to health care professionals who participate in the research study.

Disclosure of Harm

Disclosures of harm requiring action are considered unlikely to occur. If disclosures of harm are made to the recruiting research nurses or research officers or health care professionals, this will be dealt with in accordance with BCUHB policies and procedures. If disclosures of harm are made during

contact with the researcher, it will be discussed with the researcher's supervisor before reporting to the appropriate authority. Any record of disclosures will be kept.

Participant Burden

It took the patient and carer representatives between 10 and 20 minutes to complete the recruitment booklet.

The research team are aware of the number of questionnaires that need to be completed for this research study. The decision of measures to be used has been discussed at length within the research team, with the aim of minimising participant burden whilst obtaining the data needed to meet the aims and objectives of this research study. Where possible, brief measures have been chosen. For example, the Fear of relapse/recurrence scale was chosen because it is a five item measure. The socio-demographic questionnaire has been amended since phase two (IRAS number: 149889) to reduce participant burden. Regarding the discrete choice experiment, the number of potential attributes and attribute levels has been reduced to five attributes to reduce the number of possible choice sets.

To reduce participant burden further, the consent form, request for more information and questionnaire booklet will be on different coloured paper and there will be two separate booklets for participants, an English language booklet and a Welsh language booklet.

Researcher potential distress

The low score of the measures (health outcomes, quality of life, risk of recurrence) may cause emotional burden to the researcher. By asking participants in the consent form if they would like Tenovus Cancer Care to be given their contact details to see what support they can provide them with, this will reduce the potential distress on the researcher (this is not compulsory for participants, if they do not wish for Tenovus Cancer Care to be given their contact details patients and carers can still take part in the research study).

Data Storage

Participant's personal contact information will be used to contact participants for the purpose of the research study. Identifiable data will be kept in a locked filing cabinet in a lockable room at Bangor University and on the researchers encrypted university laptop on an encrypted file. These files will

be kept separate to anonymous data. Anonymous data will also be kept in a locked filing cabinet in a lockable room at Bangor University and on the researchers encrypted university laptop. The chief investigator is the only key holder for the filing cabinet. In the anonymous data set participants will be known by their non-identifiable participant number. In accordance with Bangor University Research Data Management policy (November 2-14, Final version), non-anonymous data will be destroyed a maximum of three months following completion of the research study and anonymous data will be archived for a period of three years before being destroyed. Only members of the research team will have access to the original data.

Professor Valerie Morrison's undergraduate project students will be given a sample of up to 30 **anonymous** health care professional's data sets, for comparison with a similar study they have conducted in a student population.

If participants complete the request for more information or recruitment pack during the clinic, the recruiting health care professional or research nurse may send the participants' documents to the researcher by electronic device (or by post or give it to her face to face).

For participants who consent for their contact details to be passed to Tenovus Cancer Care for Tenovus Cancer Care to offer them any support, the researcher will email the named contact in Tenovus Cancer Care the participants' contact details.

Audit and Monitoring

Monthly PhD supervision meetings are held to monitor issues arising during the research.

Monitoring and auditing of the research study will be conducted by the NHS research departments and Bangor University as is required and requested. The Chief Investigator will complete bi-annual R&D reports as requested by the NHS R&D forum. All of the research study documentation will be made available to the relevant regulators for auditing and monitoring as requested. As part of the informed consent process, participants are required to consent to this.

Dissemination

The results from this research study will be disseminated at a conference presentation and in a peer reviewed scientific journal.

Study participants will be sent an overview of the results at their request.

References

- Adams, E., Boulton, M., Horne, A., Rose, P. W., Durrant, L., Collingwood, M., Ozkrochi, R., Davidson, S. E., & Watson, E. (2014). The effects of pelvic radiotherapy on cancer survivors: Symptom profile, psychological morbidity and quality of life. *Clinical Oncology*, 26, 10 – 17. doi: <http://dx.doi.org/10.1016/j.clon.2013.08.003>
- Allirajah, D. (2010). *Demonstrating the Economic Value of Co-ordinated Cancer Services*. Retrieved from <http://www.macmillan.org.uk/Documents/AboutUs/Commissioners/MonitorResearchinManchesterbriefing.pdf>
- Andreyev, H. J. N., Davidson, S. E., Gillespie, C., Allum, W. H., & Swarbrick, E. (2012). Practice guidance on the management of acute and chronic gastrointestinal problems arising as a result of treatment for cancer. *Gut*, 61, 179 – 192.
- Bandura, A. (1977). Self-efficacy: Toward a unifying theory of behavioral change. *Psychological Review*, 84, 191-215. doi: 10.1037/0033-295X.84.2.191
- Boberg, E. W., Gustafon, D. H., Hawkins, R. P., Offord, K. P., Koch, C., Wen, K.-Y., Kreutz, K., & Salner, A. (2003). Assessing the unmet information, support and care delivery needs of men with prostate cancer. *Patient Education and Counselling*, 49, 233 - 242.
- Cancer Australia (2014). *Recommendations for the identification and management of fear of cancer recurrence in adult cancer survivors*. Retrieved from the Australian Government Cancer Australia website http://guidelines.canceraustralia.gov.au/guidelines/fear_of_recurrence/ch01.php
- Department of Health. (1997). *The new NHS: Modern, Dependable*. Retrieved from, <http://www.archive.official-documents.co.uk/document/doh/newnhs/contents.htm>.
- Department of Health. (2011). *NHS reference costs 2010 to 2011*. Retrieved from, <https://www.gov.uk/government/publications/nhs-reference-costs-2012-to-2013>
- Department of Health (2012). The NHS Outcomes Framework 2013-2014. https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/213055/121109-NHS-Outcomes-Framework-2013-14.pdf
- Department of Health (2013). *NHS reference Costs 2012 to 2013*. Retrieved from <https://www.gov.uk/government/publications/nhs-reference-costs-2012-to-2013>
- Giuffrida, A., & Torgerson, D. J. (1997). Should we pay the patient? Review of financial incentives to enhance patient compliance. *BMJ*, 315:303, doi: <http://dx.doi.org/10.1136/bmj.315.7110.703>
- Greenberg, D. B., Kornblith, A. B., Herndon, J. E., et al (1997). Quality of life for adult leukaemia survivors treated on clinical trials of Cancer and Leukaemia Group B during the period 1971-1988: predictors for later psychological distress. *Cancer*, 80(10), 1936 – 1944.
- Greer, J. A., Solis, J. M., Temel, J. S., Lennes, I. T., Prigerson, H. G., Maciejewski, P. K., Pirl, W. F. (2011). Anxiety disorders in long-term survivors of adult cancers. *Psychosomatics*, 52, 417-423.

