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DOCTOR OF PHILOSOPHY

Patient preferences in the delivery of cancer genetic services

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Award date:
2008

Awarding institution:
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PATIENT PREFERENCES IN THE DELIVERY OF CANCER GENETIC SERVICES

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SUMMARY

The NHS funded Cancer Genetics Service in Wales (CGSW) offers a resource intensive model of care, offering high risk patients multiple counselling sessions and genetic testing. Given that there is no information upon the preferences of patients for the manner in which the Welsh model of providing cancer genetic services is delivered, or data on the associated costs, research question 1 was developed. Research question 1, "What are the attributes of cancer genetics services that are important to high risk patients (the patients spending the most time in contact with the service and receiving most services i.e. genetic testing and counselling)? and what would be the cost of providing the service to comply with patient preferences?" Having examined the literature on eliciting patient preferences the relatively new and experimental technique of discrete choice modelling (DCM) was identified as the most appropriate one to use to elicit the data required to answer research question 1. The health economics literature revealed that no one had experimentally examined DCM's underlying decision theory principal of random utility theory (RUT) in conjunction with a DCM exercise. To supplement the deficiency in the health economics literature, the decision theory/psychology literature was accessed. The large body of literature on utility theory revealed that the descriptive ability of utility theory was in question. These findings resulted in research question 2, "Do respondents of DCM questionnaires make choices in accordance with Random Utility Theory?"

Empirical Aims

Experiment

- Experimentally examine respondents of a DCM exercise by means of an information manipulation to see if they are adhering to DCM's underlying decision theory principals of Random Utility Theory.

Patient survey

- Ascertain the aspects of cancer genetics services that are important to patients, and present service configurations prioritised in terms of preferences accompanied by their costs (cost-consequences analysis) for high risk patients.

Methods

Experiment

A repeated measures information manipulation experiment was conducted with a sample of 142 first year undergraduate students at the University of Wales, Bangor.

Patient survey

Patient preferences were gathered by issuing self administered discrete choice questionnaires to 30 high risk patients of the CGSW following their genetic risk assessment. Costs were estimated for the Cardiff clinic of the CGSW by administering a questionnaire to all staff, conducting an audit of clinic rooms and equipment and obtaining gross unit costs from the finance department.

Results & Conclusions

Experiment

The primary conclusion of the experiment was that respondents were not making choices in accordance with random utility theory. There is clearly a need to conduct further research into RUT as soon as possible as until the uncertainty relating to the descriptive validity of RUT is resolved DCM and other RUT based techniques are potentially invalid.

Patient survey

Given the results of the experiment, the results of the DCM survey with patients of the CGSW must be interpreted with caution. Counselling by a genetics associate accompanied by favourable levels of other attributes provided high utility and also provided substantial cost savings. These findings support the use of genetics associates for genetic counselling in response to the scarcity of qualified consultant clinical geneticists. The savings obtained from such a service configuration can be redirected to fund improvements in the service such as more staff (clinical and administrative) to reduce the waiting time between

receipt of referral and issuing patients with a risk assessment or be used in relation to other attributes or completely different health services.

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PREFACE

In 1999 the MacMillan Cancer Relief and the National Assembly for Wales required evidence upon which to base their decision of whether or not to continue to fund the provisional Cancer Genetics Service in Wales (formerly known as the All-Wales Cancer Genetics Service). Prof. Clare Wilkinson, North Wales Section of the Department of General practice (NWDGP), University of Wales College of Medicine and Dr. Rhiannon Tudor Edwards, Centre for Economics and Policy in Health (CEPHi), Institute of Medical and Social Care Research (IMSCaR), University of Wales, Bangor (U.W.B.) were commissioned to evaluate the service.

I (Gethin Griffith) was employed part time (0.8 WTE) as the principal health economic investigator on the evaluation. During the Job interview it emerged that this project would provide the ideal opportunity for me to pursue a part time Ph.D. under the supervision of Prof. J. M. G. Williams (Mark Williams; Director of IMSCaR) and Dr. Rhiannon Tudor Edwards (Director of CEPHi).

An agreement was reached with the health outcomes team [Prof. Clare Wilkinson (NWDGP), Prof. Paul Bennett (Psychology Department, University of Wales, Swansea), Dr. Jim Turner (NWDGP) and Ms. Barbara France (NWDGP)] to allow a questionnaire devoted to this Ph.D. thesis to be included with the questionnaires they were issuing to a cohort of patients referred to the CGSW. The evaluation entitled "All Wales Cancer Genetics Service Evaluation", short title "The GenQuest Evaluation" was successfully conducted between the 1st of July 1999 and the 30th of June 2002.

To date three papers have been published from this research. Two of which were designed as overlapping elements of the GenQuest evaluation and the Ph.D. thesis (papers 1 and 3 below) and one which was conducted purely as part of the GenQuest evaluation (paper 2).

1) Griffith, G. L., Edwards, R. T. and Gray, J. (2004). Cancer genetic services: a systematic review of the economic evidence and issues. *British Journal of Cancer 90, 9*, 1697-1703.

2) Griffith, G. L., Edwards, R. T., Gray, J., Wilkinson, C., Turner, J., France, B. and Bennett, P. (2004). Estimating the survival benefits gained from providing national cancer genetic services to women with a family history of breast cancer. *British Journal of Cancer 90, 10*, 1912-1919.

3) Griffith, G. L., Edwards, R. T., Gray, J., Butler, R., Wilkinson, C., Turner, J., France, B. and Bennett, P. (2005). A micro costing of NHS cancer genetic services. *British Journal of Cancer, 92, 1*, 60-71.

All other elements of this research are independent of any organisation other than the University of Wales, Bangor.

During the course of conducting this research Prof. Mark Williams left IMSCaR and took up a post with The Department of Psychiatry at the University of Oxford and Dr. Val Morison (Lecturer in Health Psychology, School of Psychology, U.W.B.) took over as the first supervisor on the Ph.D. On the retirement of Mr. Bruce Napier (Associate Director of IMSCaR), Prof. Ian Russell (Director of IMSCaR) took over as chair of the thesis committee.

ACKNOWLEDGEMENTS

I am grateful to all the patients and staff of the Cancer Genetics Service in Wales (CGSW) that participated in this research. In particular I would like to thank Ms. Elisabeth France, Ms. Rachel Butler and Ms. Sarah Maund of the Cancer Genetics Service in Wales, Mr. Andrew Lycett and Ms. Jacqueline Miller of the Cardiff and Vale NHS Trust, and Ms. Caroline Vanluttmer of Grant Thornton (formerly Cardiff and Vale NHS Trust). I would like to thank the GenQuest research team and their funders the MacMillan Cancer Relief and the National Assembly for Wales for allowing me access to the GenQuest data and issuing my questionnaire to patients.

I appreciate the assistance of Ms. Caroline White-Gwenin, Ms. Abbie Unwin and Ms. Nonn a'ch Dafydd (who were 3rd year students at the time and are now graduates of the School of Psychology, University of Wales Bangor [U.W.B.]) in conducting my experimental work with the samples of 1st year undergraduate students. I would also like to thank the undergraduate students of the School of Psychology (U.W.B.) who participated in the experiment.

I am indebted to Dr. Jonathan Gray (Welsh Assembly Government, seconded from CGSW) and Dr. Daphne Russell (Wales Organisation for Randomised Trials for Health, IMSCaR, U.W.B), both of whom have generously given of their scarce time and valuable expertise in the respective fields of medical genetics and statistics.

I would like to thank Mr. Bruce Napier and Prof. Ian Russell for chairing my research reviews. Finally, I would like to express my thanks to my supervisors, Dr. Val Morrison, Prof. Mark Williams and Dr. Rhiannon Tudor Edwards, for their prompt comments, advice, and tireless encouragement and enthusiasm.

DEFINITIONS

As this thesis deals with literature and concepts from a range of disciplines including health economics, health psychology, marketing and medical genetics amongst others, a set of definitions have been included. These brief definitions outline the primary meaning of the terms and how they are used in this thesis.

Table i

Terms and Definitions

Term	Definition
Attributes	Aspects or characteristics of a commodity or service.
Attribute levels	The subdivisions of attributes e.g. hours and minutes can be used as the levels for the attribute waiting time.
Cardinal utility	Utility that is quantifiable and has ratio data properties. See also interval and ratio data.
Conjoint analysis	A method of preference elicitation and data analysis. Responses are gathered via ranking, rating or discrete choice.
Construct validity	Construct validity is the extent to which the instrument is measuring or testing the intended theory or construct. Construct validity is most commonly addressed in the form of concurrent (convergent and discriminant) validity, criterion and theoretical validity.
Continuous data	Data measured at the ordinal or interval/ratio level.
Continuum model	A model with a continuous process or successive gradations (Reber, 1995).
Continuous preference	Improvement in one attribute can compensate for a reduction in another (Ryan, 1999b).
Concurrent/Convergent validity	The degree of agreement between different measures or tests that measure the same construct.
Criterion validity	See predictive validity.
Data coding	Transforming respondents responses into numerical codes so they can be statistically analysed using a computer.
Descriptive model	Descriptive refers to a theory's ability to describe behaviour (Howard, 1992).
Discrete data	See nominal data.
Health outcomes	The impact upon health following an intervention.
Interval data	Data on a scale where the distance between adjacent units is the same but there is no

	meaningful zero point e.g. Fahrenheit scale (Vogt, 1993).
Nominal data	Categorical or qualitative data.
Non-health outcomes	Outcomes associated with care. For example, how far a patient has to travel for health care.
Non-satiation test	A test of whether or not people prefer more benefits e.g. more product for the same price.
Normative model	An idealised description of individual decision making that should not necessarily be followed in practice (Keeney, 1992).
Ordinal data	Data measurable at the ordinal level. The values in an ordinal scale are a ranking.
Ordinal utility	Utility measurable to the ordinal level. Utility can be ranked. E.g. the utility of A is greater than the utility of B.
Penetrance	The proportion of individuals with a specific mutation that develop the associated disease.
Predictive validity	Predictive validity refers to an instrument's ability to predict changes in key variables.
Prescriptive model	The decision process to be recommended to a decision maker, even when normative rules are violated in this process.
Prevalence	The proportion of individuals in a population having a specific disease.
Process attributes	Aspect of a health service that relate to the process of care e.g. staff seen.
Ratio data	Data on a scale where the distance between adjacent units is the same and there is a meaningful zero point e.g. height. This data has ratio properties; 2 feet is exactly half of four feet (Vogt, 1993).
Reliability over attribute set	Test of the impact of the ordering of attributes upon reliability.
Reliability over scenario set	See reliability over stimulus set.
Reliability over stimulus set	Test of the impact of the ordering of scenarios upon reliability.
Reliability over time	A measure is considered to be reliable over time if it gives consistent results on more than one assessment of a construct that has not changed. Reliability over time is synonymous with test re-test.
Scenarios	In the context of discrete choice modelling, scenarios are made up of different combinations of attribute levels.
Theoretical validity	How well an instrument or measure conforms to theoretical expectations.
Transitive	Consistent ranking of commodities e.g. if A is preferred to B, and B is preferred to C, A should be preferred to C.

Utility	Relates to wellbeing, satisfaction or pleasure (Earl-Slater, 1999).
Utility function	A mathematical formula whereby a person's utility is represented by various factors that can or do affect their utility (Earl-Slater, 1999).

CHAPTER 1: PATIENT PREFERENCES IN THE DELIVERY OF CANCER GENETIC SERVICES

Genetic Cancer

For over a century clinicians have been aware that a hereditary predisposition to develop cancer exists in certain families (Steel, Smyth, Vasen, Eccles, Evans, Moller, Hodgson, Stoppa-Lyonnet, Chang-Claude, Caligo, Morrison & Haites 1999). Five percent of breast cancer cases are believed to be due to inherited genetic mutations (Lynch, Albano, Danes, Layton, Kimberling, Lynch, Cheng, Costello, Mulcahy, Wagner & Tindall, 1984). 10% - 11.7% of ovarian cancer cases (Landis, Murray, Bolden & Wingo, 1999; Malanders, Ridderheim, Masback, Loman, Kristoffersson, Olsson, Nibert & Boirg, 2004; Risch, McLaughlin, Cole, Rosen, Bradley, Kwan, Jack, Vesprini, Kuperstein, Abrahamson, Fan, Wong, & Narod, 2001) are believed to be the result of breast cancer susceptibility one and two (BRCA1/2) mutations. The hereditary genetic disorders of nonpolyposis colorectal cancer (HNPCC) and familial adenomatous polyposis coli (FAP or FAPC) are believed to be responsible for 2% - 7% and 1% of colorectal cancer cases respectively (Aaltonen, Salovaara, Kristo, Canzian, Hemminki, Peltomak, Chadwick, Kaariainen, Eskelinen, Jarvinen, Mecklin, de la Chapelle, Percesepe, Ahtola, Harkonen, Julkunen, Kangas, Ojala, Tulikoura & Valkamo, 1998; Soravia, Bapat & Cohen, 1997).

Women with a BRCA1 mutation have a lifetime risk in excess of 80% of developing breast cancer, 40% - 60% chance of developing ovarian cancer and possibly an increased risk of developing colorectal cancer (Ford, Easton, Bishop, Norad & Goldgar [Breast Cancer Link Consortium], 1994). Mutations in the hereditary genes nonpolyposis colorectal cancer (HNPCC) and familial adenomatous polyposis (FAP) predispose carriers particularly to colorectal cancer. A HNPCC germline mutation confers a lifetime risk of developing colorectal cancer of up to 80% - 90% (Vasen, Wijnen, Menko, Kleinbeuker, Taal, Griffioen, Nagengast, Meijers-Heijboer, Bertario, Varesco, Bisgaard, Mohr, Fodde & Klan, 1996; Vasen, Van Ballegooijen, Buskens, Kleibeuker, Tall, Griffioen, Nagengast, Menko & Khan, 1998; Soravia et al., 1997) and carriers of a FAP mutation have an 80% - 100% chance of developing colorectal cancer during their lifetime (Haggitt & Reid, 1986). HNPCC and FAP also convey

risks of developing cancers in the duodenum, ureteric tract and other organs (Vasen, Mecklin, Khan & Lynch, 1991; Soravia et al., 1997).

The epidemiological statistics cited above are the tip of the iceberg. The majority of individuals referred to cancer genetic services due to a family history placing them at increased risk of developing cancer do *not* have identified mutations such as BRCA1/2, FAP or HNPCC. Most high risk and all moderate risk families have as yet unidentified mutations. 18% (7% - 67%)[1] of high risk breast ovarian families have been found to have BRCA1/2 mutations (Couch, De Shano, Blackwood, Calzone, Stopfer, Campeau, Ganguly, Rebbeck & Weber, 1997; Stoppa-Lyonnet, Laurent-Puig, Essioux, Pages, Ithier, Ligot, Fourquet, Salmon, Clough, Pouillart, The ICBCG, Bonaiti-Pellie & Thomas, 1997; Eccles, Englefield, Soulby & Campbell, 1998) and 15.8% - 39.3% families with a high risk of colorectal cancer have been found to have a HNPCC mutation (Syngal, Fox, Li, Dovido, Eng, Kolonder & Garber, 1999).

Cancer Genetic Services

As a consequence of increased awareness amongst the general public, demand for genetic assessment services developed (Campbell, Mackay & Porteous, 1995; Evans, Fentiman, Mcpherson, Asbury, Ponder & Howell, 1994; Ponder, 1999; Priority Areas Cancer Team, 1998), but in a piecemeal fashion both in North America and Europe and frequently as part of academic research (Steel et al., 1999). In the late 1990s in the UK, regional cancer genetics clinics emerged in England and national services emerged in Wales and Scotland, thus enabling physicians to refer families with a history of cancer to specialist centres for risk assessment, and, if appropriate, genetic counselling and testing. It is unlikely that the expansion of cancer genetic services in the UK will desist in the foreseeable future and as a result this has substantial economic implications for the NHS.

Economic Evaluation of Cancer Genetics Services

Despite a burgeoning literature on the psychosocial impact of familial cancer and accessing genetic services upon patients (Audrain, Schwarts, Lerman, Hughes, Peshkin & Biesecker , 1998; Lerman, Hughes, Lemon, Main, Snyder, Durham, Norad

& Lynch, 1998; Brain, Gray, Norman, France, Anglim, Barton, Parsons, Clarke, Sweetland, Tischkowitz, Myring, Stansfield, Webster, Gower-Thomas, Daoud, Gateley, Moneypenny, Singhal, Branston, Sampson, Roberts, Newcombe, Cohen, Rogers, Mansel & Harper, 2000; Clarke, Bluman, Borstelmann, Regan, Winer, Rimer & Skinner, 2001; Fry, Cull, Appleton, Rush, Holloway, Gorman, Cetnarskyj, Thomas, Campbnell, Anderson, Steel, Porteous & Campbell, 2003; Geer, Ropka, Cohn, Jones & Miesfeldt, 2001; Rees, Fry & Cull, 2001), full and partial economic evaluations of cancer genetic services are sparse (see Chapter 2 and Griffith, Edwards & Gray, 2004). The systematic review of the economic evidence on cancer genetic services in Chapter 2 of this thesis reveals that economic evaluations to date have concentrated on technology assessment, looking at health outcomes and mutation identification. With the exception of the work of Wilson, Ryan and Haites (1999)[2] there is a lack of research looking into what *aspects* of cancer genetic services patients value (importance of service attributes) and how they would like services to be delivered.

A preoccupation with health outcomes fails to recognise the utility derived by patients from non-health outcomes and process attributes (Mooney, 1994; Mooney & Lange, 1993; Ryan, Farrar, Shackley, Vick & McIntosh, 1996; Ryan 1996a, 1999b; Singh, Cuttler, Shin, Silvers & Neuhauser, 1998). Since the late 1980s National Health Service policy documents have formally reflected this, advocated involvement between *service users*, clinicians, health care planners, policy makers and providers on key issues in health care such as service delivery and priority setting (DoH, 1992, 1998, 1999; Secretary of State for Health, Wales, Northern Ireland and Scotland, 1989; National Health Service in Scotland, 1992). Information upon the preferences of patients for service attributes and the costs associated with attributes is invaluable when planning and providing health services. For example should the majority of a finite budget for cancer genetic services be spent on securing as many specialist consultants as possible or would it be more effective in terms of maximising patient preference (utility) to focus the expenditure of a fixed budget upon one or more other service attributes.

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[1] The range in prevalence is a result of the variation in the criteria used to identify high risk families.
[2] O'Neill also conducted MSc research in this field but it could not be obtained from the University of Aberdeen by inter library loan.

Wilson et al. (1999) conducted an economic evaluation of the Scottish model of delivering cancer genetic services; a single counselling session with no NHS funded genetic testing for high risk patients. Patient preference (utility) was elicited using discrete choice modelling (DCM will be discussed later). The research examined patient preferences in relation to process and non-health outcomes of genetic counselling; namely 'staff seen at the appointment', 'Waiting time till appointment', 'Distance to appointment' and 'Duration of appointment'. Wilson et al. found evidence supporting the use of genetic associates and nurses rather than consultants to counsel when this resulted in improvements in other attributes.

There is no universally agreed protocol on how genetic services should be provided and as a result the findings of Wilson and et al. (1999) cannot be generalised to all NHS funded cancer genetic centres in the UK. Although there are several regional cancer genetics services in the UK the only other national service other than the one in Scotland is the one provided in Wales by the 'Cancer Genetics Service in Wales' (CGSW). The service offered in Wales is derived from the highly successful Huntington's protocol developed at the Institute of Medical Genetics in Cardiff (Griffith et al., 2005) and is substantially more resource intensive than the one offered in Scotland. The Welsh protocol offers risk assessment to all patients referred to them, genetic counselling to moderate risk patients, and in the case of high risk patients two counselling sessions and genetic testing for a cancer affected relative (to initially identify the genetic mutation) and four counselling sessions, genetic testing and the arrangement of presymptomatic surveillance for the presymptomatic patient(s)[3] initially referred to the service.

Research question 1

Clearly there is a need for a preference based economic evaluation of the Cancer Genetics service in Wales' resource intensive testing and counselling protocol for high risk patients. The main research questions raised from the economic literature on cancer genetic services for the Welsh service are "What are the attributes of cancer genetics services that are important to high risk patients (the patients spending the most time in contact with the service and receiving most services i.e. genetic testing

[3] Patient that is currently free of cancer but suspected of having a genetic mutation placing them at high risk of developing cancer.

and counselling)? and what would be the cost of providing the service to comply with patient preferences?"

Preference Elicitation

Clearly to answer research question one a reliable and valid method of collecting and interpreting the views and preferences of respondents is required. Work in this field has generally concentrated on stated preference techniques. A number of methods have been utilised within health economics to examine the multi-attribute nature of health care to date (Farrar, Ryan, Ross & Ludbrook, 2000; Ryan, 1996a), including opinion polls, satisfaction surveys visual analogue, contingent valuation, standard gamble and time-trade off (see Appendix A for a brief review of these techniques). However, all of these methods have been found to have limitations when looking at patient preferences for service provision (Ryan, McIntosh and Shackley, 1998a). Both opinion polls and satisfaction surveys provide limited information when the objective is to ascertain patient preferences; a substantial limitation of these techniques is that they do not provide any indication of strength of preference. There is also a lack of information on the relationship between attributes and they fail to incorporate opportunity cost. Techniques such as visual analogue, time-trade off and standard gamble which derive quality adjusted life years by trading years of life in various health states are not appropriate when aspects of service provision or prioritisation are the focus. Time-trade off and standard gamble have also been criticised on the grounds that the reliability of indifference answers are difficult to estimate with these techniques (Verhoef, Maas, Stalpers, Verbeek, Wobbes & van Daal, 1991). Although not solely associated with contingent valuation, the technique is particularly susceptible to six sources of bias: protest zero bids (Olsen & Donaldson, 1998), market inexperience (Drummond, 1995; Ryan, 1996b), policy or strategic bias (Johannesson, Jonsson & Karlsson, 1996), politicisation, warm glow effect and altruism (Cave et al., 1994). The six forms of bias are particularly associated with contingent valuation as respondents are asked to decide on a maximum financial value for a health intervention/service and there is reluctance on the part of some respondents to do this.

A technique utilised to obtain the views of patients, which can give valuable information on the utility of a service and inform provision and policy decisions should provide information on strength of preference, incorporate opportunity cost, provide information on attributes of the service (*process attributes* e.g. time with Dr, *non-health outcomes* e.g. distance to counselling and *health outcomes* e.g. improvement in health), and incorporate representative attributes and variation, allowing results to feed directly into policy and provision (Ryan, 1996a).

Discrete Choice Modelling (DCM)

A technique that fulfils the criteria outlined by Ryan (1996a) is discrete choice modelling. Discrete choice modelling is sometimes referred to as discrete choice conjoint analysis as it is a variant of conjoint analysis. The technique of conjoint analysis was developed within the discipline of mathematical psychology and it has been widely used in the fields of market research, transport economics, environmental economics and the economics of tourism (Ryan et al., 1996; Ryan, 1996a; Farrar, Ryan, Ross & Ludbrook, 1997; Ryan, 1999b; Ratcliffe, 2000a). Although, as early as the mid 1970s conjoint analysis had been used to address issues in health care in the USA (Ryan, 1999b), the adoption of the technique by health economists in earnest has only taken place since the early 1990s (Farrar et al., 1997; Bryan, Gold, Sheldon & Buxton, 2000, Ratcliffe, 2000a).

DCM assumes that the subject under consideration e.g. cancer genetic services, can be described in terms of its characteristics or attributes, that the utility of the service is a result of the individual utility of the attributes, that preferences are transitive[4], that preferences are continuous[5] and as such, choices are made in accordance with utility theory (Farrar et al., 2000).

As the name 'discrete choice modelling' suggests, respondents are asked to make a series of discrete choices (revealed, or in the majority of cases, stated preferences). Each choice question comprises of one (or more) scenario made up of attributes and their levels. An example of an attribute would include 'duration of consultation with a

[4] *Preferences are said to be transitive when A is preferred to B, B is preferred to C and subsequently A is preferred to C.*

clinician' and an example of a level would be '30 minute consultation'. Respondents can be asked to choose between two or more scenarios or simply if they would accept or reject a single scenario. Respondents' multiple responses are then analysed using random effect regression techniques (Ryan, 1999a). The results of the analysis reveal the utility associated with specific aspects of the service or commodity, the relative importance of attributes of a commodity or service, provides information on respondents' willingness to trade between attributes and most importantly the utility (preference) individuals have for a commodity or service with specific attributes and levels.

DCM has been successfully used in a variety of health care settings in the UK, including in vitro fertilisation (Ryan, 1999a; 1999b), orthodontic services (Health Economics Research Unit (HERU), undated a), elective surgery (HERU, undated b), general practice services and health cards (Ryan et al., 1998a) and hospital services (Jan, Mooney, Ryan, Bruggemann & Alexander, 2000). See Chapter 2 for a more detailed review of the design and implementation of discrete choice modelling.

DCM and Random Utility Theory

The underlying decision theory upon which discrete choice modelling (DCM) is based is random utility theory. Random utility theory is graphically depicted in Figure 1.1. Moving from left to right in Figure 1.1, accounting for demographic and psychological/personality traits e.g. gender and optimism, an individual weighs up the benefits and barriers of a choice (e.g. expected benefits and the likelihood of receiving the benefit) and chooses the option that maximises their subjective expected utility (provides maximum benefit). An error term (ε) is included to account for the unknown aspects of the respondent's utility (preference) function (Farrar et al., 2000).

Despite the obvious advantages of DCM, its growing popularity within the discipline of health economics and the growing literature addressing various aspects of its reliability and validity (Bryan et al., 2000; Farrar & Ryan, 1999; Ryan, McIntosh & Shaklay, 1998b), no one has experimentally examined its underlying decision theory principal of Random Utility Theory in conjunction with a DCM exercise.

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[5] *A preference is continuous when one attribute can compensate for a reduction in another.*

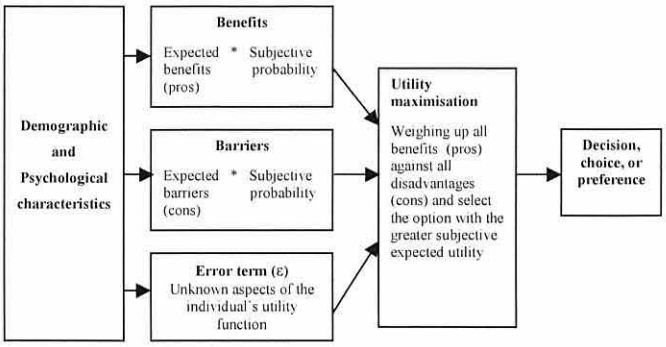

Figure 1.1. Representation of random utility theory.

Supplementing Economic Theory with Decision/Psychology Theory

In the absence of relevant data in the fields of health economics on the validity of random utility theory it was necessary to look at the decision theory/consumer choice and psychology literature. As the comparatively new academic discipline of health economics turns its attention to human behaviour in the form of patient preference and choice there is great potential to adapt and adopt theories, finding and methods that have been developed over many years in disciplines such as psychology to guide the aims and objectives of health economics.

The decision theory literature revealed that random utility theory was one of the latest utility theories, designed to redress the deficiencies identified with the preceding theories as descriptive models of choice. Expected utility and its siblings, subjective expected utility (SEU), and generalised utility in their various forms have been found to be invalid as descriptive models (Miyamoto, 1992; Keller, 1992; Camerer, 1992; Edwards, 1992) (a discussion of these theories is given in Chapter 2). Sarin (1992), Eppel, Matheson, Miyamoto, Wu and Eriksen (1992) and Schoemaker (1982) suggested that many of the violations of EU, SEU and generalised utility theories when experimentally tested were the result of psychological attributes that were not accounted for in the experiments and analysis.

The health psychologists Wroe, Salkovskis and Rimes (1998) believe that utility theory failed as a descriptive model under experimental conditions as it had been

applied in too narrow a fashion. In their contemporary revision of subjective expected utility theory they emphasise the influence of subjective beliefs (cognitive framework), proposing that the individuals' decisions are the result of a logical decision process but based upon their beliefs, transitive factors such as anxiety and the information that is available and deemed to be relevant at the time (no matter how irrational or factually incorrect it may be) (Wroe & Salkovskis, 1999).

In a series of studies Wroe et al.'s (1998) 'modified utility theory' has successfully predicted interest in obtaining genetic testing and attendance at screening (Salkovskis, Dennis & Wroe, 1999; Wroe & Salkovskis, 1999, 2000; Wroe, Salkovskis & Rimes, 1998, 2000). Wroe et al. (2000) found that 96.4% of participants were correctly classified as to their subsequent decision whether or not to take a test for bone density screening based upon their intention to have testing; both behavioural intention and actual uptake complied with modified utility theory. However, research into the impact of information upon the hypothetical decision to have genetic testing by Cameron and Diefenbach (2001) contradicts Wroe and Salkovskis' (1999) findings and raises further doubts as to the descriptive ability of utility theory in its various forms. See Chapter 2 for a review of utility theory.

Research Question 2

The literature on random utility theory (RUT) clearly shows that there is uncertainty surrounding the validity of RUT as a descriptive model of choice. Given that RUT is the underlying decision theory of DCM the validity of the results of DCM studies are called into question. Therefore, an empirical investigation of the assumption that respondent of DCM exercises make choices in accordance with random utility theory is necessary to validate the research into patient preferences for the delivery of cancer genetic services using a DCM exercise (Research question 1 above). Research question 2 is, "Do respondents of DCM questionnaires make choices in accordance with Random Utility Theory?"

Random Utility Theory and Determinants of Choice

Utility theory does not provide a detailed description of the way individuals come to make their decisions (Feather, 1982; Edwards, 1992; Jones, 1993, Frisch & Clemen, 1994; Conner & Norman, 1995), confining itself to benefits and barriers (the pros and cons) of a decision. To address this deficiency and highlight some of the determinants of choice the social/health psychology literature was accessed yet again. To extend the identification of the determinants of choice beyond the weighted benefits and barriers used in RUT, social cognition models that assume that decisions are made in accordance with utility theory (Edwards, 1954a; Conner & Norman, 1995) were examined. The models assessed are those identified by Weinstein (1993), van der Plight (1994) and Conner and Norman (1995), as being rooted in utility theory[6]; the health belief model (HBM), health locus of control (HLC), protection motivation theory (PMT), theory of reasoned action (TRA) / theory of planned behaviour (TPB) and self-efficacy theory (SET).

Two social cognition models, the Theory of Planned Behaviour (TPB) (Ajzen, 1985, 1988, 1991) and the Health Belief Model (HBM) (Hochbaum, 1958; Rosenstock, 1966, 1974; Becker et al., 1977) (see Figure 1.2) were selected as the most appropriate to identify the determinants of choice. The review of the social cognition models and the rationale for selecting the TPB and HBM is presented in Chapter 2.

Both the HBM and TPB (see Figures 1.2) assume that decisions are made in accordance with utility theory (Edwards, 1954a; Conner & Norman, 1995), weighing up the benefits and disadvantages of alternative courses of action and selecting the one with the greater subjective expected utility. Working from left to right in Figure 1.2, having accounted for variation in demographic and psychological/personality traits between individuals, both models, unlike utility theory, decompose the all encompassing benefits and barriers into the respective factors identified in each model (e.g. Perceived susceptibility, perceived severity, etc. for the HBM and Attitudes towards behaviour, subjective norm, etc. for TPB) and combine the benefits/positive[7]

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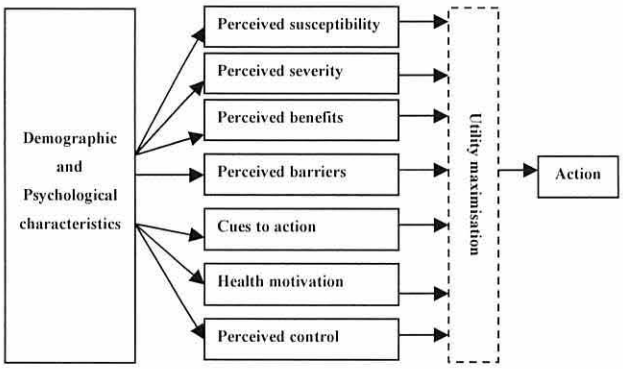
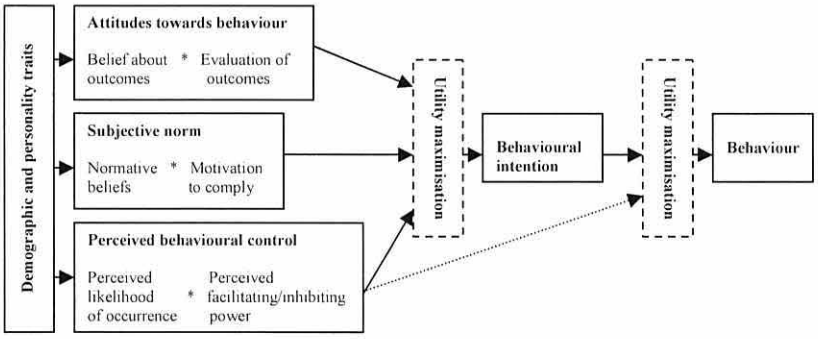
[6] Sometimes when social cognition models are discussed in the literature the term expectancy value is used interchangeably with utility theory. In that context they can be treated as equivalent as they are referring to the subjective utility and probability of an outcome.