Holmes, L. (2012) *Addressing the consequences of treatment in Gynaecological cancer patients. Care Integration Awards*. Retrieved from <http://www.ncsi.org.uk/wp-content/uploads/Article1-doc-final-version-Gynae-oncology-CNS-Lynn-Holmes.pdf>

International Society for Pharmacoeconomics and Outcome research. (2011). *Pharmacoeconomic Guidelines around the World*. Retrieved from <http://www.ispor.org/peguidelines/countrydet.asp?c=23&t=2>

Kerr-Wilson, R. H., & McCrum, A. (1995). Follow-up of patients with gynaecological cancer. *Australian and New Zealand Journal of Obstetrics and Gynaecology*, 35, 298-299.

Kew, F. M., Galaal, K., Maderville, H. (2009). Patients' views of follow-up after treatment for gynaecological cancer. *Journal of Obstetrics and Gynaecology*, 29, 135 – 142.

Kimman, M. L., Dellaert, B., Boersma, L. J., Lambin, P., & Dirksen, C. D. (2010). Follow-up after treatment for breast cancer: one strategy fits all? An investigation of patient preferences using a discrete choice experiment. *Acta Oncologica*, 49, 328 – 337.

Kocur, G., Adler, T., Hyman, W., & Aunmet, B. (1982). Guide to Forecasting Travel Demand with Direct Utility Assessment. United States Department of Transportation, Urban Mass Transportation Administration, Report UMTA-NH-11-0001-82-1, Washington DC.

Kornblith, A. B., Powell, M., Regan, M. M., et al (2007). Long-term psychosocial adjustment of older vs younger survivors of breast and endometrial cancer. *Psycho-oncology*, 16 (10), 895 – 903.

Lancaster, K. (1996). A new approach to consumer theory. *Journal of Political Economy*, 74, 132 – 157.

Leeson, S., Jones, J., Stuart, N. A., Whitaker, R., Edwards, R. T., Hall, L., Neal, R., Sylvestre, Y., Yeo, S. T., Morrison, V. (2012). Research for Patient and Public Benefit Wales: A trial of optimal personalised care after treatment for gynaecological cancer (TOPCAT –G). NISCHR, Welsh Government.

Leeson, S., Stuart, N., Sylvestre, Y., Hall, L., & Whitaker, R. (2013). Gynaecological cancer follow-up audit of current practice: national survey of current practice in the UK. *British Medical Journal open*, 3, e002859. doi:10.1136/bmjopen-2013-002859.

Lydon, A., Beavery, K., Newbury, C., Wray, J. (2009). Routine follow-up after treatment for ovarian cancer in the United Kingdom (UK): patient and health professional views. *European Journal of Oncology Nursing*, 13, 336 – 343. doi: 10.1016/j.ejon.2009.04.007

Macmillan Cancer Support (2013). Throwing light at the consequences of cancer and its treatment. Retrieved from <http://www.macmillan.org.uk/Documents/AboutUs/Research/Researchandevaluationreports/Throwinglightontheconsequencesofcanceranditstreatment.pdf>

Maddams, J., Parkin, D. M., & Darby, S. (2011). The cancer burden in the United Kingdom in 2007 due to radiotherapy. *International Journal of Cancer*, 129, 2885-2893.

Mirabeau-Beale, K., Kornblith, A., Penson, R., et al (2009). Comparison of the quality of life of early and advanced stage ovarian cancer survivors. *Gynecol Oncol*, 114(2), 353 – 359.

Morris, S., Delvin, N., & Parkin, D. (2007). *Economic analysis in Health Care*. John Wiley & Sons: Chichester, West Sussex.

Nama, V., Nordin, A., Bryant, A. (2013). Patient related outcome measures for follow-up after gynaecological cancer treatment (Protocol). *The Cochrane collaboration*. Doi: 10.1002/14651858.CD010299.