[7] The Health belief model does include the factors perceived benefits and perceived barriers. However, operationalisation of these factors differs to its operationalisation in utility theory. Barriers tend to

elements with the disadvantages/barriers/negatives into a choice using subjective expected utility to determine which course of action/choice will maximise their utility (Edwards, 1954a). Both the TPB and HBM are discussed in greater detail in Chapter 2.



Health Belief Model with the utility maximisation (RUT) component made explicit



Theory of Planned Behaviour model with the utility maximisation (RUT) component made explicit

Note: The HBM depiction is based upon Ogden (2000) and the TPB depiction is based upon Conner & Sparks (1995); utility maximisation cells have been added. For the experiment reported in chapters 3 to 5 the TPB is only applied to behavioural intention as the decision to be made is hypothetical. As is common practice, only elements of the HBM and TPB deemed study relevant are applied in the empirical analysis chapters of this thesis (Chapters 3 and 4).

Figure 1.2. Interrelationship between the HBM and RUT, and the TPB and RUT.

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focus on barriers to accessing health services or participating in health behaviour and not all negative issues associated with a certain health behaviour. Benefits are confined to those identified in the questionnaire and are mutually exclusive with the benefits associated with the other behaviour and motivation factors e.g. health motivation benefits etc.

Research question 2.1

Following on from research question two an additional question is raised, "In the event that respondents are found to adhere to RUT, can the determinants of choice be extended beyond 'benefits and barriers' (RUT) using relevant components of the theory of planned behaviour and health belief model?"

Thesis Aims

Test the theoretical validity of discrete choice modelling and utilise the technique to explore the most appropriate way in terms of costs and consequences to configure cancer genetic services in respect of high risk patients' preferences.

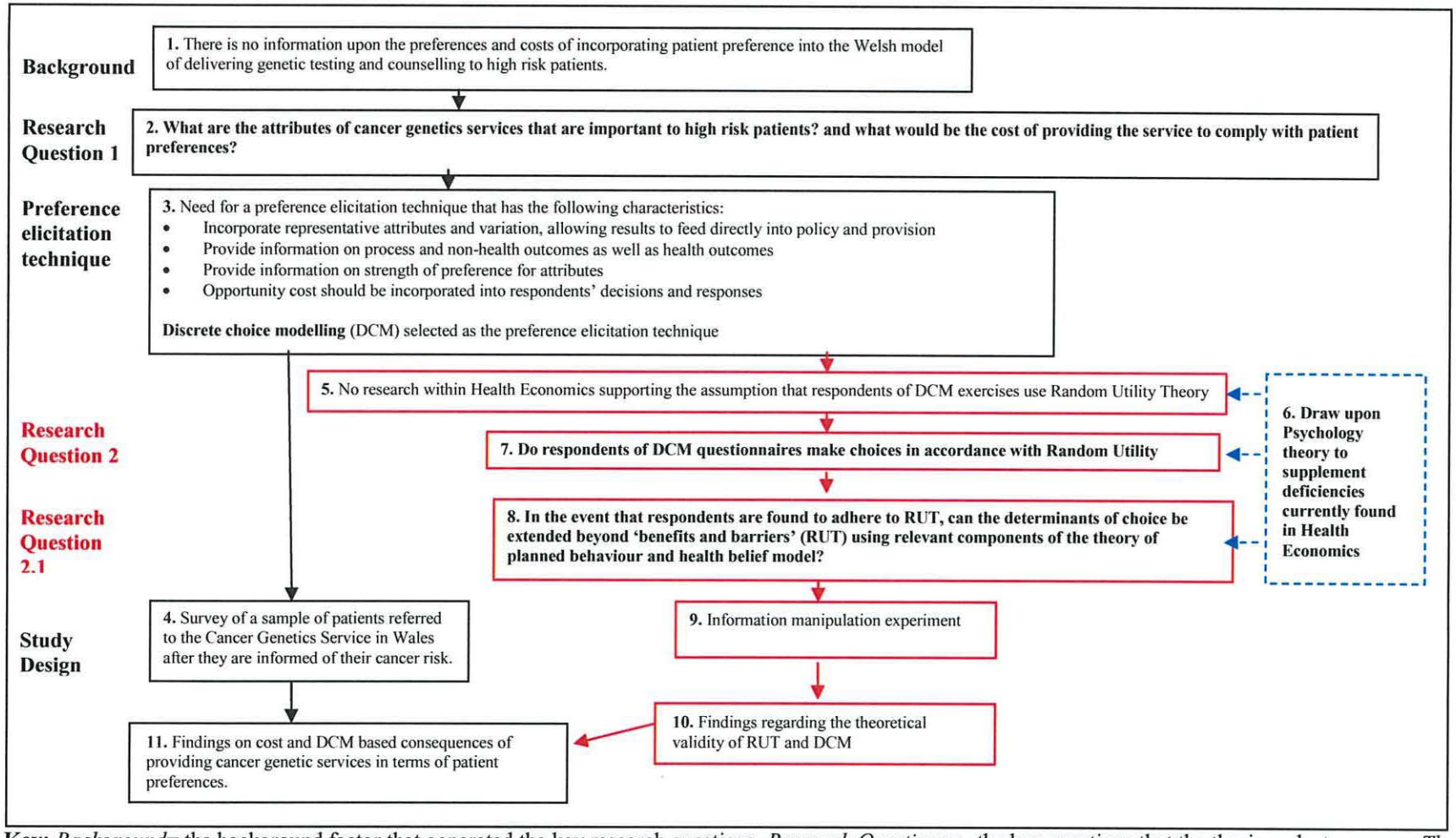
Empirical Aims

- Experimentally examine respondents of a DCM exercise by means of an information manipulation to see if they are adhering to DCM's underlying decision theory principals of random utility theory.
- Ascertain the aspects of cancer genetics services that are important to patients, and present service configurations prioritised in terms of preferences accompanied by their costs (cost-consequences analysis) for high risk patients.

Conceptual Framework of the Thesis

The conceptual framework is presented graphically in Figure 1.3. The paragraph numbers below relate to the numbers in the cells of the flow chart in Figure 1.3.

1. There are two national cancer genetics services in the UK, one in Wales and one in Scotland. Whilst the Scottish service which offers one counselling session and no genetic counselling has undergone economic evaluation accounting for patient preferences in the delivery of the service, no such evaluation has been conducted for the markedly different Welsh service. The Cancer Genetics Service in Wales (CGSW) offers a substantially more resource intensive model of care, offering high risk patients multiple counselling sessions and genetic testing. Given that there is no information upon the preferences of patients for the manner in which the Welsh model (and similar regional services in England) of providing cancer genetic services is delivered, or data on the associated costs, research question one was set.



Key: *Background*= the background factor that generated the key research questions. *Research Questions*= the key questions that the thesis seeks to answer. *Theories*= the behaviour models utilised in the thesis. *Study Design* = Research design used to elicit data for analysis to seek to answer the research questions using the theories. DCM = discrete choice modelling. RUT= random utility theory.

Figure 1.3. Conceptual framework.

2. Research question 1 "What are the attributes of cancer genetics services that are important to high risk patients (the patients spending the most time in contact with the service and receiving most services i.e. genetic testing and counselling)? and what would be the cost of providing the service to comply with patient preferences?"

3. Having examined the literature on eliciting patient preferences the relatively new and experimental technique of discrete choice modelling (DCM) was identified as the most appropriate technique to use to elicit the data required to answer research question 1.

4. The study design used to answer research question 1 was a sample of patients referred to the Cancer Genetics Service in Wales. Patients were issued a self-administered DCM questionnaire one week after they were informed of their genetic risk status. Tests of understanding and clarity were run on a sample of 115 patients (low, moderate and high risk) and cost-consequences analysis on a sample of 30 high risk patients. Full details of research methods are reported in chapter 3.

5. In order to answer research question one it is essential that a valid preference elicitation technique is used. The health economics literature revealed that no one has experimentally examined DCM's underlying decision theory principal of random utility theory in conjunction with a DCM exercise.

6. To supplement the deficiency in the health economics literature, the decision theory/psychology literature was accessed. The large body of literature on utility theory revealed that the descriptive ability of utility theory was in question. These findings resulted in research question 2.

7. Research question 2, "Do respondents of DCM questionnaires make choices in accordance with Random Utility Theory?"

The decision theory/psychology literature (cell 6 of flowchart) also revealed that the failure of utility theory as a descriptive model (explanatory model) under experimental conditions was most likely due to deficiencies in the experimental

designs applied, i.e. defining utility maximisation in too narrow a fashion and failing to account for significant mediating psychological variables.

8. Within health economics theory, specifically utility theory, the determinants of choice have been confined to the benefits and barriers of a decision. By incorporating social cognition models from social/health psychology it is possible to go beyond a list of benefits and barriers to more detailed motivating factors such as perceived susceptibility to a disease and perceptions of the severity of a disease. A review of the literature and an evaluation of social cognition models that assume that decisions are made in accordance with utility theory identified the theory of planned behaviour and the health belief model as the most appropriate for identifying relevant determinants of choice. This resulted in research question 2.1 "In the event that respondents are found to adhere to RUT, can the determinants of choice be extended beyond 'benefits and barriers' (RUT) using relevant components of the theory of planned behaviour and health belief model?"

9. The study design used to address research question 2 and 2.1 was conducting an information manipulation experiment with 142 undergraduate students. Full details of the research methods are reported in chapter 3.

10. The findings of the information manipulation experiment reveal if respondents of DCM questionnaires make choices in accordance with random utility theory. If respondents make decisions in accordance with RUT then DCM is based upon a valid descriptive decision theory and the results of DCM exercises are theoretically valid (it should be noted that other validity aspects not considered in this research may be violated). Alternatively, if respondents of DCM exercises are not adhering to RUT decision making, then the results of DCM exercises are in question. The findings relating to the theoretical validity of DCM feed directly into the results of the survey of patients to answer research question 1 (cell 11 of flowchart), revealing if the preference results of the survey are theoretically valid and appropriate to present to policy makers.

Theoretical Framework

Clearly the theoretical framework underpinning the thesis is random utility theory. Discrete choice modelling is based upon the assumption that respondents are making choices in accordance with random utility theory. When incorporating the theory of planned behaviour and the health belief model to identify the determinants of choice, it should be noted that these models are based upon utility theory and are extensions of utility theory, where the all encompassing benefits and barriers used in utility theory are decomposed into the respective beliefs identified in each model e.g. subjective norm etc.

Contribution to Health Economics Theory

This thesis has been designed and conducted with the intention of making two main contributions to health economics theory and literature. Firstly, it is the first economic evaluation of a national cancer genetics service (offering genetic testing) in terms of high risk patients' preferences for the manner in which the service can be delivered and the associated costs to the NHS (cost-consequences analysis). Secondly, the thesis experimentally tests the explicit assumption made, when using discrete choice modelling as a health economics research technique to elicit preference data, that respondents use random utility theory to make their decisions. To achieve these goals it was necessary to import and integrate contemporary work from the fields of psychology and health psychology into health economics. Contemporary work in psychology/health psychology has been incorporated into this thesis, which is rooted in health economics, in two ways. Firstly, by adopting Wroe et al.'s (1998) cognitive approach to measuring the benefits and barriers (pros and cons) of a choice; directly asking respondents what aspects of a choice are important to them and how important they are, rather than the preceding approach of applying the researcher's view of what is important and which choice represents utility maximisation. Secondly, by incorporating the theory of planned behaviour and the health belief model to extend the determinants of choice beyond random utility theory's benefits and barriers. Given the reasons cited above, this thesis is a significant contribution to the health economics theory.

CHAPTER 2: PATIENT PREFERENCES, DISCRETE CHOICE MODELLING AND UTILITY THEORY

In the preceding Chapter the background and the aims of this thesis have been outlined. The purpose of this Chapter is to give a fuller description, evaluation and discussion of the literature based arguments presented in Chapter 1. The review predominantly focuses upon three main issues. Firstly, the review highlights the paucity of evidence on patient preferences in economic evaluations of the provision of cancer genetic services for breast, ovarian and colorectal cancer. Secondly, it identifying the most appropriate research technique for collect data on patient preferences for cancer genetic services and outlines the technique. Thirdly, it examines random utility theory, the theory underpinning the data elicitation technique utilised in this thesis (discrete choice modelling) to ascertain patient preferences.

To establish the amount of research that had incorporated or focussed upon patient preferences as an outcome in economic evaluations of the provision of cancer genetic services for breast, ovarian and colorectal cancer a comprehensive literature search was required. To accomplish this a systematic review was conducted of the health economics literature. The results of the systematic review are briefly reported in this Chapter and published in full in Griffith et al. (2004). As the review of preference elicitation techniques, discrete choice modelling, utility theories and social cognition models aimed to highlight the key themes in these fields and not identify all literature on these topics, a systematic search strategy of electronic databases was not adopted for these topics.

Methods

Search strategy

A structured search was conducted to identify evidence relating to the economic evaluation of cancer genetic services for individuals/families at risk of having familial breast, ovarian or colorectal cancer. This was done by means of searching the electronic databases: BMJ Archive, BIDS, Medline, HealthPromis, DARE, EED, HTA, Cambridge Scientific Abstracts, Econobase, CINAHL, ASSIA, British Library Catalogue, OCULC WorldCat, Resource Discovery Network and the Cochran Library. Titles, abstracts and articles were searched for the keywords cancer, genetic and economic or cost. Retrieved papers were hand searched for references that had not been identified in the electronic search.

The methodological and theoretical literature (preference elicitation techniques, discrete choice modelling, utility theories and social cognition models) reviewed in this Chapter were identified by identifying relevant literature based on current knowledge and scanning the references for relevant literature "snowballing" (Greenhalgh and Peacock, 2005)[1].

Inclusion and Exclusion Criteria

The criteria for including economic literature in the review was that it was a review article, partial or full economic evaluation of treatment, detection or counselling of individuals for familial cancer. Research was excluded from the review if it: was published in any language other than English; examined sporadic cancer; examined a form of genetic cancer other than breast, ovarian or colorectal cancer such as hemochromatosis; was not an economics paper and only suggested that there may be potential economic savings, and if the term cost had been used to refer to anything other than financial cost e.g. adverse psychological events.

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[1] Many of the key papers and reviews relating to utility theory and some relating to social cognition models predate the start dates for electronic abstract databases.

The criteria for including methodological and theoretical literature in the review was that it specifically discussed preference elicitation techniques used in economic evaluations, how to conduct discrete choice modelling, specifically described and/or evaluated utility theories or social cognition models. Applications of elicitation techniques and theories were only included if they were needed as illustrative examples. As was the case for economic evaluations, research published in any language other than English was excluded from the review.

Data Extraction and Analysis

All abstracts were obtained and assessed for relevance. All articles found to be relevant or potentially relevant were obtained and reviewed in full. To asses the quality and deficiencies of economic evaluations, the elements identified in Drummond, O'Brien, Stoddart and Torrance's (1997) 10 item checklist for a sound economic evaluation (see Table 2.1) were sought. These elements were then appraised as were their interrelationships. Methodological and theoretical literature was appraised on the grounds of the strength of the arguments presented.

Table 2.1

Drummond et al.'s 10 Item Checklist for a Sound Economic Evaluation

1	Was a well-defined question posed in an answerable form?
2	Was a comprehensive description of the competing alternatives given?
3	Was the effectiveness of the programmes or services established?
4	Were all the important and relevant costs and consequences for each alternative identified?
5	Were costs and consequences measured accurately in appropriate physical units?
6	Were costs and consequences valued credibly?
7	Were costs and consequences adjusted for differential timing?
8	Was an incremental analysis of costs and consequences of alternatives performed?
9	Was allowance made for uncertainty in the establishments of costs and consequences?
10	Did the presentation and discussion of study results include all issues of concern to users?

(Drummond et al., 1997)

Results

The Economic Evidence and Issues Relating to Cancer Genetics Services

The search for economic evaluations of cancer genetic services identified 1030 papers, of which 31 fulfilled the inclusion criteria. Of the 31 papers covering 29 studies, 2 were cost-benefit studies, 5 were cost consequences, 4 were cost-effectiveness studies, 1 was a cost analysis, 2 were cost-minimization studies, 1 was a cost-utility study, 10 modelled life years (5 also considered costs) and 6 were reviews. The 31 papers are listed in Table 2.2 according to the form of economic evaluation used in the studies and the main outcome measures used. At times the classification of studies differs from those of the original authors e.g. cost-effectiveness using more than one outcome variable has been classified as cost-consequences analysis (Cohen, Barton, Gray, & Brain, 2004).

Of the 23 studies reporting primary data (see Table 2.2), 21 reported health outcomes as the only effectiveness or benefit(s) of cancer genetic testing. 12 studies reported life years saved as the outcome or one of the main outcomes of cancer genetic services, 6 reported QALYs as the main outcome (accompanies by life years saved for 5 studies) and 8 studies reported mutation detection as a key outcome. The two studies that did not report health outcomes were those of Chaliki, Loader, Levenkron, Logan-Young, Hall, and Rowley (1995) and Wilson et al. (1999).

Table 2.2

Form of evaluation

Name	Year	CB	CC	CE	CA	CM	CU	MLY	R	Outcome measure(s)	D10
Maher et al.	1993	P								Years of surveillance avoided (Life years)	4/10
Chaliki et al.	1995	P (WTP)								WTP $25 for genetic testing	1/3
Brown and Kessler	1995, 1996		P							Cases of HNPCC mutations detected & life year saved	4/10
Heimdal et al.	1999		F							Life year saved & cancers detected	7/9
(TRACE) Brain et al. Cohen et al.	2000a 2004		F							Reasons for attendance at clinic, state & trait anxiety, breast cancer worry, perceived risk of breast cancer, knowledge of breast cancer & patient satisfaction	9/9
Eccles et al.	1998			F						Mutation detection	5/9
Lidereau et al.	2000			F						Mutation detection	5/9
Debniak et al.	2000			F						Mutation detection	5/9
Sevilla et al.	2002			F						Mutation detection	8/9
Van Orsouw et al.	1999				P					Mutation detection & cost per test	3/9
Cromwell et al.	1998					P				Mutation detection	7/10
Bapat et al.	1999					P				Mutation detection	9/10
Wilson et al.	1999						F			Utility/preference for the way genetic counselling was provided	8/9
Vasen et al.	1998		✓					F		Life years saved	8/10
Syngal et al.	1998							P		Life years saved & QALYs gained	7/9
Schrag et al.	1997							P		Life years saved	6/9
Schrag et al.	2000							P		Life years saved	7/9
Grann et al.	1998		✓					F		Life years saved & QALYs gained	6/10
Grann et al.	1999			✓				F		Life years saved	7/10
Grann et al.	2000		✓					F		Life years saved & QALYs gained	7/10
Grann et al.	2002							P		Life years saved & QALYs gained	7/9
Tengs et al.	1998							P		Life years saved & QALYs gained	7/9

Tengs & Berry	2000			✓				F		QALYs gained	8/10
Lerman	1997								✓	NA	NA
Peters & Biesecker	1997								✓	NA	NA
Priority Areas Cancer Team	1998								✓	NA	NA
Hall et al.	1998								✓	NA	NA
Steel et al.	1999								✓	NA	NA
Edwards	2001								✓	NA	NA

CB = Cost-benefit analysis, CC = Cost-consequences analysis, CE= Cost-effectiveness analysis, CA = Cost analysis, CM = Cost-minimisation, CU = Cost-utility analysis, MLE = Modelling life expectancy, R = Reviews, D10 = Proportion of Drummond et al.'s (1997) 10 item checklist that were fulfilled. P indicates a partial economic evaluation, F a full economic evaluation and NA non applicable.

Wilson et al. (1999) was the only economic evaluation to account for patient preferences as an outcome of cancer genetic services[2]; using discrete choice modelling to elicit patient preferences/utility for the delivery of a single genetic counselling session. The four service attributes tested were significant; staff seen at the appointment, waiting time till appointment, distance to appointment and duration of appointment. At each of the three centres where the study was conducted, maximum utility per pound was obtained by a scenario using nurse led counselling. In terms of patient utility, genetics nurses and associates were found to be cost effective compared to doctor led counselling[3].

It should be noted that no detail was given by Wilson et al. (1999) on the assumptions made when producing cost estimates for the comparison of service provision based upon patient preferences for significant service attribute levels. Based upon the results presented it would appear that little to no provision was made in the costs to allow for the attributes of distance to counselling and waiting time. For example, for the Aberdeen clinic a £0.07 additional cost was estimated for a reduction of 4 miles in travel and 4 month waiting time suggests that travel costs, capital, labour and overheads costs have been underestimated.

Eliciting Preferences

The predominant focus on health outcomes as the primary if not sole benefit to be accounted for in economic evaluation is common (Ratcliffe, 2000a; Ryan, 1993; 1999a) and not confined solely to the evaluation of cancer genetic services. A preoccupation with health outcomes fails to recognise the utility derived by patients from non-health outcomes and process attributes (Mooney, 1994; Mooney & Lange, 1993; Ryan et al., 1996; Ryan 1996a, 1999b; Singh et al., 1998).

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[2] O'Neill conducted his/her Msc research in this field but it could not be obtained from the University of Aberdeen by inter library loan.

[3] The summary of all the economic evaluations can be accessed at http://www.bangor.ac.uk/healtheconomics/Text/cancer/litappendix

The need to elicit the preferences of service users, carers, service providers, clinicians and any other concerned party requires a reliable and valid method of collecting and interpreting the views and preferences of respondents. Work in this field has generally concentrated on stated preference techniques due to the limitations of revealed preference (Pearmain, Swanson, Kroes & Bradley, 1991). The limitations of revealed preference include: existing behaviour may not vary sufficiently to conduct statistical analysis and modelling upon the data, multicollinearity may exist, behaviour may reflect factors that are not relevant to the aims of the research, factors of interest may be swamped by irrelevant factors, new or proposed services cannot be studied, large and expensive studies may be required to obtain a sufficiently large sample of observations and although routinely collected data may be collated it may not necessarily be available to the researcher (Pearmain et al., 1991; Cave, Burningham, Buxton, Hanney, Pollitt, Scanlan & Shurmer, 1994).

Numerous methods have been utilised within health economics to examine the multi-attribute nature of health care to date (Farrar et al., 2000; Ryan, 1996a), including opinion polls, satisfaction surveys, contingent valuation, visual analogue, standard gamble and time-trade off (see Appendix A for a brief review of these techniques). However, all of these methods have been found to be unsuitable (Ryan et al., 1998a). Opinion polls and satisfaction surveys provide limited information when the objective is to ascertain patient preferences; a substantial limitation of these techniques is that they do not provide any indication of the strength of preference. There is also a lack of information on the relationship between attributes, i.e. is who you see for counselling more important than the length of the counselling session? These methods also fail to incorporate opportunity cost (if one clinical service is funded the opportunity cost is the forgone benefits of the health service(s) which were not funded). In spite of the popularity of stated preference techniques which derive quality adjusted life years (visual analogue, time-trade off and standard gamble), these techniques are not appropriate when aspects of service provision or prioritisation are the focus. It is not realistic for example to ask respondents to trade years of life against improvements in service provision. Time-trade off and standard

gamble have also been criticised on the grounds that the reliability of indifference answers are difficult to estimate (Verhoef et al., 1991). Although not exclusive to contingent valuation (willingness to pay and willingness to accept), the technique is particularly susceptible to six sources of bias: protest zero bids (Olsen & Donaldson, 1998), market inexperience (Drummond, 1995; Ryan, 1996b), policy or strategic bias – belief that the response will affect actual provision or cost of service (Johannesson et al., 1996), politicisation – political beliefs influencing response, warm glow effect – satisfaction from stating a high or low bid value, and altruism – responses influenced by perceptions of others benefits (Cave et al., 1994). For a more detailed review of the limitations of these techniques see Cave et al. (1994), Drummond (1995), Olsen and Donaldson (1998), Ryan (1996b) and Verhoef et al. (1991). The six forms of bias are particularly associated with contingent valuation as respondents are asked to decide on a maximum financial value for a health intervention/service and there is reluctance on the part of some respondents to do this.

A technique utilised to obtain the views of patients, which can give valuable information on the utility of a service and inform provision and policy decisions should provide information on strength of preference, incorporate opportunity cost, provide information on attributes of the service (*process attributes* e.g. time with Dr, *non-health outcomes* e.g. distance to counselling and *health outcomes* e.g. improvement to health), and incorporate representative attributes and variation, allowing results to feed directly into policy and provision (Ryan, 1996a). A technique that fulfils these requirements is discrete choice modelling.

Discrete Choice Modelling

Discrete choice modelling (DCM) is a variant of conjoint analysis which was developed within the discipline of mathematical psychology (Ben-Akiva & Lerman, 1985; Ryan et al., 1996). It has been widely used in a range of fields including transport economics, environmental economics, tourism and marketing. The underlying decision theory principal of DCM is Random Utility Theory (Ben-Akiva

& Lerman, 1985; Louviere, Hensher & Swait, 2000; Ryan, 1996a; Ryan et al., 1996). Random Utility Theory (RUT) (Lancaster, 1966; Luce, 1959; Marshak, 1960; McFadden, 1972; Thurstone, 1927) which will be discussed in greater detail later in this Chapter. In accordance with random utility theory DCM assumes the axioms listed in Table 2.3.

Table 2.3

Random utility theory axioms

•	The subject under consideration can be described in terms of its characteristics or attributes (Singh et al., 1998; Farrar et al., 2000).
•	The utility of services or goods are a function of the individual utility of the attributes (Singh et al., 1998; Farrar et al., 2000).
•	Each attribute level has a distinct value or utility to respondents (Singh et al., 1998).
•	Preferences are transitive e.g. If A is preferred to B and B is preferred to C then A should be preferred to C (Verhoef et al., 1991).
•	Preferences are continuous. Improvement in one attribute can compensate for a reduction in another (Ryan, 1999b).
•	Individuals are utility maximisers (Walker and Ben-Akiva, 2002). They make choices that provide them with the maximum utility.

$$V = \sum \alpha_i HO_i + \sum \alpha_j NHO_j + \sum \alpha_k P_k + e$$

V	***Estimate of measurable component of utility.***
$\alpha_{i...k}$	Coefficients for the attributes.
HO	Health outcome attribute.
NHO	Non health outcome attribute.
P	Process attribute.
e	Error term (unobservable factors in the respondents' utility function).

Source: Ryan (1999a).

Figure 2.1. A DCM regression equation

Assuming a linear utility function, the utility of a commodity or service can be expressed as a model or regression equation (Farrar et al., 1997; McFadden, 1972, 1974; Ryan, 1999b). The equation, see example in Figure 2.1, can include any characteristic (attribute) e.g. health (HO), non-health (NHO) or service/process attribute (P), including those of a qualitative nature such as the clinician's attitude. Whilst the respondent is aware of his/her utility function, the researcher is not; random utility theory includes an error term (e) in the utility function to account for the unknown aspects of the respondent's utility function (Farrar et al., 2000). An individual chooses one option over another (A over B) if the utility (V) of option A is greater than option B ($V_A > V_B$).

To account for non-random variation in coefficients (Farrar et al., 1997), such as the need to differentiate between personal characteristics of respondents e.g. psychological, sociological or economic factors, Farrar et al. (1997) recommend segmenting the model according to the characteristics of interest or if the relationship is linear, to include interaction terms in the model.

The Primary Stages in a Discrete Choice Modelling Study

There are five main stages in conducting a typical discrete choice modelling study (Cave et al., 1994; Ryan, 1999b; Ryan & Farrar, 2000); establishing the attributes,

assigning levels to the attributes, selecting scenarios to present, establishing preferences and analysing the data.

Attribute Identification

The attributes to be included in a DCM model are usually identified by one or more of the following methods:

- Literature review.
- Discussion group with a sample of the target population.
- Interviews with a sample of the target population.
- Policy issues that need addressing.

Attribute Levels

The levels must be realistic and trade-offs between them possible. Clearly, defining attribute levels is easier for empirical attributes such as waiting time or cost than qualitative attributes such as attitudes (Ryan, 1996a). Table 2.4 presents examples of service attributes and levels.

Table 2.4:

Attributes and levels used in a study of miscarriage management.

Attributes	**Levels**
The level of pain you will experience	Low, Moderate, Severe
Time in hospital receiving treatment	1 day and 0 nights, 2 days and 1 night, 3 days and 2 nights, 4 days and 3 nights,
Time taken to return to normal activities after treatment	1-2 days, 3-4 days, 5-6 days, more than 7 days
Cost to you of treatment	£100, £200, £350, £500, £600
Complications following treatment	No, Yes

(Source: Ryan and Hughes, 1997)

Selection of Scenarios

Once the attributes and levels have been established they can be combined to form scenarios as in Figure 2.2. The number of possible scenarios is a product of the number of attributes and their levels. The maximum number of scenarios being the sum of the number of levels to the power of the number of attributes, e.g. a study with 2 attributes with 3 levels and 3 attributes with 3 levels would have 243 (3^2 x 3^3)

possible scenarios. Cave et al. (1994) suggests that no more than six or seven attributes can be presented to a respondent and Smith (1995) suggests no more than four levels.

In the event of a small number of possible scenarios, all can be presented to respondents (full/complete factorial design). Presenting respondents with a large number of scenarios is to be avoided. Verhoef et al. (1991) in their discrete choice study, presented breast cancer patients with forty eight paired comparisons and noted problems with fatigue, lack of care and attention to detail and inconsistency, particularly in the latter stages of questionnaire completion.

In the event of having a large number of scenarios five methods are advocated individually or in conjunction for dealing with this problem (Pearmain et al., 1991; Ryan et. al., 1996).

- Remove options that will dominate or be dominated by the remainder of the scenarios.
- Define attributes in terms of differences between alternatives e.g. Journey time by new train system = 15 minutes less than by car.
- Separate the options into blocks, so the full choice set is completed by groups of respondents, each responding to a different subset of options. When the subsets are analysed together they reconstruct the original design.
- Carry out a series of experiments with each individual, using different attributes. At least one attribute should be common to each DCM instrument to enable comparison.
- A fractional factorial design can be employed. If some or all interactions are considered to be insignificant, then the number of scenarios to be presented can be reduced. Allowing each attribute level to be varied independently for each of the other attribute levels. However, if there are significant unforeseen interactions between attributes, this will result in the effect being loaded onto the individual main effects, giving confounded results.

Ascertaining Respondents' Preferences

Having determined the attributes, levels and scenarios to be presented, preferences need to be elicited. A unique aspect of DCM and conjoint analysis is that respondents are asked to choose amongst realistic options or scenarios rather than provide direct utilities or benefit values for each attribute level (Singh et al., 1998).

Individuals are asked to state their preference in the form of accepting or rejecting a scenario or the more common format is to ask them to choose from a choice of two or more scenarios (as in Figure 2.2). Ryan (1999a) noted that future applications of DCM in health economics should consider including an indifference or not choosing option.

• **Scenario 2**	**Surgical Treatment**	**Medical Treatment**
The level of pain you will experience	Low	Moderate
Time in hospital receiving treatment	1 day, 0 nights	1 day, 0 nights
Time taken to return to normal activities after treatment	3-4 days	3-4 days
Cost to you of treatment	£350	£200
Complications following treatment	Yes	No
	Prefer Surgical ☐	Prefer Medical ☐

(Source: Ryan & Hughes, 1997).

Figure 2.2. Scenarios used in a study of miscarriage management in Scotland.

Discrete choice is favoured in health economics studies for a variety of reasons. DCM is based upon a theory of human behaviour (RUT); "Generally speaking there can be no valid measurement without an underlying theory of the behaviour of the numbers which result from measurement" (Louviere et al., 2000, p25). DCM designs are believed to be representative of the type of decisions service users engage in daily, whilst ranking or rating exercises are rare (Ryan, 1999a). Discrete choice modelling also incorporates the concept of opportunity cost, forcing respondents to choose between types of service delivery, forgoing one option in favour of another.

In designing a discrete choice instrument it is necessary not only to consider the absolute number of scenarios which are presented but also their placement. Scenarios are paired in one of four ways; comparing all scenarios to a single scenario selected at random (Ryan, 1999a), comparing all scenarios to a scenario representing current service provision, pairing scenarios randomly or pairing scenarios directly with the intention of ensuring clear tradeoffs between attributes and levels (Farrar et al., 1997).

Analysis

As DCM questions yield data that is nominal (or discrete) the most common regression techniques used in the analysis of such data are logit or probit methods (Ryan et al., 1996; Ryan, 1996a). Use of a simple probit model assumes random independent error for observations. However, in DCM, multiple observations are recorded for each respondent, which may result in observations being correlated and as a result the assumption of random error is violated (Farrar et al., 1997; Vick & Scott, 1998).