National Cancer Intelligence Network. (2006). *One, five, and ten year cancer prevalence by cancer network, UK, 2006*. Retrieved from www.ncin.org.uk/view?rid=76

National Cancer Survivorship Initiative (2013a). *Living With and Beyond Cancer: Taking Action to Improve Outcomes*. Retrieved from <http://www.ncsi.org.uk/wp-content/uploads/howtoguide.pdf>

National Cancer Survivorship Initiative (2013b). Stratification process. Retrieved from <http://www.ncsi.org.uk/what-we-are-doing/risk-stratified-pathways-of-care/risk-stratification/>

National Health Service Improvement (2013). *Innovation to implementation: Stratified pathways of care for people living with or beyond cancer, a 'how to guide'*. Retrieved from <http://www.ncsi.org.uk/wp-content/uploads/howtoguide.pdf>

National Institute of Clinical Excellence (2005). *Supportive and Palliative Care: The Manual*. Retrieved from, <http://www.nice.org.uk/nicemedia/pdf/csgspmanual.pdf>.

Ng, A. K., & Travis, L. B. (2008). Subsequent neoplasms in cancer survivors. *The Cancer Journal*, 14, 429 – 434.

Office for National Statistics (2013). *Cancer Survival in England: patients diagnosed 2007 – 2011 and followed up to 2012*. Retrieved from, <http://www.ons.gov.uk/ons/rel/cancer-unit/cancer-survival/cancer-survival-in-england--patients-diagnosed-2007-2011-and-followed-up-to-2012/stb-cancer-survival-in-england--patients-diagnosed-2007-2011-and-followed-up-to-2012.html>

Prue, G., Rankin, J., Allen, J., Gracey, J., Cramp, F. (2006). Cancer-related fatigue: A critical appraisal. *European Journal of Cancer*, 42, 846 – 863.

Ryan, M. (2004). Discrete Choice Experiments in Health Care. *British Medical Journal*, 328, 360-361.

Ryan, M., Gerard, K., Amaya – Amaya, M. (2008). *Using Discrete Choice Experiments to Value Health and Health Care*. Springer: Dordrecht, The Netherlands.

Scientific Advisory Committee of the Medical Outcomes Trust (2002). Assessing health status and quality of life instruments: attributes and review criteria. *Quality of life research*, 11, 193 - 205.

Simba, T. (2008). *Patient preferences for breast cancer follow up: A discrete choice experiment*. Unpublished Masters Dissertation, University of Twente, The Netherlands.

Tamburini, M., Gangeri, L., Brunelli, C., Beltrami, E., Boeri, P., Borreani, C., Fusco Karmann, C., Greco, M., Miccinesi, G., Murru, L., Trimigno, P. (2000). Assessment of hospitalised cancer patients' needs by the needs evaluation questionnaire. *Annals of Oncology*, 11, 31 – 37.

Thewes, B., Butow, P., Zachariae, R., Christensen, S., Simard, S., & Gotay, C. (2012). Fear of cancer recurrence: a systematic literature review of self-report measures. *Psycho-oncology*, 21, 571 – 587

The Royal Society of Medicine. (1999). Management of Gynaecological Cancer. *Effective Health Care*, 5(3), ISSN: 0965-0288.

Van den Beuken – van Everdingen, M. H. J., de Rijke, J. M., Kessels, A. G., Schouten, H. C., van Kleef, M., & Patijn, J. (2007). Prevalence of pain in patients with cancer: a systematic review of the past 40 years. *Annals of Oncology*, 18, 1437 – 1449.

Vistad, I., Moy, B. W., Salvesen, H. B., & Livaag, A. H. (2011). Follow-up routines in gynaecological cancer-time for a change? *Acta Obstet Gynecol Scand*, 90, 707 – 718. doi: 10.1111/j.1600-0412.2011.01123.x

Welsh Cancer Intelligence and Surveillance Unit (2015). Cancer in Wales, A summary report of the population cancer incidence, mortality and survival – includes new 2013 data released as Official Statistics. Published February 2015, retrieved from <http://www.wcisu.wales.nhs.uk/documents-1>

Welsh Government (2012). *Together for Health-Cancer Delivery Plan: A delivery plan up to 2016 for NHS Wales and its Partners – The highest standard of care for everyone with cancer.* Retrieved from <http://wales.gov.uk/docs/dhss/publications/120613cancerplan>

Appendix 3.2 Example of participant information sheet (patients)

Preferences for gynaecology cancer follow up care: A questionnaire study Information about the research (patients)

We would like to invite you to take part in my PhD research questionnaire study.

What is the purpose of the study?

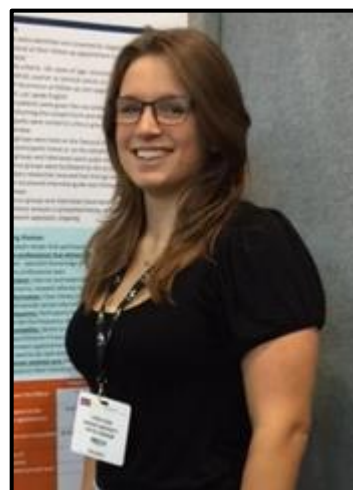
My PhD research aims to look at how gynaecological cancer up care can be improved for patients.

Why have I been invited?

You have been invited to take part in the research study because you have been treated for gynaecological cancer and now having regular check-ups (follow up care). I hope that 100 patients will take part in my PhD research study.

Do I have to take part?

It is up to you to decide whether you would like to take part in the PhD research study. Taking part or not taking part in the study and the answers you give will not affect the care you receive. If you would like to take part please fill in the consent form (on the yellow paper). If you would like more information please return the 'I am interested in the study and/or would like more information about the research study' form (on green paper) or contact me. If you change your mind about



follow

are
about

participating please let me know. Your answers to the questionnaire can be removed from the results up to the point of when I analyse the information from the questionnaires, without giving me an explanation.

What will I be asked to do if I take part?

You will be asked to fill in a questionnaire about the gynaecological cancer follow up care that you have (on the blue paper). There are 3 parts to the research study questionnaire which will take you about 30 minutes to complete. Unfortunately only a small part of the questionnaire is written in Welsh because only some of the questions have been validated in Welsh.

1. The first part asks you a few things about yourself and your cancer experience
2. The second part asks you about how you are feeling
3. The third parts asks you about what kind of follow up care you would like

If you would like help completing the questionnaire you can:

- If you were given the recruitment pack by a research nurse and are reading this and completing the questionnaire as you wait for your appointment you can ask the research nurse for guidance
- If you would prefer or if you were not given the recruitment pack by the research nurse when you were in the waiting room for your appointment you can contact me my phone (01248 382470), by email (l.j.timmis@bangor.ac.uk), or return the 'I am interested in the study and/or would like more information about the research study' form (on green paper) in the FREEPOST envelope.

It is important that you answer all the questions so that we get all the information we need. If you leave some questions blank I might contact you for the answer.

Family and friends often provide a bit of extra support during and/or after patients treatment, for example, taking you to your appointments and supporting you emotionally. These family members and friends are called informal caregivers. It is important to understand what the people that care for patients think.

Please could you ask someone who does act/has acted as an informal caregiver for you to fill in the 'caregiver' questionnaire in the 'caregiver' envelope? Thank you. If you don't want to ask an informal caregiver to take part or you don't think you have an informal caregiver, or if your informal caregiver doesn't want to take part you can still take part. In the 'caregiver' envelope is a similar recruitment pack to this one, and informal caregivers will be asked to read similar documents and complete a similar questionnaire if they wish to take part.

Will I get paid?

People who return the completed recruitment pack will be entered into a prize draw to receive one of two £50 or £25 vouchers. The winner will be chosen at random.

What are the possible disadvantages of taking part?

Filling in the questionnaires may make you think about the cancer you had and how you are feeling now. If you would like to talk to someone, please contact your GP or seek support from Tenovus Cancer Care Freephone support line, open from 8am – 8pm 7 days a week 0808 808 1010.

What are the benefits of taking part?

We cannot promise the study will help you but we hope the results will help improve gynaecological cancer follow up care for patients in the future.

Will me taking part in the study and my information be kept confidential?

With your permission we will tell your GP that you are taking part in the study. With your permission we will give Tenovus Cancer Care your contact details so that they can tell see if there is any way they can support you (if you do not agree to this, you are still able to take part in the study). Your information will not be passed to anyone else. However, if you tell us that harm has been or might be caused to someone, we are required by law to inform the appropriate authority.

What if there is a problem?

If you have a complaint about the research study please contact the school manager Mr Hefin Francis via phone 01248 38 2211 or email h.francis@bangor.ac.uk.

Who has reviewed the study?

The PhD research study has been approved by Bangor University and the NHS ethics and R&D committee.

Who has organised and funded the research?

I (Laura Timmis) have organised the research as part of my PhD project. My PhD project is funded by Tenovus Cancer Care.

What will happen to the results?

The results will be published in an academic journal. We hope that the results will be used to improve gynaecological cancer follow up care for patients. Please let me know if you would like me to send you an overview of the results in approximately a year's time.

Thank you very much for taking the time to read this information

- If you are able to help me in my PhD research study please fill in the consent form (on yellow paper) and questionnaire booklet (on blue paper) and return it to me in the FREEPOST envelope.
- If any of the questions are not clear or you would like to talk to me about the research study please complete the 'Request for more information' sheet (on green paper), email me l.j.timmis@bangor.ac.uk, or call me 01248 382470.

Appendix 3.3 Example of consent form (patients and informal caregivers)

Preferences for gynaecology cancer follow up care: A questionnaire study

Consent Form

	Please initial the box
I confirm that I have read and understand the information sheet dated 26/10/15 (version 2) for the above study. I have had the opportunity to consider the information, ask questions, and have had any questions answered satisfactorily.	<div><i>Timmis</i></div> <input type="checkbox"/>
I understand that my participation is voluntary and that I can request for my data to be withdrawn up to the point of when the information from the questionnaires is analysed without explanation	<input type="checkbox"/>
I understand that my participation and the data I provide is strictly confidential. However, I acknowledge that if I disclose harm to an individual that by law, this information has to be reported to the appropriate authority.	<input type="checkbox"/>
I agree for my GP to be informed that I am taking part in the study	<input type="checkbox"/>
I understand that relevant data collected during the study may be looked at by individuals from the research team, and for the purpose of audit and monitoring, by members of the auditing and monitoring team at Bangor University and BCUHB. I give permission for these individuals to have access to my data for this purpose.	<input type="checkbox"/>
I agree to take part in the above study	<input type="checkbox"/>
I consent to Tenovus Cancer Care being given my contact details so that they can contact me, to see if they can offer me any support (if you do not agree to this, you are still able to take part in the study)	<input type="checkbox"/>
Please send me a copy of the overview of the results	<input type="checkbox"/>

Name of Participant _____ **Date** _____ **Signature** _____

Name of Researcher Laura Timmis **Date** 07/12/15 **Signature** _____

Please return ONE consent form, and keep the other consent form for your own records

Preferences for gynaecology cancer follow up care: A questionnaire study

Questionnaire

Please complete all 3 sections of this questionnaire. The questionnaire will take approximately 20 minutes to complete.