Non random error would result in underestimation of the standard errors of the model and result in the over estimation of the statistical significance of coefficients (Ryan et al., 1996; Farrar et al., 1997; Vick & Scott, 1998). To overcome the difficulties associated with conducting regression analysis on data with multiple observations from respondents, random effect models are utilised (Farrar et al., 1997; Vick & Scott, 1998; Ryan; 1999a, 1999b; Louviere et al., 2000). See analysis section of chapter 3 for a review of the actual discrete choice regression models assessed for this thesis.

The regression coefficients gained from the regression model allow the analyst to establish; the importance of attributes, relative importance of attributes, willingness to pay (within confines of the attribute levels used), total estimated utility scores (Ryan, 1999a), cost-utility ratios (Farrar et al., 1997, 2000) and cost-benefit ratios (Ryan, 1999b).

Importance of attributes.

An attribute is considered important to respondents if it is found to contribute significantly (P <0.10) (see Appendix B for statistical significance levels) to the regression equation explaining the variance in respondents utility functions.

Relative importance of attributes.

The importance of attributes in relation to each other can be seen by comparing the size of the regression coefficients and t-scores for each of the attributes. If continuous coding is used it is important to bear in mind the size of the coded units e.g. it may be more practical to consider time in hours rather than minutes.

Indirect willingness to pay.

If a charge or contribution in the form of direct payment or taxation for the service is included as an attribute, willingness to pay can be estimated for the services (Bryan, Buxton, Sheldon & Grant, 1998; Farrar et al., 2000; Johnson, Banzhaf & Desvousges, 2000; Ryan, 1999b; Ryan & Hughes, 1997; San Miguel, Ryan & McIntosh, 1997, 2000; Scott, 2001).

Marginal rates of substitution (MRS).

Marginal rates of substitution are determined by the ratio of the regression coefficients of attributes to each other[4, 5]. MRS provides an estimate of the

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[4] It has been suggested that marginal rates of substitution using the current method $\left(MWTP = \frac{\beta_1}{\beta_p} \right.$ MWTP= marginal willingness to pay, β_1 = estimated attribute coefficient e.g. time, β_p = estimated attribute coefficient for price/cost$\left.\right)$ are inappropriate (Lancsar and Savage, 2004a, 2004b) in any circumstances other than a state of the world model where there is only one choice to be made (Ryan, 2004a; Lancsar and Savage, 2004b). It has been suggested that the alternative method of compensating variation (Hicks, 1939; Small and Rosen, 1981) should be used $\left(CV = \frac{1}{\lambda}\left[\ln \sum_{j=1}^{J} e^{V_j^0} - \ln \sum_{j=1}^{J} e^{V_j^1} \right] \right.$ λ = marginal utility of income, V_j^0 and V_j^1 = the value of the indirect utility function for choice option J before and after the quality change of the choice made,

improvement in one attribute required to compensate for a given level of another attribute e.g. Ryan et al., (1998a) found that general practice patients were willing to wait in excess of half a day (0.59 days) to see the doctor of their choice at their practice. This procedure assumes full cardinal utility and is easiest to conduct when continuous codes have been used. The theoretical merits of MRS will be discussed later in this Chapter and its practical merits will be assessed in Chapters 5 and 6.

Assuming that the DCM coefficients have cardinal properties MRS can be used to estimate cost-utility, willingness to pay and cost-benefit ratios. If cost estimates are available for the health service being evaluated they can be combined with the utility scores for statistically significant attributes to provide cost utility scores/ratios (Parker & Srinivasan, 1976; Farrar et al., 1997, 2000). By expressing the cost of a particular set of attribute levels (a mode of service delivery) as a proportion of the estimated utility score for that set of attribute levels a cost utility ratio is obtained. When a WTP attribute is statistically significant MRS can be used to estimate willingness to pay for each attribute level. When cost estimates are also available, WTP and cost estimates can be combined (MRS) to produce cost benefit ratios (Bryan et al., 1998; Farrar et al., 2000; Johnson et al., 2000; Ryan, 1999b; Ryan & Hughes, 1997; San Miguel et al., 1997, 2000). A cost benefit ratio is derived by expressing respondents' willingness to pay for a particular attribute or mode of service delivery (set of attribute levels) as a proportion of the cost of that particular attribute or mode of service delivery.

Reliability and Validity

As is the case with any research technique, DCM must satisfy the criteria of reliability and validity. Whilst much research has been devoted to establishing the reliability and validity of conjoint analysis (data gathered via ranking and rating

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J is the number of options in the choice set) as long as there are no model specification problems (Santos Silva, 2004). See Chesher and Santos Silva (2002) for specification tests.

[5] Ratcliffe, Buxton, McGarry, Sheldon and Chancellor (2004) suggest that non-parametric bootstrapping be used to generate confidence intervals around marginal rates of substitution of interest.

rather than discrete choice), particularly in the field of marketing (Acito, 1979; Bateson, Reibstein & Boulding; 1987; Leigh, Mackay & Summers, 1984; Segal, 1982), there is less evidence in relation to DCM.

Reliability

Bateson et al. (1987) propose that the reliability of a preference technique such as DCM should be assessed in terms of reliability over time, reliability over attribute set, reliability over stimulus (scenario) set and reliability over data collection procedure.

Reliability over time.

Bryan et al. (2000) found high levels of temporal reliability in terms of discrete choices recorded (input level data) (kappa coefficients of 0.71 and 0.65) and results of statistical analysis, with strong similarities in coefficient values and overlap in 95% confidence intervals.

Reliability over attribute set.

Farrar and Ryan (1999) examined reliability over attribute set or information ordering/primacy. Having issued two questionnaires, one questionnaire having attributes presented in an inverse ordered to the other to separate samples, Farrar and Ryan found no statistically significant effect as a result of the ordering of attributes.

Reliability over stimulus (scenario) set.

Whilst Ryan et al. (1998b) found no evidence that the order in which scenarios were presented to respondents (general practice patients) significantly affected the reliability of preference results in a conjoint analysis study; it has yet to be proven for a DCM exercise.

Construct validity

Construct validity is the extent to which the instrument is measuring or testing the intended theory or construct. Construct validity is most commonly addressed in the

form of concurrent (convergent and discriminant) validity, criterion and theoretical validity.

Convergent validity.

Ryan (1996a) and Ryan et al. (1996) note that convergent validity has been addressed within the context of environmental economics. Magat, Viscusi and Huber (1988) (DCM) studying the utility of reduced risk in relation to chemical products explored the convergent validity of WTP with DCM. Conventional WTP responses were found to be 25% to 58% of those found in the conjoint analysis and DCM studies. Ryan (1996c) looked at the convergent validity of DCM and WTP on assisted reproductive techniques and found DCM yielded a figure 1.5% higher than that of a closed ended WTP question.

Predicting behaviour/criterion validity.

Predictive validity refers to an instrument's ability to predict changes in key variables e.g. predict actual service use. A number of studies in transport economics and marketing have examined the predictive ability of discrete choice modelling and found they could predict behaviour (Ryan, 1996a; Ryan et al., 1996), for example Louviere and Woodworth (1983) and Louviere (1988).

Theoretical validity.

Theoretical validity (also referred to as internal consistency)[6] relates to how well the instrument conforms to theoretical expectations. As DCM assumes that preferences are transitive and that preferences are continuous, it is common to test that respondents are making choices in this manner. If a respondent *always* chooses a scenario which has a favourable outcomes for a specific attribute no matter how poor the levels of the remaining attributes, their preferences are not continuous and they do not trade between attributes. For these individuals an attribute is said to be dominant (Ratcliffe, 2000a; Ratcliffe & Buxton, 1999). If an absolute order of

[6] Some disciplines such as Psychology consider internal consistency to be reliability rather than validity.

preference for attributes is found and no degree of substitution, individuals are said to be lexicographic or adhering to a hierarchical decision rule (Parker & Srinivasan, 1976; Cave et al., 1994; Ratcliffe, 2000a). Violations of continuous and transitive preferences are tested by including specific scenarios in the DCM questionnaires, where one scenario dominates another and where a transitive structure exists between scenarios e.g. if A > B and B > C, therefore A > C. Examples of such checks are very common in the literature (Bryan et al., 1998; Ryan, 199a; Ryan, 199b; Ryan et al., 1998a; San Miguel et al., 2000). Violation of continuous and transitive preferences can also be checked by manual examination of the choices made.

Other tests of theoretical validity include convergence with established preference patterns. For example, as people do not like to wait or pay, utility coefficients should be negative related to waiting time and charges. As people using a health service want a positive outcome, utility coefficients will be positively related to the probability of a positive outcome. Examples of such checks are very common in the literature (Longworth, Ratcliffe & Boulton, 2001; Ryan & Hughes, 1997; Ryan, 1999b).

Utility Theory

Discrete choice modelling is based upon random utility theory. To highlight the issues surrounding random utility theory and as a result DCM, utility theory will be briefly reviewed in its historical context. Random utility theory developed by Thurston (1927), Marshak (1960), Luce (1959), Lancaster (1966) and McFadden (1972, 1974), is part of the large body of theory dealing with decision making or as it is more commonly referred to in the field of economics, consumer choice (Walker & Ben-Akiva, 2002). Decision theories are predominantly utilised in three ways, normatively, prescriptively and descriptively (Keeney, 1992). Normative usually refers to an idealised description of individual decision making that should not necessarily be followed in practice. Prescriptive refers to the decision process to be recommended to a decision maker, even when normative rules are violated in this

process. Not all academics adhere to this distinction; some consider prescriptive utilisation of a decision theory to be nothing more than an approximation of the normative. Descriptive refers to a theory's ability to describe behaviour (Howard, 1992). Descriptive quality is assessed by its accuracy in characterising and predicting behaviour. Although at times it will be necessary to consider normative and prescriptive aspects of utility theories, in this thesis it is the descriptive qualities that are of primary interest.

Utility theory is not solely associated with economics it has been of particular interest to philosophers, economists, mathematicians, statisticians and from the latter half of the twentieth century psychologists and management/decision theorists (Edwards, 1954a; Manstead & Hewstone, 1995). Academics have been grappling with the issue of utility since the days of Jeremy Bentham (1748 – 1832). Bentham, the political activist, legal scholar, linguist and social philosopher defined positive utility as "the property of any object whereby it tends to produce benefit, advantage, pleasure, good or happiness: or to prevent the happening of mischief, pain, evil or unhappiness to the party whose interests is considered" (Earl-Slater, 1999, p. 153). Negative utility is the converse of Bentham's quotation.

The earliest work upon utility dealt solely with risk-less choice or a sure prospect, risk-less referring to a choice between a defined set of options where there are no unknown parameters. The school of philosopher-economists pioneered by Bentham and popularised by James Mill and others (Edwards, 1954a) proposed that the goal of human activity was to maximise utility (*utility maximisation theory*). Utility maximisation theory was integrated into the formal economic analysis of the early great economists, including Jevans, Walras, Menger and Marshall (Blaug, 1985; Edwards, 1954a). Utility maximisation theory assumed that any person to whom the theory was applied was an 'economic man'. Economic man had three main characteristics; he was completely informed, was infinitely sensitive and was rational. Rationality of economic man relates to two things; the ability to weakly order choices or states and making choices with the goal of maximizing utility. To

weakly order choices economic man had to be able to state preference or indifference and preferences must be transitive e.g. if A is preferred to B (A > B) and B > C therefore (∴) A > C. The infinite sensitivity of economic man in conjunction with the choices under consideration being continuous and, like price, infinitely divisible resulted in a continuous utility function (cardinal utility). Jevans, Walras and Menger paid little attention to the precise shape of the utility function and assumed a law of diminishing marginal utility (Blaug, 1985). Walras and Menger drew linear utility curves whilst most of Jevans' curves were drawn convex from below (Blaug, 1985) (see Figure 2.3).

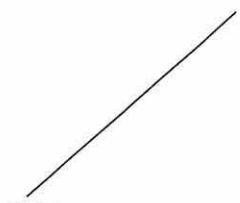
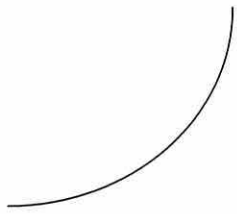
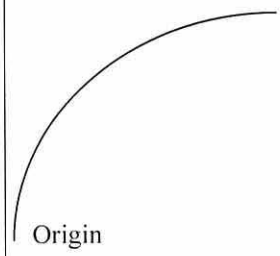
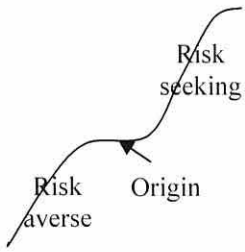
Linear utility curve	**Convex utility curve**	**Concave utility curve**	**Double inflection utility curve**
Walras and Menger	Jevans	Bournolli and Von Neumann and Morgenstern	Friedman Savage and Markowitz (1952)
			

Figure 2.3. Utility curves.

The problem with utility maximization theory was that it could not adequately deal with the relations between the utilities of different goods. Jevans, Walras, Menger and Marshall assumed an 'additive utility function where utilities of different goods were assumed to be independent and as such could simply be summed to give total utility (Edgworth, 1881; Morgan, 1945; Samuelson, 1947). This ignored the non-independent utilities of competing goods such as the large array of snack foods or completion goods such as right and left shoes or the need for a TV to utilise a DVD player. In the process of pointing out that the total utility of non-independent commodities was not an additive function Edgworth (1881) introduced the concept of indifference curves based upon the notion of measurable cardinal utility. Pareto

(Ricci, 1933) disagreed with the assumption that utility was cardinal and thus measurable up to a linear transformation. He believed individuals could prioritise states or options but not how much they preferred one over another; he proposed a utility function measurable only on an ordinal scale (ordinal utility). Pareto pointed out that indifference curves based upon ordinal utility measurement allowed the same information to be obtained as was available from marginal utilities (higher indifference curves implied greater utility, but did not reveal how much greater the utility was) and all the theorems based upon cardinal utility could also be deduced. Papers by Johnson (1913) and Slutsky (1915) examining the mathematics of ordinal utility indifference curves, added further weight to the abandonment of the concept of cardinal utility in favour of the benefits of indifference curves. Pareto's '*theory of choice*' was refined by Hicks and Allen (1934) who eliminated inconsistencies in the theory such as the claim that the sign of the utility function could be obtained from an ordinal utility function. In the *revealed preference* approach Samuelson (1938a; 1938b) developed a new analytic foundation. In essence, Samuelson proposed that indifference curves could be derived from observing choices between alternative groups of purchases available to the consumer. Wold (1943a, 1943b, 1944, 1953) concluded that Pareto, Hicks and Allen, Samuelson and Cassel's demand function approach were mathematically equivalent. Although there were several attempts to revert to cardinal utility e.g. Knight (1946) and Roberts (1952) the indifference curve approach in its various forms was firmly established as the method of accounting for risk-less choice decision making (Edwards, 1954a).

In reality there is almost always an element of risk involved in decision making e.g. when purchasing or letting a house there is no way of knowing who will move into neighbouring properties in the future and if their behaviour will cause irritation, distress etc. Theories of risky choices (or uncertain prospects) were developed to account for choices involving risk or uncertainty (probability in statistical terms). In risky choices the individual was initially assumed to be trying to maximise expected value. The expected value of a choice can be calculated by multiplying the value of each possible outcome by its respective probability of occurrence and summing the

results across all possible outcomes (Edwards, 1954a) e.g. expected value = $\sum_n p_1£_1 + p_2£_2 + ... + p_n£_n$ [7]. Clearly observation of human behaviour reveals that the public do not make decisions in accordance with this principal e.g. buying insurance when the insurance company makes a profit (for the company to make a profit they must charge more than the expected value of the policy).

Consideration of the problem of insurance and the St. Petersburg Paradox[8] led the mathematician Daniel Bournolli (1738, translated into English in 1954) to propose the assumption that individuals act to maximise expected utility and that the utility function was logarithmic and convex in shape (Blaug, 1985; Machina, 1987a)[9, 10]. Bournolli's theory did not address how to measure utility or why the expectation principal was rational (Schoemaker, 1982).

In 1926 Ramsey (1950) proposed the first general set of axioms for preference comparisons between acts. Ramsey's aim was to illustrate how beliefs could be measured on the basis of the extent to which individuals are prepared to act on them (Fishburn, 1989). To this end he proposed axioms governing preference comparisons between uncertain acts. Ramsey's work was to play a key role in the future work of Savage (1954).

The next key development in the study of risky decisions occurred with the publication of Von Neumann and Morgenstern's (1944, 1947) *expected utility theory* (EU)[11]. Schoemaker (1982) considers Von Neumann and Morgenstern's EU to be

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[7] Expected value became moral expectation in Bournolli's work and average utility in Von Neumann and Morgenstern's work (Strotz, 1953). In moral expectation and average utility, expected utility was substituted for money in the formula.

[8] St. Petersburg Paradox: although a rational gambler should pay to play a game where the entry fee is less than the expected value people will not always do this. See the St. Petersburg game at http://plato.stanford.edu/entries/paradox-stpetersburg/.

[9] Gabriel Cramer proposed the same theory independently of Bournolli (Machina, 1987a).

[10] Bournolli's utility function received some corroboration over a century later with the development of the Weber-Fechner law in Psychophysics (Blaug, 1985; Edwards, 1954a).

[11] Alchian (1953) and Fishburn (1989) notes that a closely analogous method was developed by Ramsey (1950) in 1926. Ramsey's work had one advantage over Von Neumann and Morgenstern's, it utilised subjective probability for uncertain events (Fishburn, 1989).

the major paradigm in decision making since the Second World War. Von Neumann and Morgenstern proposed that in addition to being able to state preference or indifference between states, economic man could make the same distinctions between the probabilities of states and combinations of states. This modification meant that cardinal utility could be assumed once again. However the concept of cardinal utility had matured in the interim period between Hicks and Allen (1934) and Von Neumann and Morgenstern (1944). In the interim period the debate between the vanishing cardinalists and the ascending ordinalists related to whether or not cardinality and as such quantifiable measurement was a property of utility (Strotz, 1953). This issue was spurious as "....measurement has meaning, not as a property of things, but as a predictive procedure .... Measurement is always invented and never discovered!" (Strotz, 1953, p385). Von Neumann and Morgenstern's EU implicitly assumes full cardinal utility (interval to ratio data characteristics) exists otherwise it would be impossible to psychologically determine the certainty equivalence of a risky choice (Schoemaker, 1982). However, preferences are determined by at least two factors, strength of preference for a choice and attitudes towards risk. The Von Neumann and Morgenstern EU function is a combination of these two factors without the aid of interval comparison and strength of preference measures; thus as a preference measure it is wholly ordinal, providing no more than ordinal ranking of choices (Schoemaker, 1982). This is an extremely important point which will be raised in subsequent Chapters in relation to using the results of DCM to conduct marginal rates of substitution.

Von Neumann and Morgenstern's axiomatic system representing their utility function as a theorem is presented in Appendix C. In simple terms, expected utility is the sum of the weighted utilities of the components of the risky choices (uncertain prospects) where the weights are probabilities associated with each component (Alchian, 1953). Edwards (1954a) notes three key empirical implications of expected utility theory. Firstly, like risk-less choices, risky choices can be ordered in terms of desirability e.g. $A > B$, $A < B$ and $A = B$. Secondly, the concept of expected utility is behaviourally meaningful. Finally, choices between risky alternatives are

made based upon the goal of maximising expected utility. Expected utility = $\sum p_i u(x_i)$ (Key: p = probability, u = utility, x = choice, i = i^{th} alternative).

Both Bournelli and Von Neumann and Morgenstern's utility function were convex in shape (Blaug, 1985). One of the economic applications of Von Neumann and Morgenstern's theory by Friedman and Savage (1948) was to address the question of why the same individual that buys insurance to be risk averse will also buy a lottery ticket which is risk seeking behaviour. They concluded that this could be explained by a utility curve with a double inflection (an 'S' shape. See Figure 2.3).

Edwards (1954a) identified seven problems with Von Neumann and Morgenstern's theory as a descriptive model. Firstly, the individual's subjective probabilities of a given outcome may differ to the actual (objective) probabilities. Preston and Baratta (1948), Griffith (1949), Attneave (1953) and Sprowls (1953) found this to be the case in their experimental studies. Secondly, the combination of the values of a choice with the probabilities may not be multiplicative as proposed. Thirdly, in a series of experiments where individuals chose between bets with specified probabilities Edwards (1953a, 1953b, 1954b, 1954c) established that individuals had probability preferences (not necessarily for the most positive probability of success). Fourth, the aggregation of the products of the value and the probabilities may not be additive as proposed. To date no one has successfully designed and conducted an experiment to address items two or four. Fifth, the risk or gamble involved in the choice may have a positive or negative utility in itself. Sixth, expected utility theory (and other utility maximisation theories) will only work if preferences are transitive. For the model to predict behaviour it is necessary that intransitivities in the data be infrequent enough to be considered as errors. Edwards (1954a) did not propose an acceptable level of intransitivity. However, his review of the literature revealed intransient response rates of 4% (Papandreou, 1953) to 27% (May, 1954) (the latter rate may have been lower had an indifference response category been included). Finally, people may not be making decisions with the goal of maximising their expected utility.

The next significant step in utility theory was the development of subjective expected utility. The Blackwell Encyclopaedia of Social Psychology (Manstead & Hewstone, 1995) attributes *subjective expected utility theory* (SEU) to Ward Edwards based upon his excellent review of the literature and the key issues in the field of decision making (1954a). Fishburn (1981) in his mathematically orientated review of subjective expected utility theories attributes the first complete SEU theory to Leonard J. Savage (1954)[12]. Like Edwards, Savage built upon the work of Von Neumann and Morgenstern. Savage's work was also based upon the work of Ramsey in 1931 (1950) and de Fionetti (1937) (Fishburn, 1981; Luce & Raiffa, 1957). "A primary, and elegant, feature of Savage's theory is that no concept of objective probability is assumed; rather a subjective probability measure arises as a consequence of his axioms. This in turn is used to calibrate utilities, and it is established that it can be done in such a way that expected utilities correctly reflect preferences. Thus Savage's contribution – a major one in the foundations of decision making – is a synthesis of Von Neumann and Morgenstern's utility approach to decision making and de Fionetti's calculus of subjective probabilities" (Luce and Raiffa, 1957, p304). Subjective expected utility = $\sum f(p_i)u(x_i)$ (Key: p = probability, u = utility, x = choice, i = i^{th} alternative).

Table D1 in Appendix D contains a list of 24 prominent SEU theories, developed post Savage's theory[13]. For a review of the main characteristics, advantages and disadvantages of the prominent SEU theories see Fishburn (1981, 1989), Machina (1987b), Sarin (1989) and Weber and Camerer (1987). EU and SEU have been found to be descriptively invalid (Miyamoto, 1992). EU/SEU theories have been

[12] Edwards (1955) SEU was formalized and published in 1955. Edwards used a cardinal utility measure constructed under certainty; this was inferior to Savage's cardinal utility measure constructed under risk (Schoemaker, 1982).

[13] Based upon the ability to characterise a wide range of situations realistically, simplicity and intuitive appeal of preference axioms, interpretability of structural conditions and features that can easily connect to utilities and subjective probabilities, Fishburn (1981) identified the following six SEU models in his review as the better ones: Savage (1954), Narens (1976), Suppes (1956), Pratt, Raiffa and Schlaifer (1964), Fishburn (1969), and Fishburn (1973). Despite being the first complete SEU theory, Savage's work is still regarded as one of the best.

found to be invalid as individuals violate the axioms and properties (characteristics stemming from the combination of two or more axioms) of the theories. The main axiom/property violations include (Keller, 1992): substitution violation, sure thing violation, violation of linearity in probabilities, betweenness violation, ambiguity of indifference violation, fixed reference levels violation, risk attitude and the transitivity property violation. A brief discussion of these violations is provided in Appendix E.

The next key step in relation to utility and discrete choices was the development of multiattribute utility theories. Although multiattribute additive utility had been assumed in nineteenth century consumer choice/demand theories it came to prominence in the 1960s (Fishburn, 1989). Debreu (1960) made the first rigorous connection between choice and additive utility. Debreu's work was axiomatised and developed by Luce and Tukey (1964), Luce (1966) and Scott (1964). It was found that under certain circumstances that expected utility (Von Neumann & Morgenstern, 1944, 1947) could be decomposed additively to form a multiattribute theory with linear additive or multiplicative utility (Fishbern, 1964, 1965; Keeny, 1968; Keeny & Raiffa, 1976; Pollak, 1967).

Random utility theory (RUT) sometimes called random utility maximisation and the discrete choice model (Walker & Ben-Akiva, 2002) is a multiattribute theory. In this Chapter only the general model will be considered; for a more detailed consideration of the general RUT model and variants of it see Manski (1977). The theory is most strongly associated with mathematical psychology and economic disciplines such as marketing who saw its potential for the statistical analysis of stated and revealed preference discrete choice data. Walker and Ben-Akiva (2002) ascribe the theory to Thurston (1927), Marshak (1960) and Luce (1959) who pioneered the random utility paradigm, and to Lancaster (1966) and McFadden (1972, 1974) who developed the manner of specifying utilities for the model. McFadden (1972, 1974) developed the multinomial logit model and went on to develop variants such as the nested logit

models which form the basis of the econometric software packages used to analyse DCM data today.

In essence RUT proposes that utility and probability are subjective (SEU) (Luce, 1959; Lancaster, 1966), although probability does not appear explicitly in the RUT equation below. Utility is not derived from the commodity chosen but from the characteristics of the commodity (Lancaster, 1966). The utility of the characteristics can be summed to give a total utility measure (linear additive utility). Whilst the utility derived is subjective, the characteristics or attributes and their utility measurement are the same for all consumers. One of the most distinctive features of this model is that it acknowledges that the researcher applying utility theory is unaware of consumers/respondents' idiosyncrasies of taste, assumes that these idiosyncrasies are randomly distributed amongst individuals and allows for this by including a random error term (Ben-Akiva & Lerman, 1985). The random error term also accounts for measurement and specification error[14]. The main axioms of RUT, recorded as text rather than equations were presented in Table 2.3.

$U_{in} = V_{in} + \varepsilon_{in}$ (Key: U_i = estimated utility of choice i, V_i= systemic or representative component if the utility [attributes and their levels] of choice i, ε_i= disturbance or random component of the utility of choice i).
(Source: Ben-Akiva & Lerman, 1985)

The latest Utility theories developed are collectively referred to as *generalised utility theories* (Keller, 1992). Generalised utility theories are distinguishable from their predecessors by the fact that they relax one or more of the properties/axioms that have been violated when EU or SEU have been tested experimentally (Keller, 1992; LaValle, 1992). Representative examples of generalised expected utility theories are listed in Appendix F. For a detailed discussion of generalised random utility theories see Walker and Ben-Akiva (2002). Particularly in cases of choices with small

[14] For a more detailed consideration of the general RUT model and variants of it see Manski (1977).

probabilities and high value outcomes generalised utility theories are a descriptive improvement upon EU and SEU, although at the expense of normatively appealing axioms or assumptions (Eppel et al., 1992). The most promising of the generalised utility theories are *rank dependent theories* (Camerer, 1992) and the premier model at present is believed to be Kahneman and Tversky's (1979) *prospect theory* (Eppel et al., 1992). In his review of generalised utility theories and experimental tests of these theories Camerer (1992) points out that all these theories which were specifically designed to explain violations of EU/SEU are themselves violated in other ways.

From the 11th to the 15th of June 1989 the leading academics working in the field of normative, descriptive and prescriptive utility theory (EU, SEU and generalised) participated in the 'Utility: Theories, Measurement and Applications' conference (Edwards, 1992). The delegates unanimously agreed that EU and SEU were still the *best normative* theories for decision making under risk/uncertainty (Edwards, 1992; Eppel et al., 1992) and that utility maximization (EU, SEU and generalised) is currently *indefensible as a descriptive decision theory* (Edwards, 1992).

Sarin (1992), Eppel et al. (1992) and Schoemaker (1982) suggested that many of the violations of SEU when experimentally tested were the result of psychological attributes that were not accounted for in the experiments and analysis. The generalised utility theories, *regret theory* (Bell, 1982; Loomes & Sugden, 1982) and *prospect theory* (Kahneman and Tversky, 1979) highlighted the issues of regret, subjective distortion of probabilities and utilities, choices and options being judged from a reference point and framing or context effects leading to completely different choices being made for mathematically equivalent choices. In light of this evidence Wroe et al. (1998) concluded that SEU and its forerunner of EU had failed as a descriptive model under experimental conditions as it had been applied in too narrow a fashion. They proposed utilising SEU explicitly within Becks cognitive model (Beck, 1976). Wroe et al. emphasise the influence of subjective beliefs, proposing that the individuals' decisions are based upon their beliefs and the

information that is available and deemed to be relevant to them at the time. "These premises may be totally idiosyncratic, may actually be factually wrong and may also be systematically influenced by other factors (such as anxiety)" (Wroe & Salkovskis, 1999, p20). Wroe and Salkovskis propose that apparently inconsistent decisions such as preference reversal (e.g. Lichenstein, 1973; Lichenstein & Slovic, 1971), were not the result of an illogical decision process but the information and beliefs upon which the perceived outcomes and ultimately the decisions were based.

In a series of studies (Salkovskis et al., 1999; Wroe, 2002; Wroe & Salkovskis, 1999, 2000; Wroe et al., 1998, 2000) '*modified utility theory*' has successfully predicted health behaviour including, interest in obtaining hypothetical genetic testing and attendance at screening. Wroe and Salkovskis (2000) correctly predicted the actual decision of whether or not to take a test for bone density screening at an NHS hospital for 96.4% of their study sample based upon respondents intention to have testing.

Determinants of Choice (Extension of RUT)

If respondents of DCM questionnaires adhere to RUT this will identify the decision making process, weighing up the benefits and disadvantages of each decision, however, like its predecessors RUT does not provide a detailed description of the way individuals come to make their decisions (Feather, 1982; Edwards, 1992; Jones, 1993; Frisch & Clemen, 1994; Conner & Norman, 1995). RUT does not identify the key determinants of choice e.g. perceived severity of the illness being considered. To address this deficiency and highlight some of the determinants of choice the psychology/health psychology literature should be accessed.

To extend the identification of the determinants of choice beyond the weighted benefits and barriers used in RUT social cognition models that assume that decisions are made in accordance with utility theory (Edwards, 1954a; Conner & Norman, 1995) must be examined. In social cognition models individuals decompose the all encompassing benefits and barriers into the respective beliefs identified in each

model and combine the benefits/positive[15] elements with the disadvantages/barriers/negatives into a choice using subjective expected utility to determine which course of action/choice will maximise their utility; see Figure 1.2 in Chapter 1 for a graphical representation of this relationship. The social cognition models to be assessed are those identified by Weinstein (1993), van der Plight (1994) and Conner and Norman (1995) as being rooted in utility theory[16]; the health belief model (HBM), health locus of control (HLC), protection motivation theory (PMT), theory of reasoned action (TRA)/theory of planned behaviour (TPB) and self-efficacy theory (SET). A brief critical review of the identified models is included below.

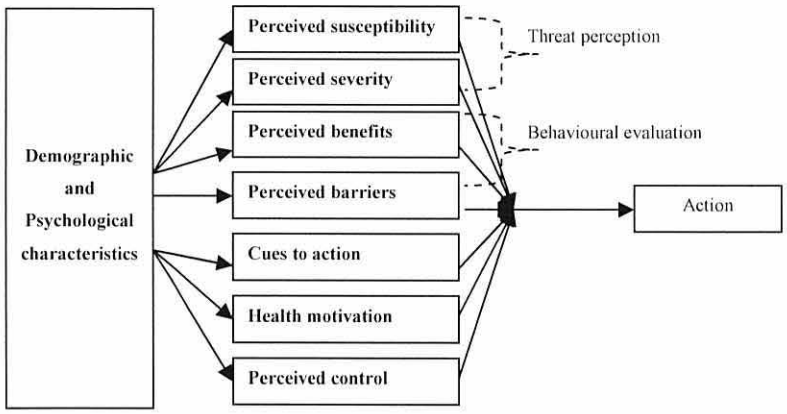

Figure 2.4. The health belief model. (Ogden 2000)

The health belief model (HBM) (Hochbaum, 1958; Rosenstock, 1966; 1974; Becker, Haefner and Mainman; 1977) was developed in response to public health research identifying that the extent to which individuals used health services or adhered to preventative health behaviour was affected by demographic characteristics (Rice,

[15] The Health belief model does include the factors perceived benefits and perceived barriers. However, operationalisation of these factors differs to its operationalisation in utility theory. Barriers tend to focus on barriers to accessing health services or participating in health behaviour and not all negative issues associated with a certain health behaviour. Benefits are confined to those identified in the questionnaire and are mutually exclusive with the benefits associated with the other behaviour and motivation factors e.g. health motivation benefits etc.

[16] Sometimes when social cognition models are discussed in the literature the term expectancy value is used interchangeably with utility theory. In that context they can be treated as equivalent as they are referring to the subjective utility and probability of an outcome.