Part 1: A bit about you, your cancer journey, and the people that support you

Part 2: How are you?

Part 3: Your opinions/preferences for gynaecology cancer follow up

Part 1: A bit about you, your cancer journey, and the people that support you

24. What is your name?

25. What is your address?
.....
.....
.....
.....

26. What is your phone number?
.....

27. What is your age?

- | | | |
|---|--|--------------------------|
| 28. What is your employment status? Are you currently.... | Employed for wages | <input type="checkbox"/> |
| | A volunteer | <input type="checkbox"/> |
| | Self-employed | <input type="checkbox"/> |
| | Out of work and looking for work | <input type="checkbox"/> |
| | Out of work but not currently looking for work | <input type="checkbox"/> |
| | A homemaker | <input type="checkbox"/> |
| | A full time Carer | <input type="checkbox"/> |
| | A student | <input type="checkbox"/> |
| | Military | <input type="checkbox"/> |
| | Retired | <input type="checkbox"/> |
| Unable to work | <input type="checkbox"/> | |

29. Who do you live with?

Alone ☐

My Husband/wife ☐

My partner ☐

Family ☐

Friends ☐

Other ☐

Please describe.....

30. What is your total income? Asking this question is very personal, but it is important to answer it as it relates to the last questionnaire in the questionnaire booklet.

Please:

- Do not deduct tax, National Insurance, any health insurance payments, all your contributions to occupational pension schemes
- Do not count loans because they are not income
- If you have a joint income, please provide the share that you receive
- If you receive a pension or benefits this is your income

Per Week

Per Year

Nil or loss

☐

Nil or less

£1 to £79

☐

£1 to £3,999

£80 to £149

☐

£4,000 to £7,999

£150 to £229

☐

£8,000 to £11,999

£230 to £329

☐

£12, 000 to £16,999

£330 to £459

☐

£17,000 to £23,999

£460 to £709

☐

£24,000 to £36,999

£710 or more

☐

£37,000 or more

31. Is this your first diagnosis of this type of cancer?

Yes ☐

No (please explain) ☐

32. What form of gynaecological cancer have you been treated for?

Endometrial (uterus or womb) cancer

☐

Cervical cancer

☐

Ovarian cancer

☐

Vulval Cancer

☐

Other

☐

(please state)

33. What stage of cancer did you have?
(If you do not know, please write 'I do not know')

.....

34. Has the cancer that you had come back?

Yes

☐

No

☐

If yes, how did you realise the cancer had come back? *Please tick all that apply*

I had symptoms that the cancer had come back

☐

I could just tell that something wasn't right

☐

In my follow up appointment I had examinations/tests which showed the cancer had come back

☐

I spoke to the specialist nurse because I was worried it had come back/I asked for an extra appointment

☐

I waited till my next scheduled appointment to tell the

Doctor I was worried the cancer had come back

☐

Other (please describe)

☐

35. What treatment did you receive? *Please tick all that apply*
(If the cancer has come back and you have received two sets of treatment, this question relates to both sets of treatment)

Surgery

☐

Chemotherapy

☐

Radiotherapy

☐☐

Brachytherapy

Other

☐

Please state

36. How long have you been receiving gynaecological cancer follow up care for? (If the cancer has come back, this is the amount of time you have had follow up appointments for since your second treatment)

Less than 6 months

☐

Between 6 months – 1 year

☐

Between 1 year – 2 years

☐

Between 2 years – 3 years

☐

Between 3 years – 4 years

☐

Between 4 years – 5 years

☐

Over 5 years

☐

37. How many gynaecological cancer follow up appointments have you had? (If the cancer has come back, this is the number of follow up appointments you have had since your second treatment)

.....

38. Where do you receive your follow up appointments? (If the cancer has come back, this is the location of appointments since your second treatment)

At the hospital I was treated at

☐

At a local, community hospital

☐

At my GP surgery

☐

Over the phone

☐

I don't have regular follow up appointments, I only see a nurse or consultant if I am worried about anything

☐

39. Who delivers your follow up appointment?
(If the cancer has come back, this is the person that delivers your follow up appointments since your second treatment)

The consultant oncologist

☐

The consultant gynaecologist

☐

A specialist nurse

☐

My GP

☐

Radiologist

☐

Alternates between the and ☐

Other (*Please state*)

☐

.....
.....

40. Currently, how often do you have follow up appointments?
(If the cancer has come back, this is the frequency of appointments since your second treatment)

Every 3 months

☐

Every 6 months

☐

Every 12 months

☐

As and when I request one

☐

41. What happens in your follow up appointment? *Please tick all that apply*
(If the cancer has come back, what happens in the follow up appointments you have had since your second treatment?)

I have an internal examination

☐

I have an external examination

☐

I have medical tests

☐

Which medical tests do you have?

.....

I am asked how I am feeling

☐

I am given a needs assessment to look at my

☐

☐

physical, psychological (mental), social and financial needs

I am referred to other services that can help me

☐

I have the opportunity to ask questions

☐

42. Are you taking part in any other gynaecology cancer research studies?

Yes

☐

No

☐

If yes, which research study are you taking part in:

.....

43. Who have you nominated as your informal carer for this research? An informal caregiver is a family member or friend who provides additional support or care for someone, due to an illness or disability they are experiencing.

.....