1998; Sheeran & Abraham, 1995; Taylor, 1999). In order for health education/promotion to influence health behaviour it was necessary to identify how socialisation characterised and demographic variables led to differing health adherence and behaviour. The solution was a model linking socialisation to action or behaviour via individual beliefs; as beliefs are not fixed and can differentiate between individuals from the same background (Sheeran & Abraham, 1995).

The original model, first applied by Hochbaum (1958), focused on two aspects of individuals' beliefs, threat perception and behavioural evaluation (see Figure 2.4). Threat perception comprises of the beliefs, perceived susceptibility to illness and perceived severity of the illness/condition. Behavioural evaluation comprised of the two beliefs, perceived benefits or efficiency of the prescribed health behaviour and the perceived barriers or costs of adhering to the prescribed health behaviour. In addition to the beliefs the model later included the motivational factor, cues to action. The model proposes that when the appropriate beliefs are held that cues to action such as health promotion campaigns or symptoms can trigger health behaviour. Later versions of the model also include health motivation (concern about health matters) (Becker et al., 1977) and perceived control (Ogden, 2000) as beliefs influencing action.

The main limitation of the HBM is that it is an incomplete social cognition model (Sarafino, 2006). There is no standardised method of measuring the components of the HBM and as a result researchers have developed different questions to measure the same factors, making the comparisons of findings across studies difficult. Several versions of the HBM have been used, with cues to action and health motivation in particular being optional factors (Morrison & Bennett, 2005). Rosenstock (1966) did not specify how the variables of the HBM interact and combine and as a result calculating factor scores and their application as predictive variables is a matter of debate (Abraham, Sheeran, Abraham & Spears, 1996; Strecher & Rosenstock, 1997; Ogden, 2004). For example, Becker et al. (1977) noted that perceived benefits should be weighted against barriers but did not specify

how this should be calculated. In addition, although a continuum relationship is assumed between 'demographic and psychosocial' characteristics and social cognition factors, the HBM is a static model when considering social cognition factors with all factors having a simultaneous influence on the individual (Ogden, 2004), later models such as the TPB suggest that there is a sequential interrelationship between factors (Morrison & Bennett, 2005).

Risk perception has not proven to be a consistent predictor of health behaviour (Morrison & Bennett, 2005). This may be a result of individuals having inaccurate risk perception when their risk increases beyond moderate levels (Sastre, Mullet & Sorum, 1999; Weinstein, 2000). Within the HBM, only limited account is taken of social influences such as peer group on health behaviour (Morrison & Bennett, 2005; Ogden, 2004). There is no accounting for habitual health behaviour such as brushing teeth, which have continued without the individual considering health threat, benefits and costs (Sarafino, 2006; Ogden, 2004). The HBM also fails to account for perceived behavioural control/self efficacy (Ogden, 2004; Morrison & Bennett, 2005), previous participation in specific health behaviour (Morrison & Bennett, 2005) and emotional factors such as fear and denial (Ogden, 2004).

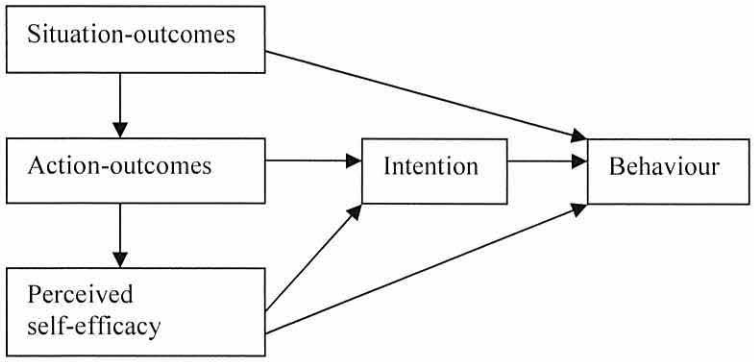

Figure 2.5. Representation of self-efficacy theory

Self-efficacy theory (Bandura, 1977) (see Figure 2.5) assumes that health behaviour/choice is based upon three types of expectancies/outcomes, situation-outcomes, action-outcomes and perceived self-efficacy (Conner & Norman, 1995). Situation-outcome expectancies are the perceived consequences of health threat

when no intervention is instigated, they are assumed to influence action-outcomes and behaviour. Action-outcome expectancy is the belief that a specific behaviour will lead to a specific outcome, and influences intention and perceived self-efficacy. Self-efficacy expectancy is the belief in the extent to which behaviour/intervention is within the individual's control and influences both intention and behaviour. Intention influences behaviour.

Although frequently incorporated into action-outcomes, the lack of an explicit inclusion of social influences on behaviour is seen as a drawback of SET (Conner & Norman, 1995). The lack of a measure of value/utility, particularly for the action-outcomes factor, is seen as a deficiency in this utility based model (Conner & Norman, 1995).

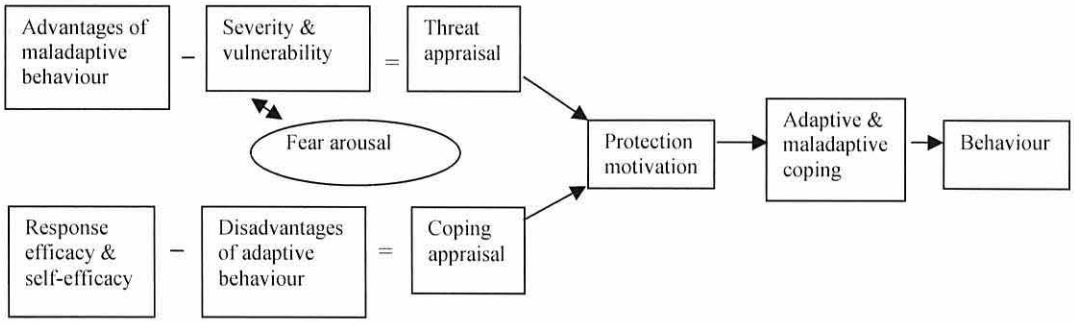

Figure 2.6. Protection motivation theory

The PMT (see Figure 2.6) was originally designed (Rogers 1975) to explain fear appraisal. In its most typical form e.g. Maddux and Rogers (1983) and Rogers (1983), the PMT describes behaviour stemming from adaptive and maladaptive coping/intention based upon protection motivation as a result of threat appraisal and coping appraisal (Conner & Norman, 1995). Threat appraisal (see Figure 2.6) is the consideration of perceived severity and vulnerability of the health threat in conjunction with the perceived advantages of maladaptive behaviour. Fear is assumed to increase perceived severity and vulnerability. Coping appraisal is the consideration of the behavioural alternatives that could reduce the health threat. Coping is presumed to be based on response (or action-outcome) efficacy

(expectation that carrying out certain behaviour(s) can remove the health threat) and self-efficacy (the individual's belief that they can successfully carry out the required actions) (Conner & Norman, 1995) considered in conjunction with the perceived disadvantages of health behaviour. Both appraisals (threat and coping) induce protection motivation, usually operationalised as the intention to perform health protective behaviour and/or refrain from health threatening behaviour e.g. smoking. Intention is assumed to influence actual behaviour. The PMT has been described as a hybrid theory (Prentice-Dunn & Rogers, 1986; Conner & Norman, 1995) with self-efficacy taken from self-efficacy theory and susceptibility/vulnerability, severity and response-efficacy coming from the HBM.

Given the close links with the HBM it is not surprising that both models share common criticisms. In particular the PMT has been criticised for not accounting for habitual behaviour or social and environmental factors (Ogden, 2004). Ogden, (2004) proposes that habitual health behaviour such as brushing teeth continue without the individual considering health threat, benefits and costs. As with the HBM however, social factors such as peer group are ignored or unsatisfactorily accounted for within PMT.

The multidimensional health locus of control (MHLC) (Wallston, Wallston & deVillis, 1978) is an extension of the two dimensional Health Locus of Control (HLC/LoC) of Rotter (1966). The three distinct dimensions of the MHLC are internal health locus of control, external/chance health locus of control and powerful others health locus of control (Morrison & Bennett, 2005)[17]. Individuals differ in regard to whether they believe that their health is controlled by them (internal health locus of control), not controlled by them but by luck or chance (external/chance health locus of control) or consider their health to be under the control of a powerful other such as a doctor (Ogden, 2004; Morrison & Bennett, 2005).

[17] Wallston (1989, 1992) has attempted to incorporate HLC into a more general theory of health behaviour (Conner & Norman, 1995) called Modified Social Learning Theory (MSLT). In MSLT health behaviour is predicted by self-efficacy when the individual values their healyth and has internal HLC.

Ogden (2004) notes that there is no clear distinction between the factors of the MHLC, is going to the doctor for help external ("the doctor is a powerful other who can make me well") or internal ("I am determining my health status by searching out an appropriate intervention")? It has been proposed that the MHLC is only appropriate as a health behaviour model for individuals who value their health (Conner & Norman, 1995). Even when the value of health has been accounted for, the MHLC has been found to be a weak explanatory model of health behaviour, accounting for a small amount of variance in health behaviour[18].

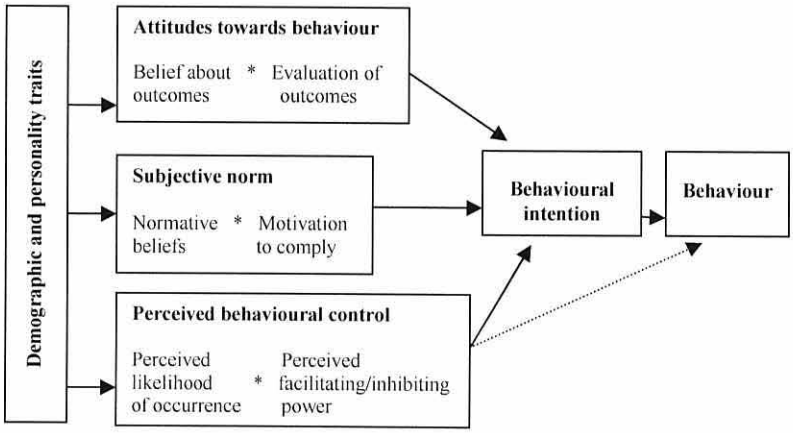

Figure 2.7. The theory of planned behaviour (Conner & Sparks, 1995).

The theory of planned behaviour (TPB, see Figure 2.7) (Ajzen, 1985, 1988, 1991) is an extension of the theory of reasoned action (TRA) (Fishbein & Ajzen, 1975; Ajzen and Fishbein, 1980) which in turn had developed from 'the principals of compatibility' (Conner & Sparks, 1995; Ogden, 2000).

In 'the principal of compatibility' (Ajzen, 1988), both attitudes and behaviour were believed to comprise of four elements (action, target, context and time). Convergence between attitudes and behaviour were believed to be at their maximum when both were measured at the same level for each element. For example a general

[18] Better results have been obtained with disease specific variants of the MHLC scales (Conner & Norman, 1995).

level of attitudes would predict general behaviour, whilst, specific attitudes would be required to predict specific behaviour. Examples of the four elements of a behaviour would be; in the case of oral hygiene, action = brush, target = teeth, context = in bathroom, time = after breakfast (Conner & Sparks, 1995). Often in the case of attitudes it is difficult to go beyond the target element.

The TRA like 'the principals of compatibility' assumes that behaviour is determined by intention to perform the behaviour which is in turn determined by attitudes towards behaviour and subjective norm. 'Attitudes towards behaviour' is the product (multiplicative product) of belief about outcomes with evaluation of outcomes. Subjective norm is the product of normative belief with motivation to comply. Non-volitional behaviour requiring skills, resources or opportunity not freely available to individuals is beyond the scope of the TRA. The TPB rectifies this deficiency by including control. However, in the absence of actual control measures it is frequently necessary to use measures of perceived behavioural control as a proxy. Perceived behavioural control is the product of perceived likelihood of outcomes with perceived facilitating/inhibiting power. PBC is assumed to influence behaviour directly[19] and indirectly due to its influence on behavioural intention[20] (see Figure 2.7). Ajzen (1991) proposed that perceived behavioural control and self-efficacy were interchangeable; however this is not a universally held view (Armitage & Conner, 2001).

The TPB has been found to be a stronger predictor of intention than behaviour (Godin & Kok, 1996; Armitage & Conner, 2001). Intentions and behaviour are only moderately related, individuals do not always act upon intentions (Sarafino, 2006). Several authors have identified emotional and 'intention of action' factors that move an individual from intention to action and increase the variance in intention and behaviour that are explained (Morrison and Bennett, 2005). The most prominent factors identified include; moral norms (Evans & Norman, 2002; Armitage and

[19] Behaviour = Behavioural Intention + perceived behavioural control.
[20] Behaviour intention = Attitudes towards behaviour + Subjective norm + Perceived behavioural control.

Conner, 1998; Manstead, 2000) e.g. safe sex and drink driving, anticipatory regret (Triandis, 1977; Bell, 1982) mediated by likely occurrence of an outcome and consequences of the outcome, self-identity (Sparks & Shepherd, 1992; Morrison and Bennett, 2005) e.g. green consumers intend to eat organic food, and implementation intention, part of the process of turning an intention into action (Morrison and Bennett, 2005). The factor, attitudes towards behaviour does not predict all health behaviours, for example attitudes have been found to predict alcohol use but not smoking and drunk driving (Stacy, Bentler & Flay, 1994).

The social/health psychology models selected to extend the determinants of health beyond the benefits and barriers used in random utility theory/health economics and the rationale for the selection is presented in the discussion.

Discussion

Outcomes of Genetic Services

To date health economics research assessing the advantages and disadvantages of cancer genetics services has concentrated upon health outcomes such as the number of cancers detected, the number of mutations detected, and in particular modelling survival and quality adjusted survival (QALYs); acknowledging death and the impact of cancer upon quality of life as the disadvantages, and diagnosis and survival as the benefits of genetic services. Exceptions to this were the TRACE project (Brain, Gray, Norman, France, et al., 2000; Cohen et al., 2004), utilising a range of psychosocial measures including reasons for attendance at a genetics clinic, Chaliki et al. (1995) looking at psychosocial predictors of willingness to pay $25 for BRCA1 (breast cancer susceptibility one) genetic testing and the work of Wilson et al. (1999) assessing the utility of genetic counselling to past users in terms of process attributes. Similar findings were reported by Hall et al. (1998) in their non-systematic review of genetic testing, in which they report that evaluations of genetic diseases have confined themselves to positive health effects and those utilising a social perspective to the total effects on individuals.

The Value and Utility of Genetic Services to Patients

At present information upon willingness to pay and the utility to patients of genetic testing and presymptomatic surveillance for familial cancer is sparse. The work of Chaliki et al. (1995) has shown that if a token charge were implemented enthusiasm for a hypothetical offer of testing declined. There is currently no information upon the monetary value placed upon cancer genetic services by users and potential service users.

Wilson et al. (1999) are the only ones to publish the results of a discrete choice modelling analysis exercise with patients of cancer genetics services. However, this study was confined to the Scottish model of delivering cancer genetics services, which at the time was confined to single counselling session and no genetic testing. In addition, there are questions relating to the costing assumptions applied in this study.

Preference Elicitation

A number of methods have been utilised to examine the multi-attribute nature of patient preferences for health care to date (Farrar et al., 2000; Ryan, 1996a), including opinion polls, satisfaction surveys, contingent valuation, visual analogue, standard gamble and time-trade off. However, all of these methods have been found inappropriate. Both opinion polls and satisfaction surveys fail to provide any indication of the strength of preference, there is a lack of information on the relationship between attributes and they do not incorporate opportunity cost. Visual analogue, time-trade off and standard gamble are not appropriate when aspects of service provision or prioritisation are the focus, and it is difficult to estimate reliability of indifference answers with time-trade off and standard gamble (Verhoef et al., 1991). Contingent valuation is susceptible to bias from protest zero bids (Olsen and Donaldson, 1998), market inexperience (Drummond, 1995; Ryan, 1996b), policy or strategic bias (Johannesson et al., 1996), politicisation, warm glow effect, and altruism (Cave et al., 1994).

Clearly DCM is a very useful technique for ascertaining and evaluating preferences amongst stakeholders (patients, carers, clinicians, providers, the general public, etc.) in health and social care. DCM allows the researcher to establish if selected attributes and their prescribed levels are significant to the sample of respondents participating in the research. The attributes tested can include non-health and process aspects of care in addition to health outcomes. Total utility in relation to the attributes tested can be estimated, allowing service configurations to be prioritised in terms of estimated utility. It is also possible to ascertain willingness to pay (WTP) within the confines of the levels set in the DCM exercise, as is the case in closed ended WTP exercises (Ratcliffe, 2000b; Ryan, 1999a). Clearly levels must be selected carefully as is the case for bid values in closed ended WTP. As is the case with any WTP exercise care must be taken with the wording of the attribute/question to minimise the possibility of politicised or protest responses such as ignoring the WTP attribute.

In spite of the favourable findings in the published literature, there is clearly a paucity of evidence in relation to the reliability and validity of DCM as a research technique in the field of health care. No research has been conducted into the reliability of DCM in relation to scenario (stimulus) set, or data collection procedure e.g. computer vs. questionnaire, or criterion validity in the form of predicting service uptake in the field of health and social care. Although Bryan at al. (2000) provided evidence of temporal reliability it is important to remember that they were presenting respondents with pairs of scenarios to choose from. Single or multiple scenarios can be presented and in particular a 'no preference' or indifference response option can be included which provides a more realistic and complete set of choice options (Johnson et al., 2000). Clearly temporal reliability should be established for DCM instruments presenting respondents with multiple choices and indifference options. Although there is evidence of concurrent validity and criterion validity (predicting behaviour) for DCM in the marketing and environmental literature amongst other sources, caution must be exercised in relation to this

evidence. Blindly accepting evidence of reliability and validity from applications in other fields as evidence of reliability and validity in health care would be reckless. In marketing in particular respondents are used to having a choice and are used to exercising choice; choice is a relatively new phenomenon in health care and the general public have less experience of exercising this choice, particularly in countries such as the UK where health is organised and funded by the central government.

Utility Theory

The brief review of utility theory, the underlying decision theory of DCM, clearly shows that the theory has undergone significant development and revision over the past two and a half centuries. Moving from risk-less to risky decision making, evolving to accommodate complementary and competing choices/goods, replacing objective utility with expected utility, replacing objective probability with subjective (perceived) probability, developing multiattribute forms of utility theory and in its generalised forms relaxing some of its normatively appealing axioms to accommodate observed contradictory human behaviour. However, by the early 1990s consensus amongst the leading psychologists and decision theorists in the field was that EU, SEU and even generalised utility theories were indefensible as a descriptive model of decision making (Edwards, 1992), confining them to be normative and prescriptive theories.

Less than a decade after delegates at the 'Utility: Theories, Measurement and Applications' conference concluded that utility theory was a descriptive failure (Edwards, 1992), Wroe et al. (1998) resurrected SEU within a cognitive framework and have enjoyed success with it as a predictive model of uptake of health services and health behaviour. The Wroe et al. (1998) and Wroe and Salkovskis (1999) papers do not go into great statistical detail about their revised SEU theory. However, in the former paper they calculate utility by deriving a weighted ratio of benefits to barriers. The weighting of a benefit or barrier is how important a reason is to a particular decision for the decision maker (in that instance anchored from 0 =

not important to 100=extremely important). Weighted ratio $=\left(\frac{\sum wben - \sum wbar}{\sum wben + \sum wbar}\right)$ (Key: wben = weighted benefit, wbar = weighted barrier). Clearly the weighted ratio calculation assumes multiattribute linear additive utility as is the case with random utility theory amongst others. This formulation obviously has some similarities with subjective weighted utility (Karmarkar, 1978)[21] which uses a weighting component in the utility calculation[22]. However, there is no separate probability element in Wroe et al.'s (1998) formulation, as is the case with random utility theory, as probability has been subjectively accounted for in the weighting provided by the respondents.

On the face of it Wroe et al.'s (1998) generalised utility theory appears to give support for the economic data collection and analysis method of discrete choice modelling (DCM) (and other techniques such as standard gamble and willingness to pay) which has been developed within a utility theory framework. However, there is a need to rigorously experimentally test for evidence of utility maximisation decision making (such as Wroe et al.'s modified SEU theory) amongst respondents of DCM questionnaires.

Determinants of Choice (Extension of RUT)

With the specific objective of addressing the deficiency in RUT of failing to provide a detailed description of the way individuals come to make their decisions i.e. identifying the key determinants of choice, two social cognition models were identified to complement each other and rectify the deficiency. The models were the TPB and the HBM; Rees et al. (2001) in their work considering theoretical perspectives which could be utilised to look at the potential impact of the breast cancer experience upon risk perceptions and response to information about genetic risk, identified the HBM and the TPB as the primary models of decision making and

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[21] Subjectively weighted utility = $\sum w(p_i)u(x_i)$

[22] Prospect theory (Kahneman & Tversky, 1979) has a weighting but the utility function is constructed under certainty.

health behaviour[23]. The HBM has been successfully used with women with a family history of breast cancer (Drossaert, Boer & Seyel, 1996; Kash, Holland, Halper & Miller, 1992; Norman & Brain, 2005); perceived susceptibility and severity proved to be particularly sensitive factors. Norman & Brain (2005) supplemented the HBM with behavioural control/self-efficacy and past behaviour. The TPB has been successfully used to predict hypothetical attendance for genetic testing for breast and colon cancer (Braithwaite, Sutton & Steggles, 2002) and the hypothetical decision to seek medical help when confronted with breast cancer symptoms (Hunter, Grunfeld & Ramirez, 2003) and attendance at breast cancer screening for the first time amongst eligible women (Rutter, 2000; Steadman, Rutter & Field, 2002).

Self-efficacy theory was rejected as the strongest element of the theory, perceived self-efficacy, has been incorporated into other models or they contain the highly correlated variable of perceived behavioural control. The multidimensional health locus of control was rejected as it is only appropriate as a health behaviour model for individuals who value their health and even when this problem is addressed it only explains a small proportion of the variance in respondents' decisions. Protection motivation theory was rejected as it only had a very limited number of factors from the wide range identified in the other models, and the PMT factors were included amongst those of more elaborate models such as the latest version of the HBM.

The TRA and the TPB both had the advantages of addressing the problem of social and environmental factors, which were associated in the PMT and the HBM, by including normative beliefs (Ogden, 2004). However, the TPB was selected rather than the TRA as it has the perceived behavioural control factor, a difficult factor to operationalise in relation to genetic testing, but relevant when considering a service which can only be accessed via a referral from a clinician and satisfying the eligibility criteria of the cancer genetic service. Although, it is not relevant in the

[23] In a study of genetic testing for prostate cancer conducted subsequently to the experiment outlined in this thesis Doukas, Localio & Li (2004) selected the TRA (the forerunner of the TPB) and the HBM as the most appropriate models for looking the hypothetical decision to have genetic cancer testing.

case of genetic testing as the test only needs to be taken once, perceived behavioural control can also account for the influential factor of past behaviour (Ogden, 2004). In Chapter 3 an experiment is described which was designed in light of this literature review. This experiment looks at predicting behavioural intention (hypothetical attendance) and therefore it was not considered necessary to include emotional and 'intention of action' variables that increase the variance explained in actual behaviour.

Disciplinary Interrelationships

Given that the focus of this thesis is eliciting patient preference there is a consistent overlap and interplay with other academic disciplines concerned with human choice and decision making throughout this chapter. Of all the disciplines concerned with decision making, the most prominent in its relevance to DCM and health economics in this literature review is psychology/health psychology. Psychology has been concerned with the issue of choice and subsequent behaviour since its inception; looking at motivation, choice, prediction and the most appropriate methodology to test hypotheses. As the comparatively new academic discipline of health economics turns its attention to human behaviour in the form of patient preference and choice, there is great potential to forgo conducting the research that psychologists have conducted to date. The body of psychological literature can be adopted and adapted to guide the aims and objectives of health economics.

For this thesis there were three primary overlapping areas between health economics and psychology. Firstly, the decision making process, the systematic weighing up of the benefits and barriers of choices and selecting the option that maximises the decision maker's utility (random utility theory). Secondly, the identification of social cognition and demographic predictors of choice (the health belief model and the theory of planned behaviour), an area that health economics has completely ignored to date whilst psychology and health psychology in particular has concentrated much effort in this area. Thirdly, the method of eliciting patient preferences, DCM; which

has its technical roots in mathematical psychology and its theoretical roots in decision theory.

Conclusions

With the exception of Wilson et al. (1999) economic evaluations of cancer genetic services have ignored patient preferences. There is a need to account for patients' views in health care provision, extending the range of outcome measures considered beyond health outcomes to non-health outcomes and aspects of process when they form part of patients' preferences (utility function). Given the finite resources available to service providers and planners it is necessary to ascertain preference data within an opportunity cost framework. Ideally preference data should be gathered in conjunction with accurate cost estimates allowing policy makers and service providers to tailor services to deliver maximum utility (benefit) to patients within budgetary constraints. Wilson et al. have provided the first step in providing this information. However, Wilson et al.'s study does have some limitations. Firstly, they have only looked at four of the potentially significant aspects of cancer genetic services. Secondly, the attributes and sample of respondents were specific to the Scottish model of delivering cancer genetic services, with one counselling session and no genetic testing. In light of these findings there is a need for further research designed to examine patient preferences for the manner in which cancer genetic services are delivered using the discrete choice modelling technique (DCM). In Chapters 4 to 7 of this thesis the results of a study designed to looking at patient preferences for the manner in which cancer genetic services are delivered using DCM is presented.

DCM has been applied to numerous health care issues and shown considerable potential as a preference elicitation technique to inform health policy and service provision. Its popularity stems from the advantages it offers over alternative methods. DCM incorporates realistic service attributes and levels (health, non-health, process [including charges/WTP]), continuous or nominal/qualitative in

nature and presents them in a straightforward choice format which is easy to understand. The choice format also allows opportunity cost to be incorporated into the decision process. The analytical results provide information on whether or not attributes and levels are significant to individuals, provides information upon the strength of preference for attribute levels and complete health care scenarios, and the relative importance of attribute levels and complete scenarios. The technique can also be utilised with revealed preference information, when the information is available, and not merely stated preference information.

Wroe et al.'s (1998) generalised 'modified utility theory' appears to give support for the economic data collection and analysis method of discrete choice modelling (DCM) which has been developed within a random utility theory framework. However, there is a need to rigorously experimentally test for evidence of utility maximisation decision making amongst respondents of DCM questionnaires. In Chapters 4 to 7 the results of an experiment designed to address this very issue are presented.

If respondents of DCM questionnaires adhere to RUT this will identify the decision making process, but it does not provide a detailed description of the way individuals come to make their decisions as RUT does not identify the key determinants of choice. To extend the identification of the determinants of choice beyond the weighted benefits and barriers to the underlying cognitions, it is proposed that theory of planned behaviour in conjunction with the health belief model be utilised.

In the future, having addressed the issue of whether or not respondents of DCM exercises adhere to its underlying decision theory principals, health economics should address the dearth of evidence upon the reliability and validity of the technique in the field of health care. For example, no research has been conducted into the reliability of DCM in relation to scenario (stimulus) set, data collection procedure, criterion validity in the form of predicting uptake in the field of health

and social care or temporal reliability for DCM instruments presenting respondents with multiple choices (3 or more) and indifference options.

CHAPTER 3: BACKGROUND AND METHODS OF THE EMPIRICAL STUDIES.

The NHS funded Cancer Genetics Service in Wales (CGSW) offers a resource intensive model of care, offering high risk patients multiple counselling sessions and genetic testing. Given that there is no information upon the preferences of patients for the manner in which the Welsh model of providing cancer genetic services is delivered, or data on the associated costs, and Chapters 1 and 2 established that there is a paucity of research upon patient preferences for the manner in which cancer genetic services are delivered, research question 1 was developed. Research question 1, "What are the attributes of cancer genetics services that are important to high risk patients (the patients spending the most time in contact with the service and receiving most services i.e. genetic testing and counselling)? and what would be the cost of providing the service to comply with patient preferences?"

Having examined the literature on eliciting patient preferences the relatively new and experimental technique of discrete choice modelling (DCM) was identified as the most appropriate one to use to elicit the data required to answer research question 1. However, in spite of the growing body of literature addressing various aspects of DCM's reliability and validity (Bryan et al., 2000; Farrar & Ryan 1999; Ryan et al., 1998b), no one has experimentally examined respondents of a DCM exercise to see if they are adhering to DCM's underlying decision theory principals of random utility theory (Thurston, 1927; Marshak, 1960; Luce, 1959; Lancaster, 1966; McFadden, 1972, 1974). Given the lack of research on random utility theory in the discipline of health economics, the decision theory/psychology literature was accessed. The large body of literature on utility theory revealed that the descriptive ability of utility theory was in question. These findings resulted in research question 2, "Do respondents of DCM questionnaires make choices in accordance with Random Utility Theory?" Following on from research question two an additional question was raised (research question 2.1). As utility theory does not provide a detailed description of the way individuals come to make their decisions (Feather, 1982; Edwards, 1992; Jones, 1993, Frisch & Clemen, 1994; Conner & Norman, 1995), confining

itself to benefits and barriers (the pros and cons) of a decision, components of two social cognition models, the theory of planned behaviour (TPB) (Ajzen, 1985, 1988, 1991) and the health belief model (HBM) (Hochbaum, 1958; Rosenstock, 1966, 1974; Becker et al., 1977) were selected to identify the determinants of choice. Research question 2.1, "In the event that respondents are found to adhere to RUT, can the determinants of choice be extended beyond 'benefits and barriers' (RUT) using relevant components of the theory of planned behaviour and health belief model?"

In the remaining chapters of the thesis two studies are reported which seek to answer research questions 1 and 2. Firstly, an experiment testing the implicit assumption made when using discrete choice modelling that respondents are using random utility theory to make their decisions (research question 2). Secondly, a survey of patients referred for cancer genetic services to establish their preferences for services delivery and the costs of prioritising services in terms of patient preferences (research question 1).

Background

Information Manipulation Experiment

In essence, random utility theory like subjective expected utility and generalised utility theory proposes that the decision maker aims to maximise their utility, where utility is the sum of the weighted subjective utilities of the components of the risky choices or uncertain prospects where the weights are subjective probabilities associated with each component.

Expected utility and its siblings, subjective expected utility (SEU) and generalised utility in their various forms have been found to be invalid as descriptive models (Miyamoto, 1992; Keller, 1992; Camerer, 1992; Edwards, 1992) (a discussion of the limitations of these theories was given in Chapter 2). Sarin (1992; Eppel et al., 1992) and Schoemaker (1982) suggested that many of the violations of EU, SEU and generalised utility theories when experimentally tested were the result of psychological attributes that were not accounted for in the experiments and analysis.

The health psychologists Wroe et al. (1998) propose that the failure of SEU and generalised utility models as descriptive models under experimental conditions stemmed from their application in too narrow a fashion; strictly imposing the experimenters' definition of choices that correspond to utility maximisation. Wroe and Salkovskis (1999) emphasise the influence of subjective beliefs, proposing that the individuals' decisions are the result of a logical decision process but based upon their beliefs, transitive factors such as anxiety and the information that is available and deemed to be relevant at the time, no matter how irrational or factually incorrect it may be (Wroe and Salkovskis, 1999). By applying a cognitive framework to collecting data upon the benefits and barriers of health behaviours Wroe and Salkovskis overcame the problem of applying utility theory in too narrow a fashion. In layman's terms, rather than imposing the researcher's definition of utility maximisation and rationality upon all respondents, they merely asked what the benefits and disadvantages of health behaviours were to respondents and how important these factors were to them.

In a series of studies Wroe et al. (1998) successfully predicted intention to have genetic testing and attendance at screening (Salkovskis et al., 1999; Wroe & Salkovskis, 1999, 2000; Wroe et al., 1998, 2000). Evidence supporting the use of utility theory within a cognitive framework in relation to the decision to pursue the hypothetical possibility of obtaining genetic testing for a range of diseases including colon cancer and breast cancer was found in a descriptive study with a sample of students and a sample of the general public that had considered genetic testing (Wroe et al., 1998). Wroe and Salkovskis (1999) proceeded to examine how benefits and barriers influenced the decision to be tested, by examining under experimental conditions the impact of information and a focussing manipulation upon the hypothetical decision to have genetic testing for breast cancer and heart disease. The results supported utility theory as a descriptive model with both the information and the focusing manipulation significantly affecting the self-prediction of opting for testing in accordance with the theory. Positive information led to greater self-prediction of testing whilst control information made no change and negative information (positive information followed by negative information) led to a decrease in the self-prediction of having testing.

Cameron and Diefenbach (2001) reported contradictory results to those of Wroe and Salkovskis (1999). In their study, where all respondents were provided with basic information about genetic cancer (standard information), increasing the amount of information provided relating to the consequences of testing, regardless of its nature (positive, negative or both), was associated with lower intention to have testing. They propose that information about the consequences of testing, regardless of its nature, may dampen initially high intention levels. This could be a result of the information leading to more careful consideration of the potential implications of testing.

Both Wroe and Salkovskis, (1999) and Cameron and Diefenbach (2001) provided some of their participants with positive and negative information. However, only Cameron and Diefenbach counterbalanced the presentation of their positive and negative information to prevent bias stemming from an ordering effect. Ordering or primacy effects have been found in information recall studies using medical statements as information (Ley & Spelman, 1967; Ley, 1972; Ley, 1982). It is possible that an ordering effect influenced the benefits and barriers considered by participants receiving positive followed by negative information in the Wroe and Salkovskis (1999) study and influenced their decision to utilise genetic testing services.

In the experiment described in this thesis an information manipulation based upon the work of Wroe and Salkovskis (1999) was conducted. If, in contrast to the findings of Cameron and Diefenbach (2001), individuals considering accessing cancer genetic services make their decision and establish utility/preference in accordance with random utility theory this will be a valuable contribution to validating DCM as a health economics and health services research technique. However, it does not give a detailed description of the way individuals come to make their choices (Feather, 1982; Edwards, 1992; Jones, 1993, Frisch & Clemen, 1994; Conner & Norman, 1995). To address this deficiency and highlight some of the determinants of choice the psychology/health psychology literature was accessed once more.

To extend the identification of the determinants of choice beyond the weighted benefits and barriers used in RUT social cognition models that assume that decisions are made in accordance with utility theory (Edwards, 1954a; Conner & Norman, 1995) were examined. The models assessed were those identified by Weinstein (1993), van der Plight (1994) and Conner and Norman (1995) as being rooted in utility theory[1]; the health belief model (HBM), health locus of control (HLC), protection motivation theory (PMT), theory of reasoned action (TRA) / theory of planned behaviour (TPB) and self-efficacy theory (SET). The Health Belief Model (HBM) (Hochbaum, 1958; Rosenstock, 1966, 1974; Becker et al., 1977) and the Theory of Planned Behaviour (TPB) (Ajzen, 1985, 1988, 1991) were selected as the most appropriate; a review of the social cognition models and the reasons for selecting the TPB and HBM are presented in Chapter 2. In their work considering theoretical perspectives which could be utilised to look at the potential impact of the breast cancer experience upon risk perceptions and response to information about genetic risk, Rees et al. (2001) identified the HBM and the TPB as the primary models of decision making and health behaviour[2].

In addition to utility theory, the TPB and the HBM, many other factors have been associated with the self-prediction of having testing: anxiety (Wroe & Salkovskis, 1999; Cameron & Diefenbach, 2001), cancer worry (Cameron & Diefenbach, 2001), having previously considered testing (Wroe et al., 1998), a family history of cancer (Cameron & Diefenbach, 2001), age at which a close relative was diagnosed with cancer (Brain, Gray, Norman, Parsons, et al. 2000) and demographic variables (part of HBM and TPB) such as age (Salkovskis et al., 1999; Brain, Gray, Norman, Parsons, et al., 2000). See the measures section for a full list of the measures included in the experiment.

The proposed research will therefore bring together contemporary work in health economics and health psychology: firstly, by experimentally testing the implicit

[1] Sometimes when social cognition models are discussed in the literature the term expectancy value is used interchangeably with utility theory. In that context they can be treated as equivalent as they are referring to the subjective utility and probability of an outcome.

[2] In a study of genetic testing for prostate cancer conducted subsequently to the experiment outlined in this thesis Doukas, Localio & Li (2004) selected the TRA (the forerunner of the TPB) and the HBM as the most appropriate models for looking the hypothetical decision to have genetic cancer testing.

assumption made in discrete choice modelling that respondents are using random utility theory to make their decisions; and secondly, by incorporating the theory of planned behaviour and the health belief model to extend the determinants of choice beyond random utility theory's benefits and barriers. See Figure 1.3 (Chapter 1) for a graphical representation of the interrelationship between economic and psychology/health psychology theory.

Survey of Patient Preferences in the Delivery of Cancer Genetic Services

The systematic review of the economic evidence on cancer genetic services in Chapter two revealed that economic evaluations to date have concentrated on technology assessment, looking at mutation identification and health outcomes. There is a lack of research looking at what aspects of cancer genetic services patients value (importance of service attributes) and how they would like services to be delivered. The only published exception is the work of Wilson et al. (1999)[3].

Wilson et al. (1999) conducted an economic evaluation looking at patient preferences for the delivery of a single genetic counselling session. They found evidence supporting the use of genetic associates and nurses rather than doctors/consultants to counsel in appropriate circumstances. Genetic associates and nurses are less expensive than doctors/consultants and were found to be acceptable to patients, particularly if this resulted in improvements in other attributes.

There is no universally agreed protocol on how genetic services should be provided and as a result the findings of Wilson and et al. (1999) cannot be generalised to all NHS funded regional and national cancer genetic centres in the UK. The only other national cancer genetics service in the UK, other than the one in Scotland, is the one provided in Wales by the 'Cancer Genetics Service in Wales' (CGSW). The service offered in Wales is substantially more resource intensive than the one offered in Scotland. The Welsh protocol provides risk

[3] O'Neill conducted his/her Msc research in this field but it could not be obtained from the University of Aberdeen by inter library loan.

assessment to all patients referred to them, genetic counselling to moderate risk patients, and in the case of high risk patients two counselling sessions and genetic testing for a cancer affected relative (to initially identify the genetic mutation) and four counselling sessions, genetic testing and the arrangement of presymptomatic surveillance for the presymptomatic patient(s)[4] initially referred to the service. Clearly there is a need for a preference based economic evaluation of the Cancer Genetics service in Wales' resource intensive testing and counselling protocol for high risk patients.

Methods

In this and the remaining chapters study specific methods, results and conclusions will be identified in the relevant headings and text. The information manipulation experiment will be referred to as the experiment and the survey of patients referred for cancer genetics services will be referred to as the survey.

Experiment Methods

Research question 2, "Do respondents of DCM questionnaires make choices in accordance with Random Utility Theory?" was operationalised into the empirical aim below.

Aims, Objectives and Hypotheses

Empirical Aim

Experimentally examine respondents of a DCM exercise by means of an information manipulation to see if they are adhering to DCM's underlying decision theory principals of random utility theory.

The empirical aim above involves theory testing, which comprises of six steps; 1) specify the theory to be tested, 2) devise a set of conceptual propositions, 3) restate the conceptual propositions as testable propositions, 4) collect data, 5) analyse the data and 6) assess the theory (de Vaus, 1996). In the 'aims,

[4] Patient that is currently free of cancer but suspected of having a genetic mutation placing them at high risk of developing cancer.

objectives and hypotheses' section of this chapter, steps one to three will be considered.

1) Specify the theory to be tested

In this instance the concept to be tested is the primary axiom of random utility theory (and all other variants of utility theory) when assessing respondent's choices; utility maximisation.

2) Devise a set of conceptual propositions

Random utility theory states that individuals are utility maximisers and weigh up the benefits and barriers of a choice and chooses the option that maximises their subjective expected utility (provides maximum benefit). Therefore, we would expect positive information about genetic testing to result in an increase in the benefits (pros) of testing and counselling reported by respondents and an increase in the behavioural intention to have genetic testing and counselling. Naturally we would expect negative information to have the contrary effect and result in respondents reporting an increase in the limitations of testing and counselling (the cons) and be less likely to want to have genetic testing and counselling. If respondents are utility maximisers we would expect accurate measures of the benefits in relation to the barriers/disadvantages (ratio of pros to cons) to be positively associated with the behavioural intention to have genetic testing and counselling. That is the more benefits relative to barriers the more likely a respondent is to want to have genetic testing and counselling.

3) Restate the conceptual propositions as testable propositions

These testable propositions outlined above have been operationalised into objectives and hypotheses below. In addition, objectives two, three, five and six have been included. Objective two is designed to accounts for a potential information ordering effect. Objective three extends objective one which looks at the pros and cons of testing and the behavioural intention to have testing, and examines the possibility that information may also affect preferences for service attributes. Objective five assesses if the information manipulation has produced any change in respondents' knowledge and health perceptions/social cognitions.

Objective six addresses research question 2.1[5] by utilising relevant components of the TPB and HBM to identify social cognition determinants of choice and not merely benefits and barriers. Objective six is distinct from objective five as the TPB and HBM are continuum models and specific relationships between variables are assumed.

Objectives

1. Ascertain if the experimental information manipulation will result in participants evaluating and making choices in relation to genetic testing and counselling for breast cancer in accordance with random utility theory, specifically in relation to: the relative importance of the perceived benefits (pros) and barriers (cons) of genetic services and the behavioural intention to have testing and counselling (see hypotheses 1, 2, 4 and 5).
2. Ascertain if there is an information ordering/primacy effect (see hypotheses 3 and 6).
3. Ascertain if the experimental information manipulation will change respondents' service attribute preferences.
4. Compare the strength of the relationship between the 'relative importance of the perceived benefits to barriers' to the behavioural intention to have testing and counselling pre and post information manipulation (see hypotheses 7).
5. Ascertain the changes, if any have taken place, in the knowledge and health perceptions/social cognitions of respondents following the information manipulation.

If respondents are adhering to RUT.

6. Ascertain which components of the TPB and HBM are significant predictors of behavioural intention.

Hypotheses

The hypotheses and a lay explanation are presented in Table 3.1. A graphic representation of the hypotheses is presented in Figure 3.1.

[5] Research question 2.1, "In the event that respondents are found to adhere to RUT, can the determinants of choice be extended beyond 'benefits and barriers' (RUT) using relevant components of the theory of planned behaviour and health belief model?"

Table 3.1

Hypotheses and Lay Explanation

Testable hypotheses	Lay explanation
1. Experiment information will produce a statistically significant change in the weighted ratio of pros to cons of testing and counselling.	
2. Positive information will increase the pros relative to the cons of genetic testing and counselling recoded by respondents, both the pos-neg (Wroe & Salkovskis, 1999, called this group the negative information group) and the neg-pos information will increase recorded cons relative to the pros, and little or no change will occur for the control group.	If individuals are adhering to utility maximization: Positive information should result in an increase in the importance of the pros of testing and counselling. Information on the limitations of testing and counselling should result in an increase in the importance of the cons. Control information should not cause change in the weighted ratio of pros to cons.
3. Neg-pos information will result in a statistically significantly greater decline in the ratio of pros to cons between assessments (baseline to follow-up) than the pos-neg information (ordering/primacy effect).	An ordering effect is assumed. The information issued first is assumed to influence the weighted ratio of pros to cons e.g. giving negative followed by positive information will result in cons having more importance than when positive information is given followed by negative information.
4. Experiment information produces statistically significant change in self-prediction and intention to have testing and counselling.	
5. Positive information will increase self-prediction and intention scores, both pos-neg and neg-pos information will reduce self-prediction and intention scores, and little or no change in scores will occur for the control group.	If individuals are adhering to utility maximization: Positive information should result in an increase in the desirability of testing and counselling. Information on the limitations of testing and counselling should result in a decrease in the desirability. Control information should not cause change in the desirability of testing and counselling.
6. Behavioural intention (intention and self-prediction) of booking an appointment for genetic testing and counselling for breast cancer will decline between assessments statistically significantly more for the neg-pos than the pos-neg information group (ordering/primacy effect).	An ordering effect is assumed. The information issued first is assumed to influence the desirability of testing and counselling e.g. giving negative followed by positive information will result in testing and counselling being less desirable than when positive information is given followed by negative information.
7. A large degree of positive correlation (0.5 – 1.0) will exist between weighted ratio scores and self-prediction and intention to have testing scores.	Individuals weigh up the pros and cons of a choice and choose the option that provides them with the most perceived benefit or minimises undesirable consequences.

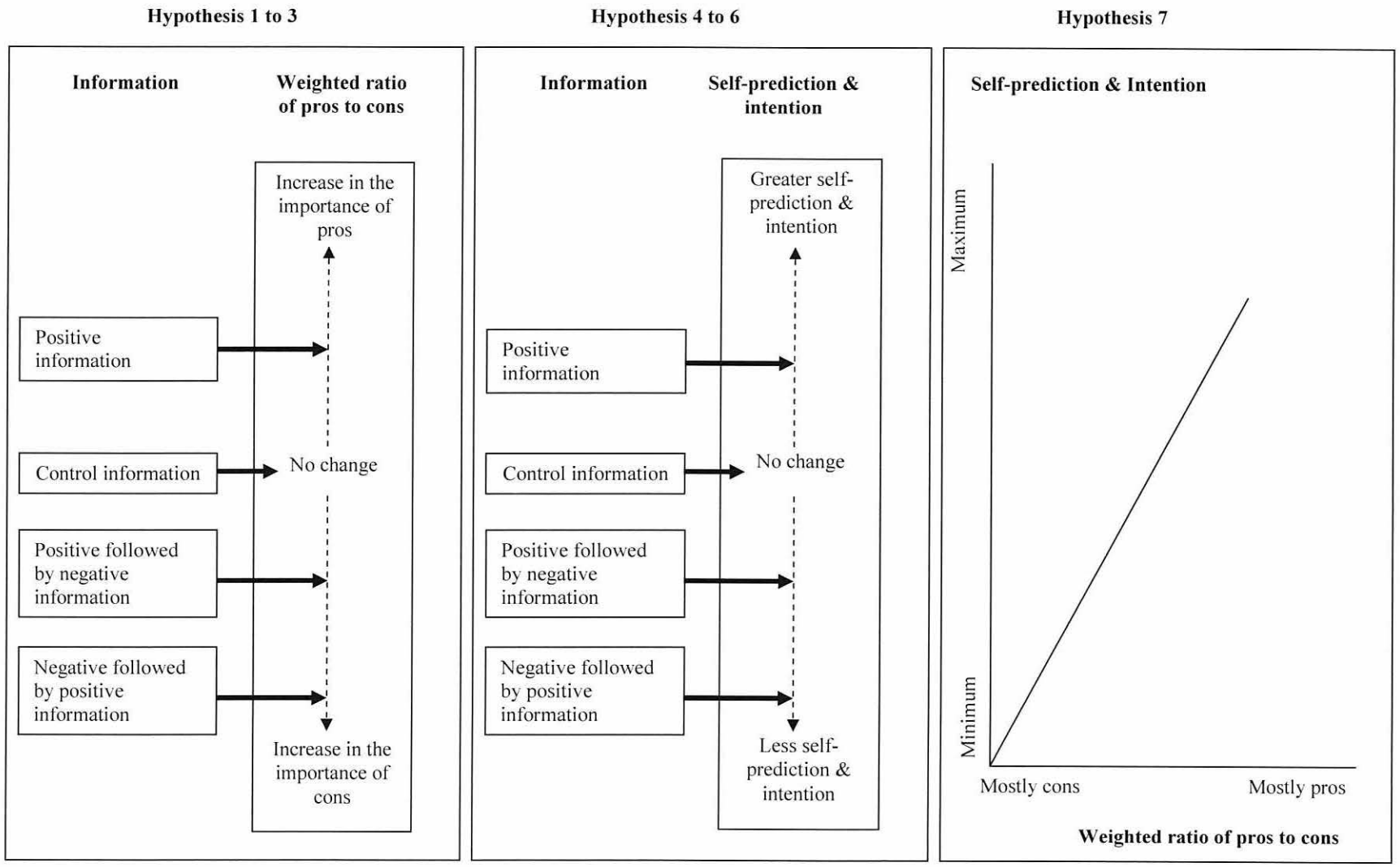


Figure 3.1. Graphic representation of hypotheses.

No hypotheses were made in relating to the responses to the DCM questionnaire. There are three main reasons for this. Firstly, as there is currently no inferential statistical test to assess if significant change has taken place between assessments, no related hypothesis could be tested. Secondly, no hypotheses were formulated in relation to the type of change that might take place in preferences following the information manipulation experiment, as the discrete choice questionnaire only has three discrete response categories and as a result is likely to be insensitive to small differences between groups. For example, differences such as those sought in hypotheses 3 and 6. Thirdly, as the sum of attributes' B coefficients (discrete choice regression coefficients) sum to zero when effects codes are used, comparison of estimated utility using a summary measure such as the mean is impractical as it will equal zero (or in the case of some levels being non-significant approximately zero).

Design

A repeated measure experimental design was used. Participants were randomised into positive, negative and control groups. The positive group were given information on detection and prevention of genetic breast cancer, the negative group received the same information in addition to information on the limitations of these methods, and the control group were given information relating to the common cold. Unlike the single negative group used by Wroe and Salkovskis (1999), two negative groups were used in this study, one received the positive information first and the other the negative information first (counterbalancing). This allowed the results to be examined for any potential ordering (or primacy/recency) effects.

Data was gathered using self-administered questionnaires (see Appendix G). Measures were taken at baseline and following the administration of the information manipulation. Conducting the pre and post information measurement within one sitting and with all the information groups at the same time in separate rooms alleviated the potentially biasing influences of discussion of the issues with others, and media coverage of relevant issues between assessments. Media coverage was a particular problem as October 2002 was breast awareness

month; local and national newspapers and national magazines such as The Daily Post, The Welsh Daily Mirror, the associated newspapers group (Femail.co.uk, Daily Mail, Mail on Sunday and Metro Newspapers) and Cosmopolitan magazine were publishing articles, supplements with information upon and references to further information on genetic breast cancer. However, there is the potential problem of recall bias, respondents recalling their original responses and answering the second set of questions in light of this.

Setting

The experiment was conducted in five research laboratories at the University of Wales, Bangor. Five rooms were used as four rooms capable of holding in excess of 55 students each were not available at a suitable time. As a result two neutral groups had to be run. The entire experiment was completed in one hour and forty minutes.

Participants

The sample consisted of 158 male and female first year undergraduate students of the School of Psychology, University of Wales, Bangor (traditional and mature). These students had not yet begun to study health psychology modules. Having gained ethical approval, all first year undergraduate Psychology students were informed that the study related to genetic testing for cancer, were given some background information and invited to participate. This was done by means of an oral presentation accompanied by handouts, made to all students attending a compulsory first year lecture. The presentation was made in the first week of the autumn term of 2002 to approximately 231 students. Copies of the script of the oral invitation and the handouts provided are presented in Appendices H and I. Participants were awarded course credits and print credits for participating in the experiment.

In order to ensure that participants' baseline experience, knowledge or psychological status did not confound the results of the experiment the following series of screening questions were posed. Participants were asked if they:

- Had ever had cancer.

- Had any of their family had cancer.
- (Clinical risk of genetic cancer estimated from reported family history)
- Had a genetic disorder.
- Had had genetic testing or counselling.
- Had considered going for genetic testing.
- Knew anyone who has a genetic disorder.
- Knew anyone who has had counselling or testing.
- Ever read genetic testing literature.
- Discussed genetic testing or counselling with any one.
- How knowledgeable they were about cancer.
- How knowledgeable they were about breast cancer.
- How knowledgeable they were about non-cancer genetic testing.
- How knowledgeable they were about genetic testing for cancer.
- How knowledgeable they were about genetic testing for breast cancer.

To assess current mood Participants were also asked to complete the Hospital Anxiety and Depression Scale (HADS) (Zigmond & Snaith, 1983).

Positive answers were compared to negative (negative and don't know in the case of a family history of cancer), knowledge scores of 50 or above were compared to those below 50 (0 = Know nothing -100 = Very Knowledgeable) and clinical scores on the HADS (11+ on a 0 – 21 point score; Carroll et al., 1993) were compared with scores of 10 or less on; their weighted ratio, self-prediction and intention scores. The respondents providing a positive response to the experience items, a rating of 50+ on the knowledge items and a score of 11+ on the HADS scales were excluded from the study if they provided substantially different responses to their counterparts.

Measures

Discrete Choice Modelling Questionnaire

Respondents' utility, both students in the experiment and patients in the survey, was measured using a discrete choice modelling questionnaire. There are four main stages in designing a typical discrete choice modelling study (see Chapter

2); establishing the attributes, assigning levels to the attributes, selecting scenarios to present and establishing preferences (present the discrete choices).

Attribute identification.

The attributes to be tested in the discrete choice model were identified from a review of the literature on patient preferences for health care, discrete choice modelling of patient preferences for health service provision, current research on attendance at cancer genetics clinics, guidance papers on the delivery of genetics services and consultation with the CGSW's Senior Consultant Cancer Geneticist, Dr. Jonathan Gray. The initial results of the search for possible attributes are presented in Table 3.2.

From the list of possible attributes in Table 3.2 six were selected to be tested in the discrete choice exercise; five process attributes (staff seen for counselling, waiting time from referral to receipt of a letter confirming risk status, duration of counselling appointments, availability of genetic testing, cost of service) and one non-health attribute (distance to appointment). The attributes 'staff seen', 'waiting time', 'distance to counselling' and 'duration of counselling' had been found to be significant to past attendees of genetic counselling (Wilson et al.,1999) and similar attributes had been successful in other health care settings (see Table 3.2). Availability of testing was included as an attribute as in the fields of genetic testing for Huntington's disease (Harper & Clarke, 1997) and breast cancer (Brain, Gray, Norman, Parsons, et al., 2000) a very strong desire to obtain testing amongst certain families was found. However, the evidence clearly shows that testing should only be available to high risk families (Griffith et al., 2004). This attribute will ascertain if there is a strong desire for testing, even among patients that would not benefit from it (the analysis of this variable for low and moderate risk patients is presented elsewhere). The attribute 'cost of service' was included to allow maximum willingness to pay within attribute levels to be estimated.

Table 3.2

Possible Attributes Identified

Attributes	Sources
Health outcomes	
Years with condition/without condition/develop condition-symptoms	Verhoef et al. (1991), Hakim and Pathak (1999)
Pain	Ryan and Hughes (1997)
Complications	Ryan and Hughes (1997), Singh et al. (1998), Jan et al. (2000)
Impact on health (amount of effect)	Singh et al. (1998), Bryan et al. (2000), Ratcliffe (2000a)
Probability of health outcomes	Singh et al. (1998), Ryan (19991, 1999b), Bryan et al. (2000)
Non-health outcomes	
Technical or skills	
Technical competence/ Good treatment/ Quality of care	Freidson (1961), Kane (1969), Parker and Srinivasan (1976)
Good equipment & facilities	Kane (1969)
Staff manner	
Taking an interest in the patient/ Good treatment/ Friendly staff/ Personal manner, personal interest, personal qualities, attitudes, or friendliness of the physician and staff towards the patient, can talk to clinician, clinician listens	Freidson (1961), Kane (1969), Parker and Srinivasan (1976), Vick and Scott (1998), Ryan (1999a, 1999b)
Image	
Physician image	Parker and Srinivasan (1976)
Referral systems (selection on the grounds of clinical or public recommendation)	Parker and Srinivasan (1976)
Patient characteristics	
For example, child's preference, age, clinical background	Singh et al. (1998), Ratcliffe (2000a)
Convenience	
Distance to appointment/ travel/travel time/travel cost	Wilson et al. (1999), HERU (undated a, b), Jan et. al. (2000), Expert Advisory Group on Cancer to the Chief Medical Officer of England and Wales (Calman-Hine) (1995)
Availability or accessibility or convenience of the care	Kane (1969), Parker and Srinivasan (1976)
Process	
Favourable admission policies/ Entry barriers	Kane (1969), Parker and Srinivasan (1976)
Staff seen at the appointment	Wilson et al. (1999), Ryan, McIntosh and Shackley (1998a), Royal College of Physicians (1991), Genetics Research Advisory Group (GRAG) (1995), ACOGT (1998a) Harper and Clarke (1997) Steel et al. (1999)
Waiting time till appointment/on list	Wilson et al. (1999); Ryan (1999a, 1999b); HERU (undated a, b); Ryan et al. (1998a), Jan et. al. (2000), Ratcliffe (2000a)
Duration of appointment	Wilson et al. (1999), Vick and Scott (1998), Semper (1995), Harper and Clarke (1997)
Time in hospital/recovery time	Ryan and Hughes (1997), Bryan et al. (2000)
Type of treatment	Singh et al. (1998)
Information	Vick and Scott (1998), Advisory Committee on Genetics Testing (ACOGT (1998a,1998b), Expert Advisory Group on Cancer to the Chief Medical Officer of

	England and Wales (1995)
Choice of treatment/input into decision	Vick and Scott (1998)
Continuity of staff	Ryan (19991, 1999b)
Follow-up support	Ryan (19991, 1999b)
Availability of clinical testing	Brain, Gray, Norman, Parsons, et al. (2000), Harper and Clarke (1997)
Provider agency	Harper and Clarke (1997), GRAG (1995), ACOGT (1998a, 1998b, 1999), J. Gray (personal communication, March 18, 2000)
Financial	
Price-charge for care	Parker and Srinivasan (1976), Ryan and Hughes (1997), Singh et al. (1998), Ryan (1999a, 1999b), Bryan et al. (2000), Jan et al. (2000)

Attribute levels.

Table 3.3

Attributes and Levels Selected

Attributes	Levels
Staff seen for counselling	Specialist genetics nurse, Consultant geneticist , Genetics associate
Waiting time for letter	1 months, 2 months, 4 months, 6 months
Distance to counselling	20 miles, 40 miles, 60 miles, 80 miles
Duration of counselling	30 minutes, 1 hour, 1 hour 30 minutes, 2 hour
Availability of genetics testing	Testing for high risk only, Testing for all.
Cost of service	£1,500, £2,000, £2,500, £3,000

The levels must be realistic and trade-offs between them possible (Farrar et. al., 1997; Parker & Srinivasan, 1976, Ryan, 1996a). All the selected levels are presented in Table 3.3. For the attribute 'staff seen', three levels were selected; the two clinical counselling categories currently in use by the CGSW, consultant geneticists and specialist genetics nurses, and a third level of genetics associates. Although genetics associates are not currently used in Wales, they have been included as a level as they were found to be acceptable to patients as counsellors (Wilson et al., 1999) and are a potential change to counselling provision within the Welsh service.

The waiting time levels selected were 1 month, 2 months, 4 months and 6 months. The levels were based on current waiting times in the three centres. The average length of time between initial referral and confirming risk status was believed to be approximately 2 months (J. Gray, personal communication, March 18, 2000).

As there are only three Cancer Genetics Centres in Wales patients are required to travel relatively large distances. The distances selected were realistic at 20, 40, 60 and 80 miles.

The levels selected for the attribute 'duration of counselling' were 30 minutes, 1 hour, 1 hour 30 minutes and 2 hours. They were selected to be representative of actual service and sufficiently different to encourage respondents to consider trading between attributes. Currently, the average length of a counselling session for individuals at high risk of developing cancer is one hour (J. Gray, personal communication, March 18, 2000).

As patients at low and moderate risk of developing familial cancer are both currently denied cancer genetics testing, and the number of levels influence the number of scenarios (see Chapter 2), the three risk categories were not used as levels for the attribute 'Availability of testing'. The levels 'High risk' and 'All' were utilised. 'High risk' represents current provision, testing available only to patients at high risk due to their family history. The level 'All' was selected to represent the availability of testing for respondents at high moderate and low risk of developing familial cancer.

The levels for the attribute 'cost of service' are based on the commercial price of genetics testing. Myriad Genetics, inc. and their subsidiary Myriad Genetics Laboratories, Inc. have made mutation testing commercially available since 1996 (Reynolds, 2000); charging $2,400 (£1,548.39 @ £1=$1.55) for BRCA analysis and $3,500 (£2,258.06 @ £1=$1.55) for rapid BRCA analysis (Myriad, 1999)[6]. The interval of £500 between levels was selected as it was found to be sufficiently large to ensure that respondents would consider trading this attribute against others in pilot testing.

[6] These prices exclude genetic counselling, although myriad do advice individuals to seek counselling and will help individuals locate genetic counsellors (Myriad, 2001)

Selection of scenarios.

If a full factorial design was employed the attributes and levels listed in Table 3.3 would produce 1,536 scenarios[7]. Clearly a discrete choice instrument of this size cannot be presented to respondents. Verhoef et al. (1991) in their study presented breast cancer patients with forty eight paired comparisons and noted problems with fatigue, lack of care and attention to detail and inconsistency, particularly in the latter stages of questionnaire completion. As has been noted in Chapter 3, there are five main approaches used, separately or in conjunction, to reduce the number of scenarios to be presented (Pearmain et al., 1991; Ryan et. al., 1996). Removing dominant and dominated options, blocking and carrying out a series of DCM studies with the same respondents were rejected as there would be too many scenarios and too few respondents. Defining attributes and levels in terms of differences between alternatives was rejected as there was no specific scenario e.g. planned service change, with which to base the comparator for change, and the number of scenarios would still be too large.

The six attributes and their levels were entered into the Speed 2.1 software (Bradley, 1991), which produced a fractional factorial design with 25 scenarios (combination of attributes and levels). The 25 scenarios are presented in Table 3.4. Examination of the scenarios revealed that only 24 scenarios needed to be used in the discrete choice modelling exercise, as scenario A and scenario U are identical. No correlation between levels of different attributes should be found in a fractional factorial design (Bradley, 1991); the speed output concurred with this requirement.

[7] $4^4*2^1*3^1=1,536$.

Table 3.4

Scenarios

	Staff seen for counselling	Waiting time for a letter	Distance to counselling	Duration of counselling	Availability of testing	Cost of service
A	Consultant geneticist	4 Months	80 Miles	120 Minutes	All	£2,500
B	Specialist genetics nurse	4 Months	60 Miles	30 Minutes	All	£3,000
C	Genetics associate	4 Months	40 Miles	90 Minutes	High	£2,500
D	Consultant geneticist	4 Months	20 Miles	60 Minutes	High	£1,500
E	Specialist genetics nurse	4 Months	80 Miles	120 Minutes	High	£2,000
F	Genetics associate	2 Months	80 Miles	30 Minutes	High	£1,500
G	Consultant geneticist	2 Months	60 Miles	90 Minutes	All	£2,000
H	Specialist genetics nurse	2 Months	40 Miles	60 Minutes	All	£2,500
I	Consultant geneticist	2 Months	20 Miles	120 Minutes	High	£3,000
J	Specialist genetics nurse	2 Months	80 Miles	120 Minutes	High	£2,500
K	Specialist genetics nurse	1 Months	80 Miles	90 Minutes	High	£3,000
L	Consultant geneticist	1 Months	60 Miles	60 Minutes	High	£2,500
M	Specialist genetics nurse	1 Months	40 Miles	120 Minutes	All	£1,500
N	Associate	1 Months	20 Miles	120 Minutes	All	£2,000
O	Consultant geneticist	1 Months	80 Miles	30 Minutes	High	£2,500
P	Specialist genetics nurse	6 Months	80 Miles	60 Minutes	High	£2,000
Q	Genetics associate	6 Months	60 Miles	120 Minutes	High	£2,500
R	Consultant geneticist	6 Months	40 Miles	120 Minutes	High	£3,000
S	Specialist genetics nurse	6 Months	20 Miles	30 Minutes	All	£2,500
T	Consultant geneticist	6 Months	80 Miles	90 Minutes	All	£1,500
U	Consultant geneticist	4 Months	80 Miles	120 Minutes	All	£2,500
V	Specialist genetics nurse	4 Months	60 Miles	120 Minutes	High	£1,500
W	Consultant geneticist	4 Months	40 Miles	30 Minutes	High	£2,000
X	Specialist genetics nurse	4 Months	20 Miles	90 Minutes	High	£2,500
Y	Genetics associate	4 Months	80 Miles	60 Minutes	All	£3,000

Ascertaining respondents' preferences.

Preferences were elicited by means of a self-administered questionnaire. Patients were asked to choose between pairs of scenarios, with the added option of stating indifference between options. All scenarios were paired against one scenario. The scenario selected was scenario H in Table 3.4. This scenario was representative of current service at the CGSW. To obtain the transitivity, dominance and non-satiation data required, scenarios M versus B and D versus W were included (they were not included in the DCM regression). To avoid 'response set bias'; that is respondents providing the same response to all questions regardless of content, it is advisable to vary response format (Bowling,

1997; de Vaus, 1996). To prevent such bias affecting the results of this study scenario H was randomly allocated to scenario option A or option B in each of the 23 choices in which it was used. The random number generation facility of Microsoft Excel was used to generate 23 number between 0 and 1, figures below 0.5 resulted in scenario H appearing in discrete choice option A, and in option B if the figure was 0.5 or above.

Prior to answering the discrete choice questions respondents were presented with a definition of the attributes and levels, asked to rank their preference amongst nominal attribute levels and rank the attributes as a whole (see Appendices G and J). The format and presentation of the discrete choice questionnaire was based on the format utilised by Farrar et al. (1997), Ryan (1999b), Jan et al. (2000), Vick and Scott (1998) and Pearmain et al. (1991) and especially Wilson et al. (1999).

Psychosocial Measures.

A number of demographic and psychological issues have been found to be significantly related to decision making in relation to cancer genetic services e.g. age, anxiety and cancer worry (Brain, Gray, Norman, Parsons, et al., 2000; Cameron & Diefenbach, 2001; Salkovskis et al., 1999; Wroe et al., 1998). Based upon these findings and the literature relating to implementing behavioural models in health care research a battery of instruments has been developed; including the New Socio-economic Classification (Rose & O'Reilly, 1998), Hospital Anxiety and Depression Scale (HADS) (Zigmond & Snaith, 1983) Cancer worry (Lerman, 2000) and the Life Orientation Test (LOT= dispositional optimism test, Scheier & Carver, 1987). The instruments used are listed in Table 3.5.