44. What is your relationship to the person you have nominated as your informal caregiver? They are my.....

Spouse

☐

Parent

☐

Sibling

☐

Friend

☐

Other

☐

Please state:

45. How long do you feel this person has been providing care beyond that of 'spouse' 'sibling' 'friend' etc?

.....

46. Do you live with this person?

Yes

☐

No

☐

47. What is the name and address of your GP?

Name: _____

Address: _____

Please turn over.

The next part of the questionnaire asks you about how you are feeling.

Part 2: We are interested in some things about you and your health. Please answer all of the questions yourself by circling the number that best applies to you

Under each heading, please tick the ONE box that best describes your health TODAY.

MOBILITY

- I have no problems in walking about ☐
- I have slight problems in walking about ☐
- I have moderate problems in walking about ☐
- I have severe problems in walking about ☐
- I am unable to walk about ☐

SELF-CARE

- I have no problems washing or dressing myself ☐
- I have slight problems washing or dressing myself ☐
- I have moderate problems washing or dressing myself ☐
- I have severe problems washing or dressing myself ☐
- I am unable to wash or dress myself ☐

USUAL ACTIVITIES (e.g. work, study, housework, family or leisure activities)

- I have no problems doing my usual activities ☐
- I have slight problems doing my usual activities ☐
- I have moderate problems doing my usual activities ☐
- I have severe problems doing my usual activities ☐
- I am unable to do my usual activities ☐

PAIN / DISCOMFORT

- I have no pain or discomfort ☐
- I have slight pain or discomfort ☐
- I have moderate pain or discomfort ☐
- I have severe pain or discomfort ☐
- I have extreme pain or discomfort ☐

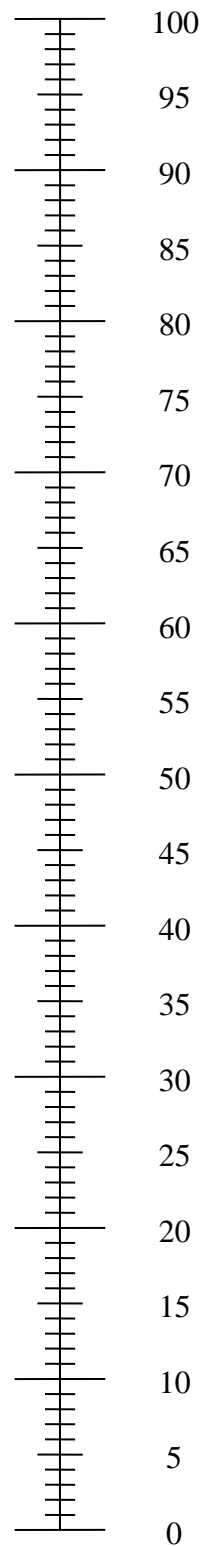
ANXIETY / DEPRESSION

- I am not anxious or depressed ☐
- I am slightly anxious or depressed ☐
- I am moderately anxious or depressed ☐
- I am severely anxious or depressed ☐
- I am extremely anxious or depressed ☐

- We would like to know how good or bad your health is TODAY.
- This scale is numbered from 0 to 100.
- 100 means the best health you can imagine.
0 means the worst health you can imagine.
- Mark an X on the scale to indicate how your health is TODAY.
- Now, please write the number you marked on the scale in the box below.

YOUR HEALTH TODAY =

The best health you
can imagine



The worst health
you can imagine



EORTC QLQ-C30

(version 3)

		Not at all	A little	Quite a Bit	Very Much
1	Do you have any trouble doing strenuous activities, like carrying a heavy shopping bag or a suitcase?	1	2	3	4
2	Do you have any trouble taking a <u>long</u> walk?	1	2	3	4
3	Do you have any trouble taking a <u>short</u> walk outside of the house?	1	2	3	4
4	Do you need to stay in bed or a chair during the day?	1	2	3	4
5	Do you need help with eating, dressing, washing yourself or using the toilet?	1	2	3	4
During the past week:		Not at all	A little	Quite a Bit	Very Much
6	Were you limited in doing either your work or other daily activities?	1	2	3	4
7	Were you limited in pursuing your hobbies or other leisure time activities?	1	2	3	4
8	Were you short of breath?	1	2	3	4
9	Have you had pain?	1	2	3	4
10	Did you need to rest?	1	2	3	4
11	Have you had trouble sleeping?	1	2	3	4
12	Have you felt weak?	1	2	3	4
13	Have you lacked appetite?	1	2	3	4
14	Have you felt nauseated?	1	2	3	4

Please go onto the next page

15	Have you vomited?	1	2	3	4
16	Have you been constipated?	1	2	3	4
17	Have you had diarrhoea?	1	2	3	4
18	Were you tired?	1	2	3	4
19	Did pain interfere with your daily activities?	1	2	3	4
20	Have you had difficulty in concentrating on things, like reading a newspaper or watching television?	1	2	3	4
21	Did you feel tense?	1	2	3	4
22	Did you worry?	1	2	3	4
23	Did you feel irritable?	1	2	3	4
24	Did you feel depressed?	1	2	3	4
25	Have you had difficulty remembering things?	1	2	3	4
26	Has your physical condition or medical treatment interfered with your <u>family</u> life?	1	2	3	4
27	Has your physical condition or medical treatment interfered with your <u>social</u> activities?	1	2	3	4
28	Has your physical condition or medical treatment caused you financial difficulties?	1	2	3	4

Please go onto the next page

For the following questions please circle the number between 1 and 7 that best applies to you

29 How would you rate your overall health during the past week?

1 2 3 4 5 6 7

Very poor

Excellent

30 How would you rate your overall quality of life during the past week?

1 2 3 4 5 6 7

Very poor

Excellent

For each question please circle the statement that you agree with.