Ethical approval was gained from the School of Psychology (U.W.B) for all the instruments used in this experiment. The TPB and HBM items used were designed by the GenQuest research team and are used with their permission.

Table 3.5

Measures

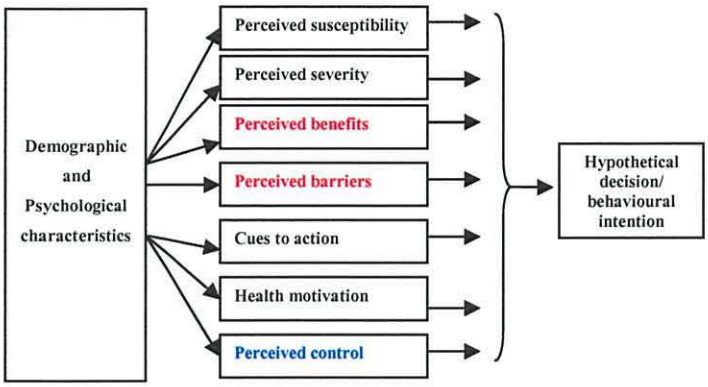
Measure	Number of items
Demographic characteristics, including the New Socio-economic Classification (Rose & O'Reilly, 1998)	4 open ended items (job description). 11 closed format items.
Previous experience of Psychology and economics	2 dichotomous items
Knowledge and experience of genetics and oncology	7 dichotomous items, 4 open ended items, 1 closed format and 5 likert items (11 point)
Family history of cancer	10 dichotomous items, potential 1 – 18 one word responses and ages
Dispositional optimism, LOT (Scheier & Carver, 1987)	12 likert items (5 point) with 4 dummy items
Anxiety	
HADS (Zigmond & Snaith, 1983)	7 likert items (4 point)
Anxiety, 3 new items.	3 likert items (11 point)
Depression, HADS (Zigmond & Snaith, 1983)	7 likert items (4 point)
Cancer worry, 2 items by Cameron and Diefenbach (2001)	2 likert (7 point)
Discrete Choice Modelling questionnaire	7 likert items (11 point), 5 ranking items, 25 discrete choices (3 response options), 1 dichotomous.
Random Utility Theory & Health Belief Model	
Benefits and disadvantages of genetic testing for breast cancer (Adapted from Wroe et al., 1998)	As many open ended responses as respondent note.
The importance of the benefits and disadvantages of genetic testing for breast cancer	One likert items (11 point) for each benefit and barrier noted.
Weighted ratio of pros to cons (Σimportance of benefits – importance of barriers/ importance of benefits + importance of barriers) (Wroe et al., 1998)	
Health Belief Model	
Perceived susceptibility	3 likert items (7 point)
Perceived severity, 1 new item and 2 by Wroe and Salkovskis (1999)	3 likert items (11 point)
Theory of Planned Behaviour	
Attitudes towards behaviour	
Belief about outcome	2 likert items (7 point)
Evaluation of outcome	1 likert items (7 point)
Subjective norm	
Normative belief	6 likert items (7 point)
Motivation to comply	3 likert items (7 point)
Perceived control	2 likert items (7 point)
Perceived risk, 1 new item and 2 by Cameron and Diefenbach (2001)	3 likert (7 point)
Coping with a positive test result	1 likert items (11 point)
Behavioural intention, 2 by Cameron and Diefenbach (2001) & 1 adapted from Wroe et al. (1998)	3 likert items (11 point)

Unless otherwise stated all multiple item measures were transformed into a single score by summing the ratings on each of the individual component items/questions. For example HADS depression was calculated by summing the results on each of the seven depression items giving a score between 0 and 21.

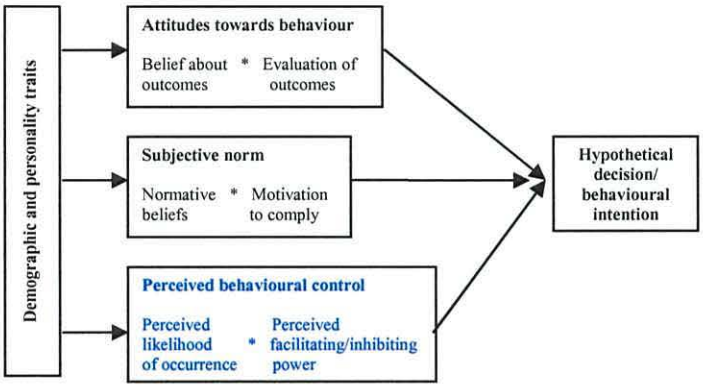
Mapping of psychological constructs to the questions administered to research participants.

Figure 3.2 displays the three models utilised in the experiment and applied to the hypothetical decision to have cancer genetic testing and counselling. As the experiment is only looking at a hypothetical decision (behavioural intention), behaviour as specified in the TPB is not relevant and is omitted in this instance (see Figures 1.2 and 3.2). Factors/psychological constructs that are common to more than one model are highlighted in colour in the respective models.

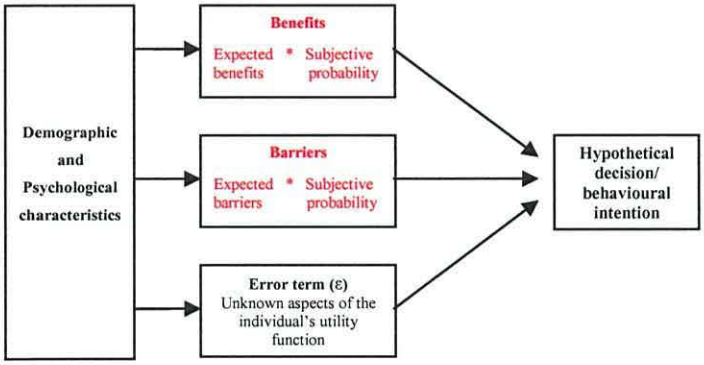
The questions used in the experiment to elicit information upon the psychological constructs of the TPB, HBM and RUT are presented in Table 3.6. All questions were worded in accordance with examples of good practice for the respective models (Ajzen, 2002; Conner & Norman, 1995; Francis, Eccles, Johnston, Walker, Grimshaw, Foy, Kaner, Smith, and Bonetti, 2004; Wroe et al. 1998).



Health Belief Model



Theory of Planned Behaviour Model



Random Utility Theory

Note: Factors common to more than one of the above models are highlighted in colour in each of the models.

Figure 3.2. HBM, TPB and RUT applied to a hypothetical decision (behavioural intention).

Table 3.6

Psychological constructs and the questions representing the constructs

Theory and/or construct	Items and (response range)
TPB	
Perceived behavioural control	**How much control do you believe you have over the following:**
	Having a genetic test carried out (1=Little /no control to 7=Complete control)
	Getting information about cancer that runs in families (1=Little /no control to 7=Complete control)
Subjective norm (Normative belief * Motivation to comply)	
Normative belief	**How much would your family like you to do the following as a way of becoming aware of cancer in its early stages?**
	a) Regular screening procedures (e.g. mammography) (1=Not at all to 7=Very much)
	b) Having a genetic test carried out (1=Not at all to 7=Very much)
	c) Getting information about cancer that runs in families (1=Not at all to 7=Very much)
	How much would your GP like you to do the following as a way of becoming aware of cancer in its early stages?
	d) Regular screening procedures (e.g. mammography) (1=Not at all to 7=Very much)
	e) Having a genetic test carried out (1=Not at all to 7=Very much)
	f) Getting information about cancer that runs in families (1=Not at all to 7=Very much)
Motivation to comply	**How much do you want to do any of the following as a way of becoming aware of cancer in its early stages?**
	g) Regular screening procedures (e.g. mammography) (1=Not at all to 7=Very much)
	h) Having a genetic test carried out (1=Not at all to 7=Very much)
	j) Getting information about cancer that runs in families (1=Not at all to 7=Very much)
Attitudes towards behaviour (Belief about outcomes *	

Evaluation of outcomes)	
Belief about outcomes	**To what extent do you agree with the following statements?**
	k) If I have genetic testing (1=It will tell me nothing to 7=I will better know my future)
	l) Genetic testing would be (1= Harmful to 7=Beneficial)
Evaluation of outcomes	**To what extent do you agree with the following statements?**
	m) How valuable would it be to know your genetic risk (1=Not valuable to 7=Extremely valuable)
HBM	
Perceived Susceptibility	How likely do you think you are of having a gene giving you an increased risk of getting breast cancer? (1=Not at all to 7=Almost certain or extremely) (Cameron & Diefenbach, 2001)
	How likely do you think it is that, at some point in your life, you will get breast cancer? (1=Not at all to 7=Almost certain or extremely) (Wroe et al., 1998)
	How vulnerable do you think you are to getting breast cancer at some point in your life? (1=Not at all to 7=Almost certain or extremely) (Cameron & Diefenbach, 2001)
Perceived Severity	How serious an illness do you think breast cancer is? (0=Not at all serious to 100=Extremely serious)
	How bad would it be to have breast cancer? (0=Not at all bad to 100=Extremely bad) (Wroe & Salkovskis, 1999)
	How bad would it be to find that you have an increased susceptibility of developing breast cancer? (0= Not at all bad to 100=Extremely bad) (Wroe & Salkovskis, 1999)
Perceived Benefits and Perceived barriers / Weighted ratio of pros to cons (**RUT**)	
Perceived benefits (pros)	Please state in the left hand column headed **"Benefits"**, all the reasons you can think of **in favour** of genetic testing and counselling for breast cancer. Do not worry about the order in which you state the reasons. (Open ended response) (Adapted from Wroe et al., 1998)

	If genetic testing and counselling for breast cancer was available to you (free of charge) by booking an appointment, how relevant would the **"Benefits"** be to your decision to book or decline genetic testing and counselling? Please, rate the relevance of each of the **"Benefits"** to **you** by circling a number from 0 to 100 in the right hand column headed **"Relevance"**.
Perceived barriers (cons)	Please state in the left hand column headed **"Disadvantages"**, all the reasons you can think of **against** genetic testing and counselling for breast cancer. Do not worry about the order in which you state the reasons. (Open ended response) (Adapted from Wroe et al., 1998)
	If genetic testing and counselling for breast cancer was available to you (free of charge) by booking an appointment, how relevant would the **"Disadvantages"** be to your decision to book or decline genetic testing and counselling? Please, rate the relevance of each of the **"Disadvantages"** to **you** by circling a number from 0 to 100 in the right hand column headed **"Relevance"**. (0=Not at all relevant to 100=Extremely relevant)
Cues to action	No question on this construct
Health motivation	No question on this construct
Perceived control	No question on this construct
Cancer worry	To what extent are you worried about getting breast cancer? (0=Not at all to 7=Almost certain or extremely) (Cameron & Diefenbach, 2001)
	To what extent are you concerned about getting breast cancer? (0= Not at all to 7=Almost certain or extremely) (Cameron & Diefenbach, 2001)
Coping	How well do you think you would cope if you had genetic testing and were told that you had an 80% chance of developing breast cancer by the age of 70? (0=Extremely badly to 100=Extremely well)
Disease specific anxiety	
Anxiety – risk of developing breast cancer	How anxious do you feel about your risk of developing cancer? (0=Not at all anxious to 100=Extremely anxious)
Anxiety – having genetic testing	How anxious would you feel about having genetic testing? (0=Not at all anxious to 100=Extremely anxious)
Behavioural intention (dependent variables)	

Intention	How interested are you in getting a genetic test for breast cancer susceptibility? (0=Definitely not to 100=Definitely) (Cameron & Diefenbach, 2001)
	I plan to have genetic testing for breast cancer susceptibility when it is available? (1= Not at all to 7=Almost certain or extremely) (Cameron & Diefenbach, 2001)
Self-prediction/Expectation	If genetic testing and counselling for breast cancer was available to you (free of charge), how likely would you be to book an appointment to have this service? (0=Definitely not to 100=Definitely) (Adapted from Wroe et al., 1998)

As the experiment is looking at a hypothetical decision, 'cues to action' (HBM) such as an invitation to attend genetic testing and counselling is inappropriate and has not been used in the experiment. Health motivation was not used in this study as it was felt that the TPB's subjective norm which includes motivation to comply was a stronger measure. As perceived control (HBM) is a component of the TPB, there was no need to incorporate this construct again for the HBM. Only the four strongest predictors of health behaviour from a meta-analysis of HBM studies, barriers, susceptibility, benefits and severity (Harrison, Mullen & Green, 1992) were selected to supplement the TPB. As the primary focus of this thesis is random utility theory, data on benefits and barriers was gathered as suggested by Wroe et al. (1998)[8].

Behavioural intentions capture the motivations influencing behaviour and reflect the efforts individuals are willing to devote to performing a health behaviour (Ajzen, 1991). Intentions have been measured primarily in three ways, desires, intentions and self-predictions (or expectations) (Conner & Norman, 1995; Armitage & Conner, 2001). In this experiment a two item intention measure designed by Cameron & Diefenbach, 2001) and a single item self-prediction measure (based upon a question developed by Wroe et al., 1998) were used. Intentions and self-predictions have been found to be better predictors of actual

[8] Becker et al. (1977) noted that perceived benefits should be weighted against barriers in the HBM but did not specify how this should be done.

behaviour than desire (Armitage & Conner, 2001). The intentions and self-predictions questions used in this experiment (see Table 3.6) were not combined into a single behavioural intention measure as Shepperd, Hartwick and Warshaw's (1988) meta-analysis suggests that self-prediction measures may be a better predictor of behaviour than intention measures. The Intention score was calculated by transforming the results of the 0 to 100 item to a 7 point rage like its counterpart question (multiply results by 7/100) and summing the results on both questions (Cameron & Diefenbach, 2001).

The TPB measures used were designed by the GenQuest team for use with patients referred to the Cancer Genetics Service in Wales and reflect the protocols of that service. The items used in the TPB were generated in respect to previous research conducted by members of this team with patients at increased risk of developing genetic cancer, genetic surveillance and cancer screening (Bennett, P., Phelps, C., Brain, K., Hood, K. and Gray, J., 2007; Lowe, R., Vedhara, K., Bennett, P., Brookes, E., Gale, L., Munnoch, K., Schreiber-Kounine, C., Fowler, C., Rayter, Z., Sammon, A. & Farndon, J., 2003; Phelps, C., Bennett, P., Iredale, R., Anstey, S. & Gray, J., 2005).

For the TPB, perceived behavioural control was measured directly and the remaining two constructs, attitudes towards behaviour and subjective norm, were measured indirectly. Indirect measurement of perceived behavioural control (PCB) is difficult to operationalise in relation to cancer genetic services as the service is highly organised and follows set protocols driven by a patients' family history/genetic risk. Due to the highly organised nature of the service 'perceived likelihood of occurrence' is rendered inappropriate in relation to the two areas upon which questions focussed, 'having genetic testing carried out' and 'getting information about cancer'. Having genetic testing carried out is completely governed by risk status (number of family members with potential genetic cancer) and having access to a blood sample from a cancer affected relative. Information on genetic cancer is automatically issued when a patient is referred to the cancer genetics service. Given that measuring perceived likelihood of occurrence was impractical in light of the limitations noted above, indirect measurement of PCB was considered inappropriate.

Usually direct measurement of perceived behavioural control comprises of capability and controllability items. Capability questions capture the individuals' ability to perform the behaviour and controllability items capture the individuals' belief that the performance of behaviour is under their control (Ajzen, 2002). Due to the organised nature of issuing information and conducting genetic testing capability questions were not used. The only potentially relevant capability issues related to obtaining information and having genetic testing were approaching their GP to discuss genetic testing (if their GP, an oncologist, surgeon or member of the genetic service via a relative had not already contacted them) leading to referral and automatically being issued with information on genetic cancer, and attending a genetics clinic or having a home visit to give a blood sample accompanied by counselling. Given that all patients should have the capability to fulfil these tasks it was felt by the GenQuest research team that in this instance capability items could be excluded. In a study of genetic testing for prostate cancer, published subsequently to the experiment outlined in this thesis being designed and implemented, Doukas, Localio and Li (2004) opted to completely omit perceived behavioural control and opted to use the forerunner of the TPB, the theory of reasoned action.

Perceived behavioural control in the form of controllability alone was produced by summing the results of both controllability items. Indirect measurement of subjective norm and attitudes towards behaviour were calculated in the conventional way by multiplying the components factors together and summing the results e.g. attitudes towards behaviour = $\sum$belief about outcomes * evaluation of outcomes (Ajzen, 2002). The motivation to comply questions were designed to correspond to both sets of normative belief questions, reducing the number of motivation to comply questions required to three rather than six (subjective norm = questions in Table 3.6 [a*g]+[b*h]+[c*j]+[d*g]+[e*h]+[f*j]). One 'evaluations of outcomes' question was used as it was designed to be equally applicable to both beliefs about

outcomes questions (behavioural intention = questions in Table 3.6 [k*m]+[l*m])[9][10].

As Rosenstock (1966) did not specify how the variables of the HBM interact and combine, calculating factor scores and their application as predictive variables is a matter of debate (Abraham, Sheeran, Abraham & Spears, 1996; Strecher & Rosenstock, 1997; Ogden, 2004). Perceived susceptibility and perceived severity were treated as independent factors.

Benefits and barriers have been combined into a weighted ratio as suggested by Wroe et al. (1998) for utility theory. Weighted ratio of pros to cons (Wroe et al., 1998) was calculated by subtracting the weighted barriers from the benefits and dividing by the sum of the weighted benefits and barriers.

$$\text{Weighted ratio} = \frac{\sum \text{Benefit Importance} - \sum \text{Barrier Importance}}{\sum \text{Benefit Importance} + \sum \text{Barrier Importance}}$$

For example, if a respondent had 3 benefits (pros) for genetic testing with respective importance to them of 80, 80 and 90, and 3 disadvantages (cons) with respective importance weightings of 70, 80, 90 the weighted ratio score would be 0.02 [(80+80+90)-(70+80+90)/(80+80+90+70+80+90)].

Family history of cancer.

The anonymous research participants were categorised as having a family history of breast or breast ovarian cancer if their family history complied with the Cancer Genetics Service in Wales's referral guidelines (Griffith, Edwards, Gray, Butler, Wilkinson, Turner, France & Bennett, 2005). See Table 3.7 below for family history referral guidelines.

[9] Ajzen (2002) does not specify a minimum number of questions to be used to measure a factor, but he does emphasise that only items with good internal consistency should be used.
[10] Unipolar coding was used for all TPB items rather than bipolar (Ajzen, 2002) e.g. 1 to 7 (see Table 3.6).

Table 3.7

Cancer Genetics Service in Wales's Referral Guidelines

Cancer	**Family history criteria for referral** (on the same side of the family)
Breast Cancer	• 1 first degree relative diagnosed at 40 years or less. • 2 first degree relative diagnosed at 60 years or less. • 3 first or second degree relative diagnosed at any age. • 1 first degree male breast cancer. • A first degree relative with bilateral breast cancer.
Breast/Ovarian Cancer	• At least one breast and one ovarian cancer in first degree relatives (breast cancer diagnosed under 50 if only one of each cancer). • A first degree relative who has both breast and ovarian cancer.

Source: Griffith et al. (2005).

Tests of dominance and transitivity.

To reiterate briefly from Chapter 2, random utility theory dictates that preferences must be transitive[11] and continuous[12]. Violation of the axiom of continuous preference was tested for by looking for dominant preference patterns. Dominance was identified using the same method as proposed by Scott, (2002). The 23 paired choices presented to respondents were checked and respondents were deemed to have a dominant preference for an attribute if they always selected the scenario with the most favourable value of a specific attribute in each paired choice and had ranked that attribute as the most important of all six attributes.

Four paired choices were included in the questionnaire where one of the two scenarios was dominant. Three of the four choices in conjunction, choices 3, 13 and 25, formed a test of transitivity. The choices comprise of scenarios H, M and B (Table 3.4) where M > H > B and as a result respondents would be expected to select 'option B' in all three choices. The remaining test was purely a test of non-satiation, where one scenario was perceived by the author to be superior to the other as it was equal or better on each attribute.

All un-validated items were piloted with convenience samples of students and staff members of the Institute of Medical and Social Care Research for acceptability, clarity and ease of use.

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[11] *If A > B and B > C, then A > C.*

[12] *If utility is continuous compensatory decision making (trade-offs) takes place, individuals are prepared to accept more of a specific attribute in compensation for less of another.*

Procedures

Students who wished to participate in the study were asked to register that they were going to participate by signing up for the study. 173 students signed up for the study. To ensure random assignment to the experimental and control groups 173 numbers (1 to 173) were printed and shuffled repeatedly. These numbers were placed in a bag, selected at random and handed to participants as they entered the lecture theatre where the experiment was initially convened. Participants were then allocated to information groups according to the random numbers they had been issued with (see Table 3.8).

Table 3.8

Randomised Groups

Random numbers issued to participants	Information given	Number of participants
1 to 43	Positive	42
44 to 86	Positive then negative	39
87 to 129	Negative then positive	36
130 to 152	Neutral	23
153 to 173	Neutral	18
Total number of participants		158

An experiment protocol was produced (Appendix K) and the following procedures were followed by each of the group coordinators with their respective groups. Students were asked to switch off their mobile phones, not to talk to each other until the experiment was completed and to remain seated until every one in their group had handed in their questionnaires. They were also informed that they would receive further information at the end of the study and their printer credits.

The coordinators showed participants page 31 of the questionnaire which read "Please Stop here! You will be given some information shortly. Once you have read the information the researcher coordinating your group will ask you to turn to the next page and answer the remaining questions. Thank you." Participants were told verbally that they should stop answering questions when they got to that point in the questionnaire and that they would be asked to continue when it was appropriate.

The questionnaires were then handed out. Participants were asked to read and sign the informed consent form and hand them back to the coordinator before answering the anonymous questionnaire (see Appendix L). Coordinators then observed the class ensuring there was no talking, no visual communication and assisting anyone who had a problem or question. Once everyone had completed the first half of the questionnaire the relevant information sheet(s) were handed out face down. The information provided to participants was the information used by Wroe and Salkovskis (1999) (see Appendix M). In the case of the negative groups, the information sheets were stapled in the order in which the participants were supposed to read them. Once everyone had an information sheet(s), the overhead projector (OHP) with a copy of the information on acetate was switched on, participants were then asked to turn their information sheets face up and the coordinator read the contents of the information sheet(s) aloud to the class. This ensured that any visual or hearing impairment on the part of the participants did not affect the experiment. The OHP was then switched off and participants were asked to put their information sheets to one side. This ensured that all participants were exposed to the information simultaneously and for the same length of time. Participants were then told to forget about their previous answers and not to look back at them; they could then answer the remainder of the questionnaire. The coordinators then collected all the information sheets.

Once all the questionnaires had been completed and everyone had ticked their names on the attendance list to allow course credits to be allocated to them, debriefing sheets (see Appendix N) and printer credits were handed out.

Sample Size and Power

158 undergraduate students participated in this experiment (68.4% response rate). Preliminary power analysis during the design of the experiment indicated that a sample of 100 respondents or more would be adequate to find moderate effect sizes with α's of 0.05 or less at 80% power for the statistical hypotheses (hypotheses 1 and 4) specified. The final sample size having excluded those respondents failing to pass the screening questions was 142. The following post hoc power analysis has been conducted on the least favourable parameters

(maximum number of independent variables and smallest N) to ascertain the minimum sensitivity (largest effect size) of the experiment at 80% power.

The smallest recorded N for a multivariate test was 107 and the largest number of independent variables (IV's) was 25. With α set at 0.05 and power at 80% a moderate effect size of R^2=0.21 to 0.23/f^2=0.26 to 0.30[13] could be detected in either multiple linear regression or GLM analysis (Cohen, 1988).

The smallest N and number of discrete choice observations was recorded by the Neg-pos group at N=31 and observations=712. With α set at 0.05 and a confidence interval of 10% choice probabilities (the probability of making a certain choice) as low as 0.36 (range 0 to 1) can be detected by discrete choice regression. For the entire sample (pos, pos-neg, neg-pos, control = 3254 observations) choice probabilities as low as 0.11 can be detected (Louviere et al., 2000).

Coding

The attribute levels of the discrete choice scenarios presented to respondents (independent variables) were effects coded. See Table 3.9 for the effects codes used. Attribute levels of the constant comparator scenario in the DCM questionnaire were taken as the baseline and were all coded with -1s. Attribute levels for the no preference option were all coded as 0. The merits of effects coding relative to more popular or conventional coding methods will be discussed in the discussion section. All other variables were coded using conventional coding methods e.g. dummy or ordinal coding.

[13] The lower estimate had 24 IV's specified and the upper estimate 30 IV's specified in the calculations.

Table 3.9

Discrete Choice Attribute Levels and their Effects Codes

Variable/Attribute	Levels			
		Code 1	Code 2	Code 3
Staff seen	Consultant geneticist	1	0	-
	Specialist genetics nurse (Baseline)	-1	-1	-
	Genetics associate	0	1	-
Waiting time for letter	1 month	1	0	0
	2 month (Baseline)	-1	-1	-1
	4 month	0	1	0
	6 month	0	0	1
Distance to counselling	20 miles	1	0	0
	40 miles (Baseline)	-1	-1	-1
	60 miles	0	1	0
	80 miles	0	0	1
Duration of counselling	30 minutes	1	0	0
	1 hour (Baseline)	-1	-1	-1
	1 hour 30 minutes	0	1	0
	2 hour	0	0	1
Availability of testing	High risk	1	-	-
	All (Baseline)	-1	-	-
Cost of service	£1,500	1	0	0
	£2,000	0	1	0
	£2,500 (Baseline)	-1	-1	-1
	£3,000	0	0	1

Note: The dependent variable, scenario chosen (option A, option B or no preference) was coded as 1 for the selected scenario and 0 for the rejected scenarios.

Analysis

In data screening (removing respondents that fail the screening questions) and establishing continuity of information groups, tests for independent samples were used (Chi-square, independent t test, Wilcoxon-Mann-Whitney U test, Median test, One-way analysis of variance test or the Kruskal-Wallis one-way ANOVA by ranks test). Reliability and validity of the psychosocial measures used in this experiment were assessed using Cronbach's alpha and correlation (Pearson or Spearman). The strength of the relationship between the weighted ratio of benefits to dis-benefits (weighted by importance) with the behavioural intention to opt for testing was assessed by correlation (Pearson or Spearman). Correlation coefficients were interpreted in accordance with Cohen's classification (1988), coefficients of 0.1 to 0.29 are small, 0.30 to 0.49 are medium and 0.50 to 1.00 are large. The number of reasons for, against and the importance weightings were compared pre to post information by means of pair-wise tests (related t test, Wilcoxon signed ranks test or the sign test). To establish if there was a difference

in the benefits and barriers and the desirability of opting for testing by information group and other potentially significant independent variables repeated measure General Linear Models (GLM) were conducted. Preferences for the manner in which cancer genetic services are provided (elicited from discrete choices) were established by means of discrete choice regression (multinomial logit model, heteroscedastic extreme value model, random parameters mixed logit model or the multinomial probit model). In the absence of an established inferential statistical test to determine pair-wise change in preferences (DCM) for attribute levels over time, substantial change was defined as change in attribute level coefficients as a ratio of baseline standard error (approximate Z score) which exceeded ±1.96 (the critical value of two tailed z statistic at α=0.05). Any change in social cognition variables from pre to post information was assessed using repeated measure GLM analysis (analysis of variance/covariance). Establishing the strongest predictors of self-prediction and intention to have genetic testing was done by means of multiple linear regressions.

In the case of multivariate GLM, repeated measures GLM with multiple independent variables and multiple linear regression analysis the final models presented in the results section are those with a parsimonious specification of the independent variables. Only variables that were statistically significant ($P \leq 0.05$) or approached statistical significance ($P \leq 0.1$) were included in the final specification of GLM, linear regression and discrete choice models. In the case of univariate and multivariate GLM analysis, descriptive statistics are presented in the form of estimated marginal means (EMM) and not observed means. Estimated marginal means are adjusted for the covariates, if there are any. In the absence of covariates EMM's and observed means are the same. All of the above tests and their tenets/assumptions are discussed in detail in Appendix B. Discrete choice regression was conducted on Limdep 7.0/NLogit 2.0 software; all other analysis was conducted on SPSS 13.0.

Survey Methods

Research question 1, "What are the attributes of cancer genetics services that are important to high risk patients (the patients spending the most time in contact with the service and receiving most services i.e. genetic testing and counselling)? and what would be the cost of providing the service to comply with patient preferences?" was operationalised into the empirical aim below.

Aims and Objectives

Empirical Aim

Ascertain the aspects of cancer genetics services that are important to patients, and present service configurations prioritised in terms of preferences accompanied by their costs (cost-consequences analysis) for high risk patients.

Objectives

- Establish if service attributes identified from literature are important to patients.
- Calculate maximum willingness to pay for cancer genetics service (within the attribute levels presented).
- Establish the relative utility of service configurations based upon significant service attributes.
- Estimate the costs of providing cancer genetic testing and counselling (service configurations) derived from significant service attributes.

Design

A survey of patients of the CGSW post issuing them with their clinical risk assessment. Data was gathered approximately one week after the CGSW had informed the patients (high and moderate risk) and the referrer (in the case of low risk patients) of their status. The data was gathered with the aid of the GenQuest research team (Turner, France, Wilkinson, Griffith & Edwards, 2002) who were evaluating the CGSW and issuing their own questionnaires to these patients.

Setting

Patient data was gathered by self-administered questionnaires sent to patient's homes.

Participants

The sample comprised of 120 adult (18+) male and female patients of the Cancer Genetics Service in Wales (CGSW) at high, moderate and low risk of developing genetic cancer (breast, ovarian and colorectal) recruited as part of the GenQuest research project. Patients were recruited from all three of the CGSW centres in Wales (University Hospital of Wales, Cardiff; Singleton Hospital, Swansea and Ysbyty Glan Clwyd, Bodelwyddan, North Wales). Data capture for this survey took place between May 2001 and the January 2002.

Measures

Three types of information were collected in the survey of CGSW patients, demographic, clinical and service preference. The demographic data included age, gender, qualifications, work status and Rose and O'Reilly's (1998) New Socio-economic Classification. Clinical data comprised of the cancer centre a patient was referred to, the cancer they were at risk of developing and their clinical risk of developing genetic cancer[14]. Preferences were gathered using the same DCM questionnaire as was administered in the information manipulation experiment to students. The research project and all the measures used were awarded ethical approval by the national (MREC) and all the regional (LREC) ethics committees.

Procedures

All patients referred to the CGSW between February 2001 and January 2002 were invited to participate in the GenQuest study. Having reviewed a referral letter the CGSW issue patients with information upon genetic cancer and a family history questionnaire by mail. The GenQuest information sheet and informed consent form were issued to patients in a sealed envelope along with the CGSW information (see Appendix J). Upon the sealed envelope with the

[14] Data on age, gender, cancer centre, suspected cancer type and risk status was collected by the Cancer Genetics Service in Wales.

GenQuest paperwork was an invitation to participate in the research. Patients returning a signed informed consent sheet and a questionnaire were enrolled in the study.

The survey questionnaire was issued to patients in the mail by the GenQuest research team (see Appendix J). Questionnaires were issued approximately one week after the CGSW had informed the patients (high and moderate risk) and the referrer in the case of low risk patients.

Sample size & power

220 DCM questionnaires were issued by the GenQuest team, 120 patients returned a discrete choice questionnaire giving a response rate of 54.5%. Of the 120 returned questionnaires 5 did not complete the discrete choice items. For the entire sample (N=115, observations = 2633) choice probabilities (the probability of making a certain choice) as low as 0.13 (range 0 to 1) can be detected by discrete choice regression with α set at 0.05 and a confidence interval of 10% (Louviere et al., 2000). For the high risk breast cancer patients used in the cost-consequences analysis (N=30, Number of observations=678) choice probabilities of 0.37 and above can be detected.

Coding and analysis were conducted in the same fashion as for the experiment.

Costs

Resource use data for the micro costing was gathered by means of an interviewer administered questionnaire administered to all clinical, administrative and laboratory staff, and conducting an audit of the clinic rooms and laboratory of the Cardiff clinic of the CGSW (see Appendix O). Resource use (labour, capital, overhead and consumables) and the unit costs of the resources were estimated in 2002/2003 £ sterling from the provider's perspective, the UK National Health Service. Costs were estimated on a per patient basis for all clinical event pathways, full details of the methods and costs are presented in (Griffith et al., 2005). Unit costs for staff time, capital, overheads, equipment and consumables were provided by the finance department of the Cardiff and Vale NHS trust.

Equipment costs were transformed into an annual cost assuming a 6% discount rate, a 5 year working life and payment in arrears.

In order to conduct cost-consequences analysis upon the alternative ways of providing cancer genetic services to high risk patients, cost estimates were required for service attributes and service configurations that do not currently exist. As a result certain assumptions must be made to facilitate the production of cost estimates. The assumptions applied in this study are outlined below.

All costs relate to high risk breast cancer patient, the first member of a nuclear family approaching the CGSW. It is assumed that upon testing a cancer affected relative that a BRCA1 or BRCA2 mutation is found and the presymptomatic patient referred to the service is then tested at a cost of £2,510 (Griffith et al., 2005).

For the attribute 'staff seen', the cost of seeing a consultant geneticist was calculated using the unit costs for an MC21/02 grade and genetics associates were costed at the unit costs for an NP51/05 grade.

The current service can provide a waiting time of 4 to 6 months (current mean wait = 3.075 months, N = 40 high risk patients). As the discrepancy between a 1 and 2 month waiting time will depend mainly on the patient returning the family history questionnaire promptly and prompt issuing of medical records for cancer affected relatives, the cost of adapting the service to facilitate a 1 and 2 month waiting time was assumed to be equal. For the waiting time for a letter to be reduced to 1 to 2 month it would be necessary to reduce the time lag between receipts of paperwork e.g. referral letter, family history questionnaire (pedigree) and medical records, and the administrative and clinical staff reviewing this paperwork and establishing clinical risk. To do this more staff would be required. To account for the increased staff input in the cost per presymptomatic patient (including cancer affected relative costs), labour and the accompanying capital and overhead costs were inflated by 50%.

The largest statistically significant level of the attribute 'distance to counselling' was taken as the baseline cost. A reduction in distance to counselling was assumed to require the clinician to travel by car at a cost of £0.45 per mile for the reduced distance to and from counselling, plus travel time for the clinician (time estimated on travelling by car at 30 miles per hour; time=distance/speed) and the cost of securing a counselling room at a district general hospital or health centre. It was assumed that the counselling room at the CGSW centre would be unused and the associated capital charges would be incurred by the CGSW and as a result included in the estimated costs. The costs associated with the duration of counselling were estimated by multiplying the presymptomatic counselling unit costs by the duration time.

CHAPTER 4: RESULTS – RELIABILITY, VALIDITY AND SAMPLE CHARACTERISTICS

Information Manipulation Experiment

Exclusions

In order to ensure that participants' knowledge and experience of cancer and genetically inherited disease, and respondents' psychological wellbeing did not confound the results of the experiment a series of screening questions were asked. (Descriptive statistics for the initial 158 respondents that attended the experiment are listed in Appendix P).

Table 4.1

Experience

	Yes	No	Don't know
Had had cancer	1 (0.6%)	157 (99.4%)	-
Any of family had cancer	101 (63.9)	40 (25.3%)	17 (10.8%)
Clinical risk of genetic cancer	2 (1.3%)	156 (98.7%)	-
Have a genetic disorder	1 (0.7%)	145 (99.3%)	-
Had genetic testing or counselling	3 (1.9%)	155 (98.1%)	-
Have considered going for genetic testing	11 (7.1%)	143 (92.9%)	-
Know anyone who has a genetic disorder	50 (31.6%)	108 (68.4%)	-
Know anyone who has had counselling or testing	39 (24.8%)	118 (75.2%)	-
Ever read genetic testing literature	34 (21.5%)	124 (78.5%)	-
Discussed genetic testing or counselling with any one	39 (24.7%)	119 (75.3%)	-

Respondents with and without experience of each of the 10 items listed in Table 4.1 were compared upon the weighted ratio of pros to cons they reported for genetic testing and counselling for breast cancer and their self-prediction and intention to obtain genetic testing and counselling. As the number with experience of 'having had cancer', 'with a clinically significant family history', 'having a genetic disorder' and 'having genetic testing or counselling' were so small, comparison using inferential statistics was not possible.

The participant that had suffered with cancer had a substantially lower weighted ratio score and was far *less likely* to have genetic testing than the remainder of the participants on the self-prediction of having testing and counselling item and

the genetic testing intention score. The two patients at increased risk of developing cancer due to their family history and the participant with a genetic disorder were far *more likely* to have testing and gave higher weighted ratio scores than the remainder of the participants. Participants that did not have a genetic disorder but had received counselling and/or testing were as likely to have testing as the remainder of the participants and provided similar weighted ratio scores. Descriptive statistics are presented in Table 4.2 The individual that had suffered with cancer did not suffer from breast cancer and neither the individual that had a genetic disorder or the three that had received counselling and/or testing had a cancer mutation or were tested for a cancer mutation. No other details are provided on these respondents in order to maintain their anonymity.

The remaining variables presented in Table 4.1 were assessed using statistical tests for independent samples. The only item upon which a statistically significant difference was found was 'have you ever considered going for genetic testing?' Median tests revealed that respondents that had considered going for testing reported significantly higher positive weighted ratio scores (N=142, Yate's continuity corrected: chi-square=6.307, df=1, p=0.012), were more likely to have testing on the self-prediction of having testing and counselling item (N=154, Yate's continuity corrected: chi-square=5.729, df=1, p=0.017) and the genetic testing intention score (N=153, Yate's continuity corrected: chi-square=7.609, df=1, p=0.006)[1] than respondents who had not considered going for testing. Descriptive statistics are presented in Table 4.2. Six of the 11 that had considered obtaining genetic testing had done so for genetic breast cancer.

[1] A median test was used due to the small number that had considered going for genetic testing (N=11), the skewed distribution of scores on the dependent variables could not be transformed satisfactorily and the distributions were not the same shape for the independent variable categories.

Table 4.2

Genetic Testing Weighted ratio, Self-prediction and Intention by Experience

		N	Mean	Median	Standard deviation	Response range
Weighted ratio of pros to cons of testing and counselling						
Had cancer	Yes	1	0.2500	0.2500	-	-1 to +1 (min=-0.65, max= 1.0)
	No	144	0.5471	0.5858	0.36932	
At clinical risk	Yes	2	0.7249	0.7249	0.13195	
	No	143	0.5425	0.5714	0. 37066	
Have a genetic disorder	Yes	1	1.0000	1.0000	-	
	No	133	0.5436	0.5439	0. 35699	
Had genetic testing or counselling	Yes	2	0.4167	0.4167	0.58926	
	No	143	0.5468	0.5833	0.36783	
Have considered going for genetic testing*	Yes	11	0.8167	1.0000	0.24331	
	No	131	0.5254	0.5273	0.36935	
Self-prediction of having testing						
Had cancer	Yes	1	10	-	-	0 = Definitely not – 100 = Definitely
	No	157	57.07	60	29.19	
At clinical risk	Yes	2	85	85	21.21	
	No	156	56.41	60	29.30	
Have a genetic disorder	Yes	1	90	-	-	
	No	145	56.48	60	28.74	
Had genetic testing or counselling	Yes	3	58.33	47.5	22.55	
	No	155	56.74	60	29.51	
Have considered going for genetic testing*	Yes	11	78.18	90	30.27	
	No	143	55.14	60	29.00	
Genetic testing intention scores						
Had cancer	Yes	1	1.7	-	-	1 = Not at all/definitely not – 14 = Almost certainly/Definitely
	No	156	7.31	7.5	3.36	
At clinical risk	Yes	2	10.8	10.8	0.28	
	No	155	7.31	7.5	3.38	
Have a genetic disorder	Yes	1	10.9	-	-	
	No	145	7.31	7.5	3.36	
Had genetic testing or counselling	Yes	3	7.58	5.73	4.06	
	No	155	7.35	7.5	3.38	
Have considered going for genetic testing**	Yes	11	10.19	10.60	3.52	
	No	142	7.13	7.35	3.30	

Statistically significant * $p<0.05$, ** $p<0.01$.

Table 4.3

Knowledge

	Mean	Median	Standard deviation	N	Answering range
Knowledge of cancer	45.9	50	21.8	158	0 = Know nothing -100 = Very Knowledgeable
Knowledge of breast cancer	43.4	40	23.0	158	
Knowledge of non-cancer genetic testing	18.9	10	19.5	158	
Knowledge of genetic testing for cancer	19.9	15	19.8	158	
Knowledge of genetic testing for breast cancer	21.8	20	21.4	158	

Descriptive statistics for knowledge scores are presented in Table 4.3. Knowledge scores were split into scores below 50 and scores of 50 and above. No statistically significant differences were found between high ($\geq$ 50) and low (< 50) scorers on any of the knowledge items upon the weighted ratio of pros to cons, the self-prediction item or the genetic testing intention score. In addition baseline scores on the self-prediction of having testing and counselling and intention to have genetic testing scores were compared for respondents with and without clinical ratings of anxiety or depression (Clinical rating = 11+) on the HADS (see Appendix P for HADS and other demographic characteristics). No significant difference emerged on anxiety scores. However, the one respondent with a clinically significant depression score was far more pro testing and counselling than the remainder of the respondents, providing a rating of 70 on the self-prediction of having testing and counselling item, 8.9 on the genetic testing intention score and a weighted ratio score of 0.8889, compared to the mean ratings of 56.69 (std. dev. 29.41), 7.34 (std. dev. 3.39) and 0.5426 for the remainder of the respondents.

In light of the findings on the baseline weighted ratio of pros to cons score and two behavioural intention (self-prediction and intention) by the screening questions on experience, knowledge and psychological wellbeing, it was necessary to remove 16 respondents from the initial sample of 158 before analysing the experiment results. The participants that were excluded were: one that had had cancer, two with a family history of cancer, one with a genetic

disorder, 11 who had considered going for genetic testing and one with a clinically depressed rating on the HADS scale.

Reliability and Validity of Psychosocial Measures

Reliability

Internal consistency

Tables 4.4 and 4.5 contain the Cronbach alpha internal consistency coefficients for the composite measures used in the experiment. It has been proposed that alphas coefficients should not fall below 0.7 (Guilford, 1956; Kline, 1993; Nunnally, 1978), and for the majority of items in this experiment this criteria has been met. In the final sample (16 respondents having been excluded) (see Table 4.5) HADS depression does have a coefficient just below 0.7 at 0.6877. Perceived control has a coefficient of 0.5968 at baseline but falls within acceptable limits at 0.7233 in the follow-up assessment. Finally the weighted ratio score has a low coefficient at baseline (0.4588) and follow-up (0.4468). As the weighted ratio scores comprise of the completely opposing variables of the sum of the weighted pros and the sum of the weighted cons of genetic testing and counselling, the low alpha coefficients are to be expected. Based upon these results the internal consistency of the measures used in this experiment are acceptable.

Table 4.4

Cronbach Alpha's for Composite Measures Prior to Exclusions

Item	Baseline		Follow-up	
	N	Standardised item α	N	Standardised item α
Anxiety (HADS)	155	0.7976	-	-
Depression (HADS)	157	0.7160	-	-
Dispositional optimism (LOT)	153	0.8752	-	-
Weighted ratio	145	0.4776	148	0.3956
Intention	157	0.8885	158	0.9139

Table 4.5

Cronbach Alpha's for Composite Measures Following Exclusions

Item	Baseline		Follow-up	
	N	Standardised item α	N	Standardised item α
Anxiety (HADS)	139	0.7941	-	-
Depression (HADS)	141	0.6877	-	-
Dispositional optimism (LOT)	137	0.8717	-	-
Weighted ratio	129	0.4588	131	0.4468
Intention	141	0.8831	142	0.9142
Perceived control (TPB)	141	0.5968	139	0.7233
Subjective norm (TPB)	137	0.8710	140	0.8936
Attitudes towards behaviour (TPB)	140	0.7351	141	0.8498
Susceptibility (HBM)	141	0.9050	142	0.9290
Severity (HBM)	142	0.7912	142	0.8256
Cancer worry	138	0.9379	141	0.9203

Inter rater reliability

In Chapter 5 the perceived benefits and barriers of having cancer genetic testing as stated by respondents are post-coded into categories and reported. Post-coding into categories was done by the author. As a test of the quality of coding of similar open ended responses into categories, 50 questionnaires were randomly selected by computer (SPSS software) and divided between three undergraduate students (17, 17 and 16 questionnaires respectively) for coding of the first three benefits and barriers recorded by respondents into the categories identified from the data. The degree of agreement between the undergraduates' coding and the coding used in this thesis was tested using the Kappa test (Cohen 1960). Bakeman and Gottman (1986) view Kappa coefficients below 0.7 with concern whist Fleiss (1971) considers coefficients of 0.40 - 0.60 as fair, 0.60 – 0.75 as good and 0.75 of greater as excellent. The coefficients calculated are reported in Table 4.6 and range between 0.68 and 0.79. Based upon these results the degree of agreement with the coding of open ended responses in this thesis is acceptable.

Table 4.6

Kappa tests of agreement

	Kappa value	Asymp. Std. Error(a)	Approx. T(b)	N	Approx. Sig.
1st benefit recorded	0.79	0.066	13.865	45	0.000
2nd benefit recorded	0.68	0.076	13.285	43	0.000
3rd benefit recorded	0.72	0.078	12.506	37	0.000
1st barrier recorded	0.72	0.082	11.837	38	0.000
2nd barrier recorded	0.72	0.088	12.147	28	0.000
3rd barrier recorded	0.76	0.117	9.272	14	0.000

a Not assuming the null hypothesis.
b Using the asymptotic standard error assuming the null hypothesis.

Validity

Table 4.7

Concurrent Validity Correlations

	Anxiety (HADS)	Depression (HADS)	Dispositional optimism (LOT)	Anxious about getting breast cancer	How anxious about genetic testing
Anxiety (HADS)	-	.507**	-.551**	.375**	.239**
Depression (HADS)	.507**	-	-.584**	.102NS	-.038NS
Dispositional optimism (LOT)	-.551**	-.584**	1.00	-.186*	-.090 NS
Anxious about getting breast cancer	.375**	.102NS	-.186*	-	.378**
How anxious about genetic testing	.239**	-.038 NS	-.090 NS	.378**	-

N= 140 – 142. * Correlation is significant at the 0.05 level (2-tailed), ** Correlation is significant at the 0.01 level (2-tailed), NS Not statistically significant.

When examining concurrent or predictive validity, comparing variables that we would expect to be associated with each other, it has been suggested that only correlation coefficients of 0.3 or above are acceptable (Kline, 1993). A strong (or large) positive relationship was found between self-prediction and intention scores (0.864, N=141, P=0.000) (see Table 4.7). Examples of concurrent validity in Table 4.7 include positive correlations between HADS anxiety and HADS depression (0.507, N=142, p=0.000), HADS anxiety and anxiety about developing breast cancer (0.375, N=137, P=0.000), and anxiety about developing breast cancer and anxiety about genetic testing (0.378, N=140, p=0.000). As

would be expected a negative relationship was found between dispositional optimism and: HADS anxiety (-0.551, N=142, p=0.000) and HADS depression (-0.584, N=142, p=0.000).

In Table 4.8 anxiety about developing breast cancer correlated positively with cancer worry (0.706, N=141, p=0.000). Anxiety about genetic testing correlated positively with cancer worry (0.392, N=141, p=0.000). Subjective norm (TPB) correlated positively with: attitudes towards behaviour (TPB) (0.375, N=140, p=0.000) and weighted ratio scores (0.370, N=129, p=0.000). Attitudes towards behaviour correlated positively with weighted ratio scores (0.381, N=128, p=0.000). As would be expected perceived susceptibility correlated positively with: anxiety about developing breast cancer (0.505, N=140, p=0.000), anxiety about genetic testing (0.419, N=140, p=0.000), and cancer worry (0.562, N=137, p= 0.000) (see Table 4.9).

Correlation within the HBM and TPB are to be expected. However, with the exception of attitudes towards behaviour (TPB) and subjective norm (TPB) (0.375, N=140, p=0.000) (see Table 4.8) no statistically significant correlations were found. For the health belief model no factors (including weighted ratio) correlated significantly with each other and gave a coefficient of 0.3 or above (see Table 4.9).

Table 4.8

Concurrent Validity Correlations

	Anxious about getting breast cancer	How anxious about genetic testing	Perceived control (TPB)	Subjective norms (TPB)	Attitudes towards behaviour (TPB)	Perceived susceptibility (HBM)	Perceived severity (HBM)	Weighted ratio	Cancer worry	Coping
Anxious about getting breast cancer	-	.378**	-.124 NS	.286**	.200*	.505**	.213*	.123 NS	.706**	-.118 NS
How anxious about genetic testing	.378**	-	-.104 NS	-.025 NS	-.170*	.419**	.163 NS	-.058 NS	.392**	-.133 NS
Perceived control (TPB)	-.124 NS	-.104 NS	-	-.007 NS	.029 NS	-.074 NS	.076 NS	-.026 NS	-.075 NS	.184*
Subjective norms (TPB)	.281**	-.019 NS	-.009 NS	-	.375**	.153 NS	.208*	.376**	.258**	.014 NS
Attitudes towards behaviour (TPB)	.200*	-.170*	.029 NS	.375**	-	.032 NS	.084 NS	.381**	.052 NS	.099 NS

N= 128 – 142. * Correlation is significant at the 0.05 level (2-tailed), ** Correlation is significant at the 0.01 level (2-tailed), NS Not statistically significant.

Table 4.9

Concurrent Validity Correlations

	Anxious about getting breast cancer	**How anxious about genetic testing**	**Perceived control (TPB)**	**Subjective norms (TPB)**	**Attitudes towards behaviour (TPB)**	**Perceived susceptibility (HBM)**	**Perceived severity (HBM)**	**Weighted ratio**	**Cancer worry**	**Coping**
Perceived susceptibility (HBM)	.505**	.419**	-.074 NS	.156 NS	.032 NS	-	.162 NS	.119 NS	.562**	.009 NS
Perceived severity (HBM)	.213*	.163 NS	.076 NS	.206*	.084 NS	.162 NS	-	.235**	.283**	-.254**
Weighted ratio	.123 NS	-.058 NS	-.026 NS	.370**	.381**	.119 NS	.235**	-	.136 NS	-.039 NS
Cancer worry	.706**	.392**	-.075 NS	.269**	.052 NS	.562**	.283**	.136 NS	-	-.265**
Coping	-.118 NS	-.133 NS	.184*	.010 NS	.099 NS	.009 NS	-.254**	-.039 NS	-.265**	-

N= 125 – 142. * Correlation is significant at the 0.05 level (2-tailed), ** Correlation is significant at the 0.01 level (2-tailed), NS Not statistically significant.

Table 4.10

Concurrent Validity Correlations

	Knowledge of cancer	Knowledge of breast cancer	Knowledge of non-cancer genetic testing	Knowledge of genetic testing for cancer	Knowledge of genetic testing for breast cancer
Knowledge of cancer	-	.748**	.450**	.511**	.496**
Knowledge of breast cancer	.748**	-	.403**	.515**	.559**
Knowledge of non-cancer genetic testing	.450**	.403**	-	.791**	.706**
Knowledge of genetic testing for cancer	.511**	.515**	.791**	-	.914**
Knowledge of genetic testing for breast cancer	.496**	.559**	.706**	.914**	-

N= 142. * Correlation is significant at the 0.05 level (2-tailed), ** Correlation is significant at the 0.01 level (2-tailed), NS Not statistically significant.

All the knowledge items (Table 4.10) correlated positively with coefficients ranging between 0.403 and 0.914.

Validity of Discrete Choice Measures

Acceptability and ease of completion.

All 142 respondents completed the discrete choice questions at baseline and follow-up. Respondents found the discrete choice questions moderate to easy to answer, providing a mean difficulty rating of 5.8 (median = 5) (1=very difficult, 5=moderate, 10=very easy). At follow-up respondents found the DCM questions significantly easier to answer than at baseline (N=141, t=-2.755, df=140, p=0.007), providing a mean rating of 6.3 (Median = 6).

No systematic preference selection for option A or B was found at baseline or follow-up. However, at baseline one respondent stated that they had no preference for all of the 25 discrete choices presented to them. Although this

individual was issued with positive information, they reiterated their initial preference at follow-up; indifference between all of the scenarios presented to them. A further two respondents reported that they had no preference between any of the pairs of choice scenarios presented to them at follow-up. One of these respondents had been issued with positive information and the other with pos-neg information. All three respondents were female. Visual examination of the remaining demographic characteristics revealed no differences with the remainder of the respondents participating in the study.

Non response.

Table 4.11 contains the frequency counts of the choices made at baseline and follow-up from the 23 choices[2] presented to respondents. Very little missing data was recorded for the 23 choices (142 – Total N, in Table 4.11). Only one respondent provided missing data at baseline, a member of the positive group did not give a response to choice 23. At follow-up six respondents provided missing data; a member of the pos-neg group did not answer choices 13 to 18, one member of the pos-neg group did not answer choice 4 and another did not answer choice 24, one member of the positive group did not answer choice 8 and another did not answer choice 9, and a member of the neg-pos group did not answer choice 21.

Response options.

It is important to remember that in addition to allowing respondents to select scenario A or B, this study also had a 'no preference' response option. In this respect, this study differs with the majority of applications of discrete choice modelling in health economics (Bryan et al., 1998; Ryan, 1999a). The 'no preference' response option was used by 2.8% to 8.5% of respondents in each of the choices presented to them (see Table 4.11).

[2] Choices 1 and 25 were included and used solely to test for dominance and transitivity.

Table 4.11

Choices Made at Baseline and Follow-up

	Baseline (T1)						Follow-up (T2)						Total N
	Option A		Option B		No preference		Option A		Option B		No preference		
Choice	**N**	**%**	**N**	**%**	**N**	**%**	**N**	**%**	**N**	**%**	**N**	**%**	
2	37	26.1	99	69.7	6	4.2	35	24.6	101	71.1	6	4.2	142
3	0	0	136	95.8	6	4.2	2	1.4	134	94.4	6	4.2	142
4	106	74.6	29	20.4	7	4.9	109	76.8	24	16.9	8	5.6	141
5	69	48.6	66	46.5	7	4.9	82	57.7	49	34.5	11	7.7	142
6	119	83.8	18	12.7	5	3.5	110	77.5	21	14.8	11	7.7	142
7	39	27.5	92	64.8	11	7.7	33	23.2	99	69.7	10	7.0	142
8	41	28.9	92	64.8	9	6.3	40	28.2	91	64.1	10	7.0	141
9	51	35.9	84	59.2	7	4.9	55	38.7	79	55.6	7	4.9	141
10	20	14.1	110	77.5	12	8.5	19	13.4	116	81.7	7	4.9	142
11	107	75.4	25	17.6	10	7.0	97	68.3	34	23.9	11	7.7	142
12	74	52.1	57	40.1	11	7.7	76	53.5	59	41.5	7	4.9	142
13	7	4.9	127	89.4	8	5.6	4	2.8	133	93.7	4	2.8	141
14	116	81.7	17	12.0	9	6.3	121	85.2	15	10.6	5	3.5	141
15	47	33.1	85	59.9	10	7.0	45	31.7	87	61.3	9	6.3	141
16	10	7.0	125	88.0	7	4.9	10	7.0	123	86.6	8	5.6	141
17	123	86.6	13	9.2	6	4.2	111	78.2	22	15.5	8	5.6	141
18	122	85.9	16	11.3	4	2.8	110	77.5	26	18.3	5	3.5	141
19	117	82.4	15	10.6	10	7.0	120	84.5	14	9.9	8	5.6	142
20	48	33.8	82	57.7	12	8.5	42	29.6	88	62	12	8.5	142
21	101	71.1	31	21.8	10	7.0	104	73.2	27	19	10	7.0	141
22	31	21.8	106	74.6	5	3.5	26	18.3	107	75.4	9	6.3	142
23	109	76.8	23	16.2	9	6.3	108	76.1	26	18.3	8	5.6	141
24	22	15.5	113	79.6	7	4.9	18	12.7	118	83.1	5	3.5	141

Construct validity.

Table 4.12

Discrete Choice (Multinomial Logit) Model for Attribute Main Effects at Baseline

Attributes	Levels	Coefficients (B)	Standard error	t-ratio	P value
Staff seen	Consultant geneticist	0.843261	0.0689592	12.2284	2.88658e-015
	Specialist genetics nurse	-0.47997	0.0869375	-5.52086	3.37348e-008
	Genetics associate	-0.363292	0.0934029	-3.88951	0.000100446
Waiting time for letter	1 month	1.41628	0.0955571	14.8213	2.88658e-015
	2 month	0.927039	0.0801314	11.569	2.88658e-015
	4 month	-0.55868	0.0961259	-5.81196	6.17455e-009
	6 month	-1.78464	0.14652	-12.1802	2.88658e-015
Distance to counselling	20 miles	0.269486	0.108007	2.49508	0.0125928
	40 miles	0.82404	0.10167	8.10502	2.88658e-015
	60 miles	-1.04442	0.109689	-9.5217	2.88658e-015
	80 miles	-0.0491044	0.0950789	-0.516459	0.605534
Duration of counselling	30 minutes	-1.3567	0.13322	-10.1839	2.88658e-015
	1 hour	0.224441	0.101384	2.21377	0.0268448
	1 hour 30 minutes	0.526961	0.0959232	5.49358	3.93876e-008
	2 hour	0.6053	0.0835327	7.24626	4.28546e-013
Availability of testing	High risk	-0.151346	0.0607661	-2.49063	0.0127518
	All	0.151346	0.0607661	2.49063	0.0127518
Cost of service	£1,500	1.10392	0.0945193	11.6793	2.88658e-015
	£2,000	0.525168	0.105662	4.97025	6.68657e-007
	£2,500	0.0240341	0.0889284	0.270263	0.786958
	£3,000	-1.65312	0.132718	-12.4559	2.88658e-015
N = 142, Number of observations=3254, Log likelihood function=-2386.441, Restricted log likelihood (Log-L for Choice model) = -2386.4407, R^2(McFadden's R^2)=0.33244, Adjusted R^2(Adjusted McFadden's R^2)=0.33090.					

Note: By specifying an alternate baseline level it is possible to calculate the coefficients for the levels used as a baseline in the base case model. As this analysis is based on a pre to post experimental design missing data in a baseline or follow-up choice has necessitated the removal of the matching data from the corresponding time point.

Construct validity is the extent to which the instrument is measuring or testing the intended theory or construct. Construct validity is evaluated in terms of concurrent (positive concurrent and discriminant) validity and theoretical validity.

Concurrent.

The statistically significant ($p<0.05$) B coefficients in Table 4.12 revealed, for the nominal attribute of availability of testing, that testing available to all was preferred to testing only being available to high risk patients. In terms of staff seen for counselling ($p<0.001$), respondents would prefer to be counselled by a genetics associate rather than a nurse, and a consultant geneticist rather than a

genetics associate. In order to validate the results for these two attributes respondents were asked to rank each of their levels before choosing between the discrete choice scenarios presented to them. Ranking of the availability of testing (see Table 4.13) complied with the coefficients produced by the multinomial logit regression model. The ranking of staff seen for counselling (Table 4.14) did not correspond with the regression coefficients; whilst the consultant was the first choice in both the regression and the ranking exercise; on the ranking exercise a nurse was preferred to a genetics associate.

Table 4.13

Ranking of Genetic Testing Availability

	Mean	Median	N	Missing
Test all	1.1849	1.0000	119	23
Test high risk	1.8151	2.0000	119	23

Note: The preferred option was ranked 1 and the least preferred was ranked 2.

Table 4.14

Ranking of Preferred Counsellor

	Mean	Median	N	Missing
Consultant	1.5104	1.0000	96	46
Nurse	2.0645	2.0000	93	49
Associate	2.3936	3.0000	94	48

Note: The preferred option was ranked 1, the second preference 2 and the least preferred was ranked 3.

Theoretical/internal validity.

Theoretical validity also referred to as 'a priori theory' or internal validity relates to how well the instrument conforms to theoretical expectations.

Theoretical expectations for the un-segmented model in Table 4.12 include:

- Utility increases as cost declines.
- Utility increases as waiting time declines.
- Utility increases as duration of counselling increases.
- Utility increases as distance to counselling declines.
- Preferences are transitive.
- Preferences are continuous (improvement in one attribute can compensate for a reduction in another).

The size and sign of the attribute level coefficients (B) relative to the baseline values coded in the multinomial logit model for the continuous attributes of waiting time, duration of counselling and cost of service were in accordance with expectations. In terms of waiting time utility increased as the waiting time for a letter confirming risk status declined. Utility increased as duration of counselling session increased and utility increased as the cost of the service declined. Distance to counselling did not comply with expectations as greater utility was associated with travelling 40 miles than 20 miles.

Dominance, transitivity and non-satiation tests.

Three respondents always selected a scenario where the waiting time was lowest regardless of the other attribute levels. 24 respondents always selected the scenario where testing was available to all (no one consistently selected the option where testing was only available to high risk patients). No dominant preference patterns were found for the remaining attributes. As 'staff seen' is a nominal attribute with three categories this was not assessed for dominance as there was no a priori assumptions upon which to base a dominance check. When the dominant preference patterns were compared with respondents rankings of which attribute was the most important to them it emerged that six respondents had a dominant preference according to Scott's (2002) criteria. Two respondents had a dominant preference for the attribute waiting time for a letter and four respondents had a dominant preference for the attribute availability of testing (preference for testing for all regardless of their risk status).

Table 4.15

Baseline Transitivity Tests

	Choice 1		**Choice 3**		**Choice 13**		**Choice 25**	
	N	**%**	**N**	**%**	**N**	**%**	**N**	**%**
Option A	135	95.1	0	0	7	4.9	4	2.8
Option B	0	0	136	95.8	127	89.4	133	93.7
No preference	7	4.9	6	4.2	8	5.9	5	3.5
Missing	0	0	0	0	0	0	0	0

Note: The dominant option in choices 1, 3, 13 and 25 were respectively, option A, option B, option B and option B.

The majority of respondents (89.4% - 95.8%) passed each of the four dominated tests at baseline (see Table 4.15), selecting the clearly superior scenario. In

conjunction, choices 3, 13 and 25 formed a test of transitivity. Respondents with transitive preferences would be expected to select 'option B' in all three choices. 20 respondents failed to select option B in one or more of the three choices. Additionally, three respondents passed the transitivity tests but failed the non-satiation (dominated choice) test in choice 1.

In total 28 respondents failed the baseline dominance, transitivity and non-satiation tests conducted. Respondents that failed the tests were aggregated into one group in order to compare them with the remaining 114 that passed the tests. No significant difference was found between those passing and failing the utility theory axiom tests on the reported difficulty of completing the discrete choice questions. Comparison by demographic and psychological characteristic revealed that there were significantly more mature students (two tailed Fishers exact test, $p=0.017$) and parents (all mature students) (two tailed Fishers exact test, $p=0.004$) amongst the respondents failing the axiom tests. Respondents failing the test also had significantly higher optimism scores, 20.9 comported to 17.5 ($N=142$, $t=-2.367$, $df=140$, $p=0.019$). No significant differences were found by information group, or on the outcome measures of the weighted ratio of pros to cons, self-prediction and intention to obtaining genetic testing and counselling at baseline.

Relative importance of attribute levels.

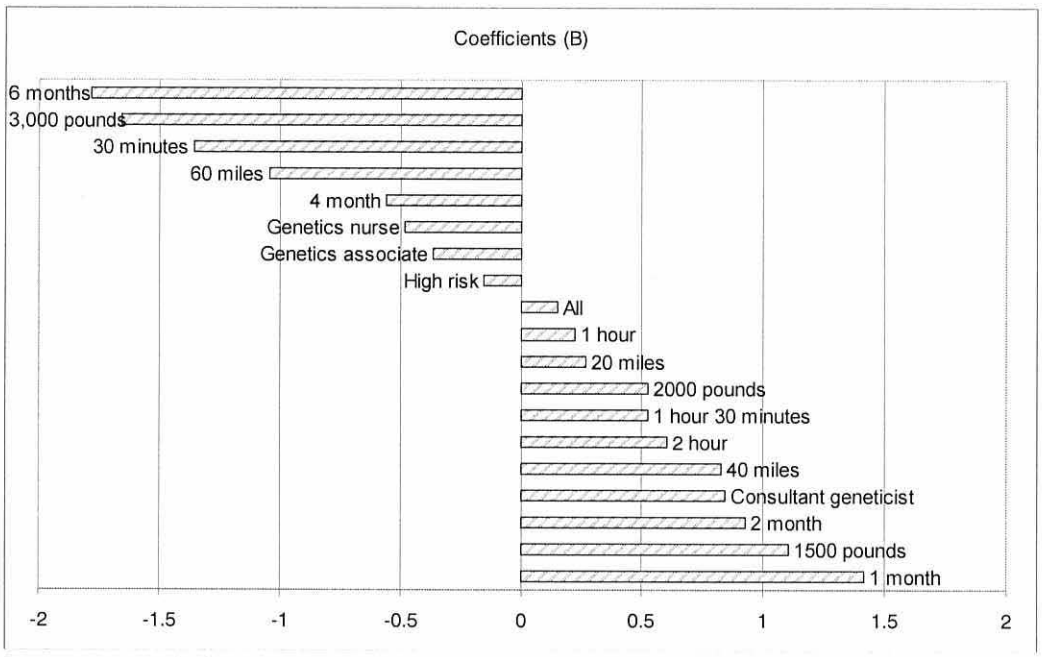


Figure 4.1. Relative importance of attribute levels.