1) I worry about future diagnostic tests

Not at all

A little bit

Somewhat

Very Much

2) I worry about another type of cancer

Not at all

A little bit

Somewhat

Very Much

3) I worry about my cancer coming back

Not at all

A little bit

Somewhat

Very Much

Guidance for Part 3 of questionnaire

Aspects

The types of gynaecological cancer follow up are described by the following aspects:

➤ Personalised care

This describes having your care personalised to your physical and mental needs, with referrals to other services to support you if you need this.

➤ Information received

This describes the amount of information you receive. Brief information means a one page sheet about the signs and symptoms of the cancer coming back and the contact details for your key worker/specialist nurse. Comprehensive information means a ten page booklet that also tells you about the physical and mental effects of the cancer for you and your informal caregiver, how you can make changes to your lifestyle to reduce the risk of the cancer coming back, and a list of local support groups and charities that can help you and your informal caregiver.

➤ Medical examination

Patients have an examination to check how things are internally, and to see if there are any signs of the cancer coming back. This aspect describes whether you would like an examination if it can/cant detect whether the cancer has come back if you have no symptoms.

➤ Health care professional

This aspect describes who you would like to see for your follow up care appointments. Either the GP, specialist nurse, consultant or different professionals in the multidisciplinary team depending on your needs (e.g. the consultant and community nurse, or the specialist nurse and a charity support worker)

➤ Frequency of care

This describes how often you would like to have gynaecological cancer follow up appointment

Describing gynaecological cancer follow up

Five main aspects of follow up are presented. Each aspect has more than one choice. In the questionnaire please assume that any aspects of follow up care that are not described here, do not change. For example, the length of time the person you spend in your appointment. To help you complete Part 3 in the questionnaire, please read the descriptions of the aspects and possible choices below.

Aspects	Possible Choices
Personalised care	I have the same medical care as everyone else who has had gynaecological cancer
	The care I receive is specific to my physical and mental needs
Information received	I receive basic information
	I receive detailed information
Medical examination	I have an internal examination even if it can't detect whether the cancer has come back. I don't have any symptoms
	I have an internal examination if it can detect whether the cancer has come back. I don't have any symptoms
Health care professional seen	I see the GP for my appointments
	I see the specialist nurse for my appointments
	I see the consultant for my appointments
	I see different professionals in the multidisciplinary team for my appointments depending on my needs
Frequency of appointments	I have an appointment every 12 months
	I have an appointment every 6 months

Part 3: Preferences for gynaecology cancer follow up

In this section, we would like you to think about your gynaecology cancer follow up. You will be presented with 9 questions that describe two different types of follow up.

For each question, please tick whether you would prefer 'Follow up Care A' or 'Follow up Care B'. There is no right or wrong answer.

Question 1

Please tick ONLY ONE GREEN BOX below for which service you would prefer

Follow up A		Follow up B
The care I receive is specific to my physical and mental needs		I have the same medical care as everyone else who has had gynecological cancer
I receive comprehensive information		I receive basic information
I have an internal examination if it can detect whether the cancer has come back. I don't have any symptoms		I have an internal examination even if it can't detect whether the cancer has come back. I don't have any symptoms
I see the GP for my appointments		I see the specialist nurse for my appointments
I have an appointment every 6 months		I have an appointment every 12 months

Tick here if you prefer Follow up A <input type="checkbox"/>	OR	Tick here if you prefer Follow up B <input type="checkbox"/>
--	----	--

Question 2

Please tick ONLY ONE GREEN BOX below for which service you would prefer

Follow up A	Follow up B
The care I receive is specific to my physical and mental needs	I have the same medical care as everyone else who has had gynecological cancer
I receive comprehensive information	I receive basic information
I have an internal examination if it can detect whether the cancer has come back. I don't have any symptoms	I have an internal examination even if it can't detect whether the cancer has come back. I don't have any symptoms
I see the GP for my appointments	I see the specialist nurse for my appointments
I have an appointment every 6 months	I have an appointment every 12 months

Tick here if you prefer Follow up A	<input type="checkbox"/>	OR	Tick here if you prefer Follow up B	<input type="checkbox"/>
-------------------------------------	--------------------------	----	-------------------------------------	--------------------------

Question 3

Please tick **ONLY ONE GREEN BOX** below for which service you would prefer

Follow up A	Follow up B
I have the same medical care as everyone else who has had gynaecological cancer	The care I receive is specific to my physical and mental needs
I receive basic information	I receive comprehensive information
I have an internal examination if it can detect whether the cancer has come back. I don't have any symptoms	I have an internal examination even if it can't detect whether the cancer has come back. I don't have any symptoms
I see the specialist nurse for my appointments	I see the consultant for my appointments
I have an appointment every 6 months	I have an appointment every 12 months

Tick here if you prefer Follow up A <input type="checkbox"/>	OR	Tick here if you prefer Follow up B <input type="checkbox"/>
--	-----------	--

Question 4

Please tick ONLY ONE GREEN BOX below for which service you would prefer

Follow up A	Follow up B
The care I receive is specific to my physical and mental needs	I have the same medical care as everyone else who has had gynaecological cancer
I receive comprehensive information	I receive basic information
I have an internal examination even if it can't detect whether the cancer has come back. I don't have any symptoms	I have an internal examination if it can detect whether the cancer has come back. I don't have any symptoms
I see the specialist nurse for my appointments	I see the consultant for my appointments
I have an appointment every 12 months	I have an appointment every 6 months

Tick here if you prefer Follow up A <input type="checkbox"/>	OR	Tick here if you prefer Follow up B <input type="checkbox"/>
--	----	--

Question 5

Please tick ONLY ONE GREEN BOX below for which service you would prefer

Follow up A
I have the same medical care as everyone else who has had gynaecological cancer
I receive comprehensive information
I have an internal examination even if it can't detect whether the cancer has come back. I don't have any symptoms
I see the consultant for my appointments
I have an appointment every 6 months

Follow up B
The care I receive is specific to my physical and mental needs
I receive basic information
I have an internal examination if it can detect whether the cancer has come back. I don't have any symptoms
I see different people in the multidisciplinary team for my appointments depending on my needs
I have an appointment every 12 months

Tick here if you prefer Follow up A	<input type="checkbox"/>	OR	Tick here if you prefer Follow up B	<input type="checkbox"/>
-------------------------------------	--------------------------	----	-------------------------------------	--------------------------

Question 6

Please tick ONLY ONE GREEN BOX below for which service you would prefer

Follow up A	Follow up B
The care I receive is specific to my physical and mental needs	I have the same medical care as everyone else who has had gynaecological cancer
I receive basic information	I receive comprehensive information
I have an internal examination if it can detect whether the cancer has come back. I don't have any symptoms	I have an internal examination even if it can't detect whether the cancer has come back. I don't have any symptoms
I see the consultant for my appointments	I see different people in the multidisciplinary team for my appointments depending on my needs
I have an appointment every 12 months	I have an appointment every 6 months

Tick here if you prefer Follow up A <input type="checkbox"/>	OR	Tick here if you prefer Follow up B <input type="checkbox"/>
--	----	--

Question 7

Please tick ONLY ONE GREEN BOX below for which service you would prefer

Follow up A	Follow up B
I have the same medical care as everyone else who has had gynaecological cancer	The care I receive is specific to my physical and mental needs
I receive comprehensive information	I receive basic information
I have an internal examination if it can detect whether the cancer has come back. I don't have any symptoms	I have an internal examination even if it can't detect whether the cancer has come back. I don't have any symptoms
I see different people in the multidisciplinary team for my appointments depending on my needs	I see the GP for my appointments
I have an appointment every 12 months	I have an appointment every 6 months

Tick here if you prefer Follow up A <input type="checkbox"/>	OR	Tick here if you prefer Follow up B <input type="checkbox"/>
--	----	--

Question 8

Please tick ONLY ONE GREEN BOX below for which service you would prefer

Follow up A	Follow up B
The care I receive is specific to my physical and mental needs	I have the same medical care as everyone else who has had gynaecological cancer
I receive basic information	I receive comprehensive information
I have an internal examination even if it can't detect whether the cancer has come back. I don't have any symptoms	I have an internal examination if it can detect whether the cancer has come back. I don't have any symptoms
I see different people in the multidisciplinary team for my appointments depending on my needs	I see the GP for my appointments
I have an appointment every 6 months	I have an appointment every 12 months

Tick here if you prefer Follow up A <input type="checkbox"/>	OR	Tick here if you prefer Follow up B <input type="checkbox"/>
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Question 9

Please tick ONLY ONE GREEN BOX below for which service you would prefer

Follow up A	Follow up B
The care I receive is specific to my physical and mental needs	I have the same medical care as everyone else who has had gynecological cancer
I receive comprehensive information	I receive basic information
I have an internal examination if it can detect whether the cancer has come back. I don't have any symptoms	I have an internal examination even if it can't detect whether the cancer has come back. I don't have any symptoms
I see the GP for my appointments	I see the specialist nurse for my appointments
I have an appointment every 6 months	I have an appointment every 12 months

Tick here if you prefer Follow up A <input type="checkbox"/>	OR	Tick here if you prefer Follow up B <input type="checkbox"/>
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Question 10

We are interested in finding out how much you value your gynaecological cancer follow up care appointments. Your answers will be useful to inform the government in making decisions about how resources are allocated within the NHS. **This question is completely hypothetical.**

Imagine that there is no NHS, so you pay less in tax and national insurance contributions. In return, you must pay for every medical service out of your own pocket. As you have known this for a long time, over time you have saved the equivalent of a year's income so that you are prepared for any medical costs.

The amount you would be willing to pay for your gynaecological cancer follow up care *if you had to*, gives an indication *of how much you value your gynaecological cancer follow up care appointments* compared with other things you might want to spend your money on.

Please tick the appropriate box if you would be willing to pay this amount each year, not willing to pay this amount each year, or not sure if you would be willing to pay this amount each year for your gynaecological cancer follow up care appointments, if there was no NHS, and if you had the equivalent of one year's income in your savings account?

	Willing to pay this amount each year	Not willing to pay this amount each year	Not sure if I would be willing to pay this amount each year
£50			
£75			
£100			
£125			
£150			
£175			
£200			
£225			
£250			
£275			
£300			

£500			
£1000			

We are interested in what would be the *highest* amount you would pay each year for your gynaecological cancer follow up care appointments, if there was no NHS, and if you had the equivalent of one year's income in your savings account?

£ _____