Figure 4.1 contains the attribute levels ranked in order of their subjective expected utility (random utility), from least to most utility maximising (desirability). Five attributes appear among the six attribute levels providing the greatest utility. The mix in the order of the attribute levels confirms that the respondents as a whole had continuous preferences; a weak level of one attribute can be compensated for with a more desirable level of another attribute.

Table 4.16

Multinomial Logit Models for Attribute Main Effects by Utility Theory Axiom Test Results

		Failed axiom tests				Passed axiom tests			
Attributes	**Levels**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**
Staff seen	Consultant geneticist	0.628945	0.133386	4.71524	2.41426e-006	1.00326	0.0830017	12.0872	2.88658e-015
	Specialist genetics nurse	-0.531207	0.160074	-3.31851	0.000905001	-0.508045	0.10387	-4.89116	1.00242e-006
	Genetics associate	-0.0977379	0.169511	-0.576589	0.564217	-0.495216	0.113567	-4.36056	1.29733e-005
Waiting time for letter	1 month	0.325647	0.182326	1.78607	0.0740877	1.97316	0.125751	15.691	2.88658e-015
	2 month	0.600908	0.152952	3.92874	8.53937e-005	1.12519	0.0950089	11.843	2.88658e-015
	4 month	-0.345103	0.167577	-2.05937	0.0394587	-0.655453	0.117009	-5.60172	2.12241e-008
	6 month	-0.581451	0.213266	-2.72641	0.00640279	-2.4429	0.189294	-12.9053	2.88658e-015
Distance to counselling	20 miles	0.271203	0.18388	1.47489	0.140242	0.163503	0.134021	1.21998	0.222472
	40 miles	0.357169	0.166836	2.14084	0.032287	1.17975	0.134594	8.76525	2.88658e-015
	60 miles	-0.440477	0.208989	-2.10766	0.0350607	-1.36088	0.127409	-10.6812	2.88658e-015
	80 miles	-0.187895	0.16794	-1.11882	0.263217	0.0176255	0.116136	0.151765	0.879372
Duration of counselling	30 minutes	-0.369176	0.20766	-1.7778	0.0754375	-1.83258	0.159339	-11.5011	2.88658e-015
	1 hour	0.0636204	0.174615	0.364347	0.715599	0.225693	0.130249	1.73278	0.0831346
	1 hour 30 minutes	0.0589636	0.200551	0.294008	0.768751	0.794676	0.109639	7.2481	4.22551e-013
	2 hour	0.246592	0.154816	1.59281	0.111202	0.812207	0.105453	7.70208	1.33227e-014
Availability of testing	High risk	-0.374641	0.106778	-3.50859	0.000450481	-0.0545563	0.0725164	-0.75233	0.451853
	All	0.374641	0.106778	3.50859	0.000450481	0.0545563	0.0725164	0.75233	0.451853
Cost of service	£1,500	0.394676	0.179582	2.19775	0.0279669	1.53586	0.127855	12.0125	2.88658e-015
	£2,000	0.196418	0.20366	0.96444	0.334825	0.642542	0.126395	5.08358	3.70379e-007
	£2,500	0.131286	0.153907	0.853023	0.393647	-0.109252	0.107633	-1.01504	0.310086
	£3,000	-0.72238	0.224353	-3.21983	0.00128265	-2.06915	0.159431	-12.9784	2.88658e-015
	N =28, Number of observations=636, Log likelihood function=-585.3825, Restricted log likelihood (Log-L for Choice model) =-585.3825, R^2=0.1622, Adjusted R^2=0.15221.					N =114, Number of observations=2618, Log likelihood function=-1724.766, Restricted log likelihood (Log-L for Choice model) =-1724.7663, R^2=0.40032, Adjusted R^2=0.39860.			

Table 4.16 contains the baseline attribute main effects for those passing the utility theory axiom tests and those failing the tests. Comparison of the un-segmented baseline model (Table 4.12) with the baseline models segmented according to passing and failing the axiom tests (Table 4.16), by means of a likelihood ratio test revealed that there was a significant difference (N=142, χ^2 = 152.585, df=15, p<0.001)[3]. There was therefore a significant difference between the preferences of those respondents passing the tests and those failing the dominance and transitivity tests.

Examination of the attribute main effects in Tables 4.12 and 4.16 reveal where the differences lie. As was the case for the un-segmented model (N=142), the attributes of cost of service equal to £2,500 and distance to counselling equal to 80 miles were statistically non-significant for those passing (N=114) and those failing (N=28) the axiom tests. For those passing the tests, the choice model differed from the un-segmented model by the fact that the attribute level 20 miles distance to counselling and both levels of the attribute availability of testing were non-significant.

The respondents that failed the axiom (transitivity, dominance and non-satiation) tests had a very different utility/choice function to those that passed. In addition to cost=£2,500 and distance=80 miles, the attribute levels: Staff seen = Genetics associate; distance to counselling = 20 miles; Duration of counselling = 1 hour, 1 hour 30 minutes, 2 hours and cost=£2,000 were statistically non-significant.

The utility function for the respondents that failed the dominance and transitivity test had the lowest number of significant attributes (p<0.10) at thirteen (21 attribute levels in total). This utility function explained 15% (adjusted R^2=0.152213) of the variance in the discrete choices made. The utility function for the respondents passing the dominance and transitivity tests had sixteen significant predictors attribute levels (p<0.10) and explained 40% (adjusted R^2=0.39860) of the variance in the discrete choices made. Excluding the respondents that failed the tests therefore increased the explained variance in

[3] Likelihood ratio test = 2[(-2386.441) - [(-585.3825) + (-1724.766)]] = -152.585.

choices by 7% (un-segregated model's adjusted R^2=0.33090). The remainder of the analysis will be conducted with all 142 respondents and no exclusions (see discussion for an explanation).

Sample Characteristics and Pre-experimental Information Group Comparisons (N=142)

Continuity within Information Groups

As the controls were split into two groups due to a lack of one room sufficiently large to run the experiment with them as a whole, the first step was to assess if there were any major differences between both groups. No statistically significant differences were found in terms of age, gender, number of respondents that were parents, ethnicity, social class, anxiety, depression or dispositional optimism. As no significant differences were found between the control groups on the main outcome measures of the weighted ratio of pros to cons, self-prediction or intention to obtaining genetic testing and counselling at baseline or from baseline to post information manipulation they have been combined and treated as one group for the remainder on the analyses.

Hypothesis 3 states that the neg-pos information will result in a statistically significantly greater decline in the ratio of pros to cons between assessments (baseline to follow-up) than the pos-neg information (ordering/primacy effect). This pattern would also be expected to appear in respondents' self-prediction and intention to have genetic testing and counselling with both groups showing a decline in ratings of intention and self-prediction with the neg-pos group showing the greatest decline (hypothesis 6). However if this hypothesis is incorrect and there is no information ordering/primacy effect both groups can be combined.

No statistically significant differences were found in terms of age, gender, number of respondents that were parents, ethnicity, social class, anxiety, depression or dispositional optimism. No significant differences were found between the pos-neg and neg-pos groups on the outcome measures of weighted ratio of pros to cons, self-prediction or intention to obtain genetic testing and counselling at baseline or from baseline to post information manipulation.

Table 4.17

Weighted Ratio, Self-prediction and Intention Variables' Estimated Marginal Means at Baseline (T1) and Follow-up (T2)

Information group	**Assessment**	**Weighted ratio**			**Self-prediction**			**Intention**		
		N	Mean	Std. error	N	Mean	Std. error	N	Mean	Std. error
Pos-neg	Baseline	35	0.590	0.68	37	54.865	4.973	37	7.032	0.572
	Follow-up	35	0.373	0.68	37	55.541	4.846	37	7.446	0.569
Neg-pos	Baseline	26	0.508	0.78	31	50.161	5.433	31	6.632	0.625
	Follow-up	26	0.196	0.79	31	45.484	5.295	31	6.123	0.622

Although no statistically significant differences were found between the pos-neg and neg-pos groups, the descriptive statistics in Table 4.17 show that these two groups have not reacted in a uniform pattern or in full accordance with expectations. Both groups recorded a decline in their weighted ratio scores and the largest decline was recorded by the neg-pos group. In contrast to the weighted ratio results the pos-neg group recorded an increase in self-prediction and intention scores whilst the neg-pos recorded a decline on both scores.

Despite no significant differences being found between the pos-neg group and the neg-pos group, the results recorded for both groups on the weighted ratio, self-prediction and intention scores suggest that there is an information ordering effect and as a result these groups will not be combined for any analyses.

Sample Characteristics

Table 4.18

Sample Characteristics (N=142)

		Pos	Pos-neg	Neg-pos	Control	All
	N	38	37	31	36	142
Demographic characteristics						
Age	Mean	19.3	21.9	19.1	20.6	20.2
	Median	18.5	18.0	18.0	18.0	18.0
	Standard deviation	3.0	9.0	1.7	6.0	6.0
Gender	Male	4 (10.5%)	6 (16.7%)	5 (16.1%)	8 (22.2%)	23 (16.3%)
	Female	34 (89.5%)	30 (83.3%)	26 (83.9%)	28 (77.8%)	118 (83.7%)
Parents	Yes	1 (2.6%)	4 (10.8%)	0 (%)	3 (8.3%)	8 (5.6%)
	No	37 (97.4%)	36 (89.2%)	31 (100%)	33 (91.7%)	135 (94.4%)
Ethnicity	White	36 (94.7%)	34 (91.9%)	27 (87.1%)	31 (88.6%)	128 (90.8%)
	Black African	1 (2.6%)	0 (0%)	0 (0%)	0 (1%)	1 (0.7%)
	Black Caribbean	0 (0%)	0 (0%)	1 (3.2%)	0 (0%)	1 (0.7%)
	Indian	1 (2.6%)	0 (0%)	0 (0%)	0 (0%)	1 (0.7%)
	Bangladeshi	0 (0%)	0 (0%)	0 (0%)	1 (2.9%)	1 (0.7%)
	Chinese	0 (0%)	0 (0%)	2 (6.5%)	1 (2.9%)	3 (2.1%)
	Japanese	0 (0%)	0 (0%)	1 (3.2%)	0 (0%)	1 (0.7%)
	Armenian	0 (0%)	0 (0%)	0 (0%)	1 (2.9%)	1 (0.7%)
	Persian	0 (0%)	0 (0%)	0 (0%)	1 (2.9%)	1 (0.7%)
	Other	0 (0%)	3 (8.1%)	0 (0%)	0 (0%)	3 (2.1%)
New socio economic classification	Higher managerial	1 (2.6%)	5 (13.5%)	6 (21.4%)	2 (6.1%)	14 (10.3%)
	Professionals	15 (39.5%)	12 (32.4%)	7 (25.0%)	14 (42.4%)	48 (35.3%)
	Lower managerial & professional	9 (23.7%)	7 (18.9%)	7 (25.0%)	8 (24.2%)	31 (22.8%)
	Intermediate	1 (2.6%)	2 (5.4%)	1 (3.6%)	3 (9.1%)	7 (5.1%)
	Small employer & own account workers	3 (7.9%)	6 (16.2%)	1 (3.6%)	3 (9.1%)	13 (9.6%)
	Supervisors/craft related	4 (10.5%)	1 (2.7%)	3 (10.7%)	2 (6.1%)	10 (7.4%)

	Semi-routine occupations	3 (7.9%)	3 (8.1%)	2 (7.1%)	1 (3.0%)	9 (6.6%)
	Routine occupations	2 (5.3%)	1 (2.7%)	1 (3.6%)	0 (0%)	4 (2.9%)
Psychological characteristics						
HADS anxiety scores	Mean	9.2	8.5	8.3	9.2	8.9
	Median	10.0	8.0	8.0	9.0	8.0
	Standard deviation	3.2	3.3	3.8	4.1	3.6
	Normal (0-7)	13 (34.2%)	17 (45.9%)	12 (38.7%)	13 36.1%)	55 (38.7%)
	Mild (8-10)	10 (26.3%)	12 (32.4%)	12 (38.7%)	12 (33.3%)	46 (32.4%)
	Moderate (11-14)	15 (39.5%)	4 (10.8%)	4 (12.9%)	7 (19.4%)	30 (21.1%)
	Severe (15-21)	0 (0%)	4 (10.8%)	3 (9.7%)	4 (11.1%)	11 (7.7%)
HADS depression scores	Mean	3.8	3.7	4.0	4.1	3.9
	Median	4.0	4.0	4.0	4.0	4.0
	Standard deviation	2.5	2.5	2.9	2.4	2.5
	Normal (0-7)	35 (92.1%)	34 (91.9%)	25 (80.6%)	34 (94.4%)	128 (90.1%)
	Mild (8-10)	3 (7.9%)	3 (8.1%)	6 (19.4%)	2 (5.6%)	14 (9.9%)
	Moderate (11-14)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
	Severe (15-21)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
LOT dispositional optimism score	Mean	18.1	19.8	17.1	17.6	18.2
	Median	17.5	21.0	17.0	18.0	19.0
	Standard deviation	6.7	6.6	6.5	7.5	6.9

Demographic characteristics.

Sample characteristics for all respondents, including those excluded are presented in Appendix P. Of the 142 first year psychology students meeting the inclusion criteria 23 were male (16.3%) and 118 were female (83.7%) (missing data = 1). The mean age of respondents was 20.2 years (min = 17, max =53, median =18, standard deviation = 6.0 years), 16.4% were mature students at 21 years of age or greater (7.9% were 25 plus). Eight participants had children (min = 1, max = 4). In terms of ethnicity (see Table 4.18) the majority of participant were white (90.8%); 2.1% categorised themselves as other (not one of the ten groups listed in Table 4.18) and did not state their ethnic background. It should be noted that the Japanese and other cultures with an interdependent construal of self have been found to associate wellbeing with belonging or a group perspective rather than an individual perspective (Kitayama et al., 1994; Heine & Lehman, 1995). However, as there is only one Japanese respondent in the study

this should not cause a problem. No participants were of Ashkenazi Jewish origin. The prevalence of BRCA1/2 mutations is much higher for Ashkenazi Jews than any other ethnic group at 1.0% - 2.5% (Struewing, Abeliovich, et al.,1995; Oddoux et al., 1996; Tonin et al., 1996) compared to 0.25% - 0.5% in the general public (Brook, 1999). Based upon the New Socio Economic classification (Rose & O'Reilly, 1998) of household heads, participants in this study are atypical of the national populations of Wales and England (see Appendix Q). There is a strong bias towards professional backgrounds amongst the participants in the study; a finding that is also common amongst other European familial breast cancer clinics (Steel et al., 1999).

Psychological characteristics.

There were four outliers in terms of anxiety scores; 2 outliers below and two above the median. No extreme values were found. Anxiety levels were high with a mean score of 8.3 to 9.2, scores of 11 and above are clinically significant. Even the lowest mean score of 8.3 was significantly higher than the norm of 6.14 (Crawford et al., 2001) (one sample t test: t=3.163, df=30, p=0.004); this finding remained even when the extreme values were excluded from the analysis. The norms in terms of mean and standard deviation scores on the dispositional optimism (LOT) test for undergraduate students are, 21.03 (sd 4.56) based on 357 males and 21.41 (sd 5.22) for 267 females (Scheier & Charles, 1985). Dispositional optimism scores were lower than the established norms for both men and women at 19.13 and 17.92 women (mean scores) respectively. The score for women was significantly lower than the established norm (one sample t test: t=-5.514, df=117, p=0.001).

Demographic and psychological characteristics by information groups.

No significant differences were found between the information groups (positive, pos-neg, neg-pos and control) on age, gender, number of respondents that were parents, social class, anxiety, depression or dispositional optimism.

Survey of Patients Referred for Cancer Genetics Services

Validity of Discrete Choice Measures

Acceptability and ease of completion.

Respondents found the discrete choice questions moderate to easy to answer, providing a mean difficulty rating of 6.3 (N=115) (1 = Very difficult, 5 = Moderate, 10 = Very easy). The 'no preference' response option was used by 6.1% to 13.9% (n= 7 to 16) of respondents in each of the choices presented to them and only 5% (n=6) of returned DCM questionnaires had missing data. Given the low number of missing values and 'no preference' responses in the latter half of the DCM questionnaire, it is unlikely that boredom or fatigue had set in and were affecting responses (San Miguel et al., 2005; Verhoef et al., 1991).

Construct validity.

Table 4.19

Discrete Choice (Multinomial Logit) Model for Attribute Main Effects at Baseline

Attributes	Levels	Coefficients (B)	Standard error	t-ratio	P value
Staff seen	Consultant geneticist	0.752182	0.0805994	9.33236	2.88658e-015
	Specialist genetics nurse	-0.244905	0.0979457	-2.50042	0.0124047
	Genetics associate	-0.507277	0.11217	-4.52241	6.11382e-006
Waiting time for letter	1 month	1.22785	0.0969927	12.6592	2.88658e-015
	2 month	0.679595	0.0957352	7.09869	1.25944e-012
	4 month	-0.609606	0.107205	-5.68634	1.29792e-008
	6 month	-1.29784	0.147065	-8.82493	2.88658e-015
Distance to counselling	20 miles	0.60168	0.108364	5.5524	2.81775e-008
	40 miles	0.521625	0.111965	4.65881	3.18037e-006
	60 miles	-0.954636	0.131349	-7.26791	3.65041e-013
	80 miles	-0.16867	0.101865	-1.65581	0.0977597
Duration of counselling	30 minutes	-0.972312	0.139727	-6.95863	3.43592e-012
	1 hour	0.234426	0.102848	2.27934	0.0226468
	1 hour 30 minutes	0.437806	0.110226	3.97188	7.13078e-005
	2 hour	0.30008	0.0919916	3.26204	0.00110612
Availability of testing	High risk	-0.381895	0.0687416	-5.55551	2.76796e-008
	All	0.381895	0.0687416	5.55551	2.76796e-008
Cost of service	£1,500	0.485889	0.108545	4.47638	7.59205e-006
	£2,000	0.40624	0.122193	3.32458	0.000885524
	£2,500	0.0810544	0.09374	0.864673	0.387218
	£3,000	-0.973184	0.137877	-7.05836	1.68487e-012
N = 115, Number of observations=2633, Log likelihood function=-1934.156, Restricted log likelihood (Log-L for Choice model) =-1934.1555, R^2(McFadden's R^2)=0.33135, Adjusted R^2(Adjusted McFadden's R^2)=0.32944.					

Concurrent.

Table 4.19 reveals for the attribute 'availability of testing' that testing available to all was preferred to testing only being available to high risk patients. In terms of staff seen for counselling, respondents would prefer to be counselled by a specialist genetics nurse than a genetics associate, and a consultant geneticist rather than a genetics associate. Ranking of the staff seen for counselling and availability of testing (see Tables 4.20 and 4.21) prior to completing the DCM exercise complied with the coefficients produced by the multinomial logit regression model.

Table 4.20

Ranking of Genetic Testing Availability

	Mean	Median	N	No Preference	Missing
Test all	1.25	1.0	88	21	6
Test high risk	1.75	2.0	88	21	6

Note: The preferred option was ranked 1 and the least preferred was ranked 2.

Table 4.21

Ranking of Preferred Counsellor

	Mean	Median	N	No Preference	Missing
Consultant	1.56	1.0	34	59	22
Nurse	1.88	2.0	34	59	22
Associate	2.56	3.0	34	59	22

Note: The preferred option was ranked 1, the second preference 2 and the least preferred was ranked 3.

Theoretical/internal validity.

The theoretical expectations for the un-segmented model in Table 4.19 were:

- Utility increases as cost declines.
- Utility increases as waiting time declines.
- Utility increases as duration of counselling increases.
- Utility increases as distance to counselling declines.

The size and sign of the attribute level coefficients (B) relative to the baseline values coded in the multinomial logit model for the continuous attributes of 'waiting time' and 'cost of service' complied with expectations. Utility increased as the waiting time for a letter confirming risk status declined and cost of service

declined. The level £2,500 was non-significant (p=0.378) for the attribute 'cost of service'.

The non-significant (correcting to two decimal places) coefficient for the level '80 miles distance to counselling ' did not comply with expectations; providing a coefficient indicating less dissatisfaction with travelling 80 miles than travelling 60 miles. The remaining levels of the attributes complied with expectations, with utility increased as distance to counselling venue declined. 'Duration of counselling' complied with theoretical expectations for the three shortest time intervals (30 minutes, 1 hour, 1 hour 30 minutes) with utility increasing as duration did. The longest counselling interval of '2 hours' did not comply with expectations, yielding a coefficient value between those of '1 hour' and '1 hour 30 minutes'.

Dominance, transitivity and non-satiation tests.

In total 59 (49.2%) respondents either failed the dominance, transitivity and/or non-satiation tests conducted. As was noted previously in relation to the analysis of the experiment, these patients were retained in the analysis (see discussion for an explanation).

Sample Characteristics

Patients' clinical and demographic characteristics are presented in Table 4.22. 42% of patients referred to the CGSW were based in South East Wales and referred to the University Hospital of Wales (Cardiff), 32.8% were based in the South west and referred to Heath Hospital (Swansea) and 25.2% were based in North Wales and referred to Ysbyty Glan Clwyd.

72.3% of patients had a family history of breast cancer. 33% of the patients were at high risk of developing genetic cancer due to their family history, 41% were at moderate risk and 26% were at low risk (population risk). The sample was almost entirely female at 98.3%. The mean age of respondents was 44.5 years; 15 years below the minimum eligibility age for UK national breast screening programs. Compared to the Welsh national average, the patient sample comprised of a greater proportion of individuals with academic qualifications.

Whilst the national figures for 2001 National Population Census (Office for national statistics (ONS, 2003)) were 33% with no qualifications and 17% with a qualification at degree level or above, the patients had only 26.5% with GCSE qualifications at grade three or below and 23.1% with a qualification at degree level or above. Substantially more patients were in employment (77.4%) than the national average for Wales in 2001 of 55.2%(ONS, 2003). In terms of the New Socio Economic Classification (Rose & O'Reilly, 1998) the sample of patients does differ to the national population of Wales. The most marked differential was in terms of the percentage of individuals categorised as routine occupations; only 4% of the patients were in this category, whilst Wales as a whole has 14.8%.

Table 4.22

Clinical Characteristics and Information

Clinical characteristics & demographic characteristics		**N (%)**
Cancer centre	Ysbyty Glan Clwyd, Bodelwyddan, North Wales	30 (25.2%)
	University Hospital of Wales, Cardiff	50 (42.0%)
	Heath Hospital, Swansea	39 (32.8%)
	N	119
Cancer family history	Breast & Ovarian	13 (10.9%)
	Breast	86 (72.3%)
	Ovarian	10 (8.4%)
	Colorectal	7 (5.9%)
	Other	3 (2.5%)
	N	119
Risk	High	40 (33.3%)
	Moderate	49 (40.8%)
	Low	31 (25.8%)
	N	120
Age	Mean	44.53
	Median	44.50
	Standard deviation	10.77
	N	120
Gender	Male	2 (1.7%)
	Female	118 (98.3%)
	N	120
Educational attainment	GCSE grade 3 or below	31 (26.5%)
	O level - NVCQ 2	38 (32.5%)
	A level - NVQ level 3	14 (12.0%)
	Higher diploma - HND	5 (4.3%)
	Degree or above	27 (23.1%)
	Other	2 (1.7%)
	N	117
Work status	Working	89 (77.4%)
	In full-time education	1 (0.9%)
	Looking after home/family	3 (2.6%)
	Permanently sick/disabled	5 (4.3%)
	Retired	13 (11.3%)
	Other/unemployed	4(3.5%)
	N	115
New socio economic[4] classification	Higher managerial	5 (4.4%)
	Professionals	11 (9.7%)
	Lower managerial & professional	24 (21.2%)
	Intermediate	17 (15.0)
	Small employer & own account workers	8 (7.1%)
	Supervisors/craft related	15 (13.3)
	Semi-routine occupations	28 (24.8%)
	Routine occupations	5 (4.4%)
	N	113

[4] In order to improve coverage Rose and O'Reilly (1998) recommend that when employing the new socio economic classification that those not in employment are allocated according to their last main job.

CHAPTER 5: RESULTS OF AIMS OBJECTIVES AND HYPOTHESIS TESTING

Information Manipulation Experiment

The Perceived Benefits and Barriers of Having Cancer Genetic Testing

As random utility theory proposes that individuals weigh up the benefits and barriers (pros and cons) of their options when making a choice and select the option that provides them with the maximum subjective expected utility, experiment participants were asked to list all the benefits and barriers of cancer genetic testing they could think of. Reasons given by participants in favour (benefits) and against (barriers) testing and counselling were post-coded according to their meaning.

The benefits of testing and counselling at baseline and follow-up all fell into twelve categories (see Table 5.1). At baseline the disadvantages of testing and counselling were coded into 14 categories (see Table 5.2). After issuing participants with the experiment information, an additional category of response emerged on seven occasions. The new category was "Increased health risk from having mammography". The frequency with which each of the benefits and barriers were cited at baseline (prior to issuing experimental information), are presented in Tables 5.1 and 5.2 in descending order of the frequency with which they were cited.

By far the most frequently cited benefits at baseline were "prevention/early detection" and "promoting greater understanding of breast cancer", 49.9% of all the benefits listed at baseline fell into these two categories. The third most popular category was "discover susceptibility status". The most frequently cited barrier of genetic testing and counselling was "increased worry/anxiety/distress/depression", which accounted for 31.1% of all the barriers reported. The remaining barriers were far less frequently cited with the second most popular category "life insurance problems/financial issues/test expensive/time/travel" only accounting for 10.2% of the barriers cited. (For

information purposes follow-up benefits and barriers are presented in Appendix R).

Table 5.1

Frequency with Which Each Benefit was Cited at Baseline

Benefit	**Reasons**												
	1st	**2nd**	**3rd**	**4th**	**5th**	**6th**	**7th**	**8th**	**9th**	**10th**	**11th**	**Total**	**%**
Prevent, surveillance, early treatment & inc health behaviour	55	47	20	7	4	3	3	1	0	0	0	140	30.9
Promote greater understanding of breast cancer/ awareness	21	22	16	11	10	1	1	1	1	1	1	86	19.0
Discover breast cancer susceptibility status	21	6	9	2	1	0	0	0	0	0	0	39	8.6
Social support / counselling	5	6	11	10	3	2	1	0	0	0	0	38	8.4
Come to terms with the possibility of breast cancer /prepare	8	6	10	5	2	2	0	0	0	0	0	33	7.3
Help family come to terms with the prospect of breast cancer	1	5	7	7	3	2	0	0	1	0	0	26	5.7
Other biomedical	4	9	7	1	0	1	1	0	0	0	0	23	5.1
Other psychological	4	5	4	3	4	1	0	0	0	0	0	21	4.6
Reduce psychological distress/worry	3	6	3	2	3	1	0	1	0	0	0	19	4.2
Other miscellaneous	2	3	3	2	0	0	0	0	0	0	0	10	2.2
Family planning/children	1	2	4	2	0	0	1	0	0	0	0	10	2.2
Save lives	5	2	1	0	0	0	0	0	0	0	0	8	1.8
Sub Total	**130**	**119**	**95**	**52**	**30**	**13**	**7**	**3**	**2**	**1**	**1**	**453**	**100.0**
Missing	12	23	47	90	112	129	135	139	140	141	141	-	-
Total	142	142	142	142	142	142	142	142	142	142	142	-	-

Table 5.2

Frequency with Which Each Barrier was Cited at Baseline

Disadvantages	**Reasons**							
	1st	**2nd**	**3rd**	**4th**	**5th**	**6th**	**Total**	**%**
Increased worry/ anxiety/distress/ depression	41	20	8	2	2	0	73	31.1
Life insurance, problems/ financial issues/ test expensive	10	6	3	5	0	0	24	10.2
No longer worth living if susceptible/ suicidal	8	8	1	0	1	1	19	8.1
May not want to know genetic status	11	4	3	0	0	0	18	7.7
Issues relating to the accuracy of the test/ No guarantee	4	8	3	2	0	0	17	7.2
Rejection/ genetic discrimination/ treated differently	9	3	2	1	0	0	15	6.4
Playing god	4	8	3	0	0	0	15	6.4
Other psychological	6	3	2	1	0	0	12	5.1
Not being able to plan/ waiting for cancer to occur	3	6	1	0	0	0	10	4.3
Implications for the rest of the family	2	3	3	1	1	0	10	4.3
Wary of health professionals/ counselling	2	4	2	1	0	0	9	3.8
Make patient fell uncomfortable/ not feel oneself	3	3	1	0	0	0	7	3.0
Other biomedical	2	1	2	0	0	0	5	2.1
Become complacent about lifestyle if not susceptible	0	1	0	0	0	0	1	0.4
Sub Total	**105**	**78**	**34**	**13**	**4**	**1**	**235**	**100.0**
Missing	37	64	108	129	138	141	-	-
Total	142	142	142	142	142	142	-	-

Table 5.3

Number of Benefits and Barriers Associated with Testing and Counselling

	N	Mean	Median	Std. dev.	Minimum	Maximum
Baseline						
Number of benefits (pros)	142	3.2	3.0	1.9	0.0	11.0
Number of barriers (cons)	142	1.7	2.0	1.4	0.0	6.0
Follow-up						
Number of benefits (pros)	142	3.0	3.0	1.4	0.0	7.0
Number of barriers (cons)	142	1.7	2.0	1.3	0.0	5.0

Table 5.4

Importance of Benefits and Barriers

	N	Mean	Median	Std. dev.	Minimum	Maximum
Baseline						
Importance of benefits (pros)	129	283.9	250.0	151.4	20.0	1040.0
Importance of barriers (cons)	104	129.6	130.0	82.4	0.0	400.0
Follow-up						
Importance of benefits (pros)	131	260.7	260.0	115.4	50.0	660.0
Importance of barriers (cons)	115	141.4	130.0	82.9	0.0	460.0

At baseline the mean number of benefits of testing and counselling reported was 3.2, with a maximum of 11 benefits reported (see Table 5.3). The Mean number of barriers reported was 1.7 with a maximum of 6. No significant change was seen in the number of benefits or barriers between baseline and follow-up, with a mean of 3 benefits and 1.7 barriers being recorded at follow-up.

The small reduction in the mean number of benefits reported between baseline and follow-up was accompanied by a reduction in the mean of the sum of the importance rating provided by respondents for all the benefits reported (the importance of each benefit and barrier was rated on a 0-100 likert scale). The latter change was accompanied by an increase in the mean sum of the importance ratings of the barriers noted by respondents (see Tables 5.3 and 5.4). These changes were not statistically significant.

Weighted Ratio of Pros to Cons

Hypothesis 1: Experiment information will produce a statistically significant change in the weighted ratio of pros to cons of testing and counselling.

Hypothesis 2: Positive information will increase the pros relative to the cons of genetic testing and counselling recoded by respondents, both the pos-neg (Wroe & Salkovskis, 1999, called this group the negative information group) and the neg-pos information will increase recorded cons relative to the pros, and little or no change will occur for the control group.

Hypothesis 3: Neg-pos information will result in a statistically significantly greater decline in the ratio of pros to cons between assessments (baseline to follow-up) than the pos-neg information (ordering/primacy effect).

Six respondents (4.2%) did not provide data which enables the weighted ratio scores to be calculated at either assessment, two only responded at baseline (1.4%), eight only responded at Follow-up (5.6%) and 126 (88.7%) provided data at both assessments. No statistically significant differences were found between the respondents that provided data for the weighted ratio items at both assessments and those that did not on age, gender, number of respondents that were parents, ethnicity, social class, anxiety, depression or dispositional optimism.

Hypothesis one was tested by means of a repeated measures general linear model (GLM) and the accompanying post hoc tests. The dependent variable change in weighted ratio of pros to cons from baseline to follow-up (post information) was compared by a list of independent variables and the interaction of the independent variables. The independent variables comprised of information group, demographic characteristics (gender, age and social class), psychological characteristics (anxiety [HADS], depression [HADS] and dispositional optimism [LOT]) and the potentially biasing factor of failing the utility theory axiom tests (dominance, transitivity and non-satiation). The independent variables anxiety, depression and dispositional optimism were continuous variables; all other independent variables were nominal.

Examination of the unstandardised residuals revealed one outlier for the baseline data and one outlier and an extreme for the follow-up data (two respondents in total); both residual outliers were excluded. Repeated measures GLM detected a significant effect of time [F(1,118)=16.330, P=0.000], dispositional optimism with time [F(1,118)=7.026, P=0.009] and information group by time [F(3,118)=4.930, P=0.003].

Table 5.5

Estimated Marginal Weighted Ratio Scores Predicted from Dispositional Optimism Scores

	N	**Mean**	**Std. error**	**95% Confidence interval**	
				Lower bound	Upper bound
Weighted ratio score (baseline)	123	0.3967	0.00062	0.3955	0.3980
Weighted ratio score (follow-up)	123	0.3829	0.00680	0.3694	0.3963

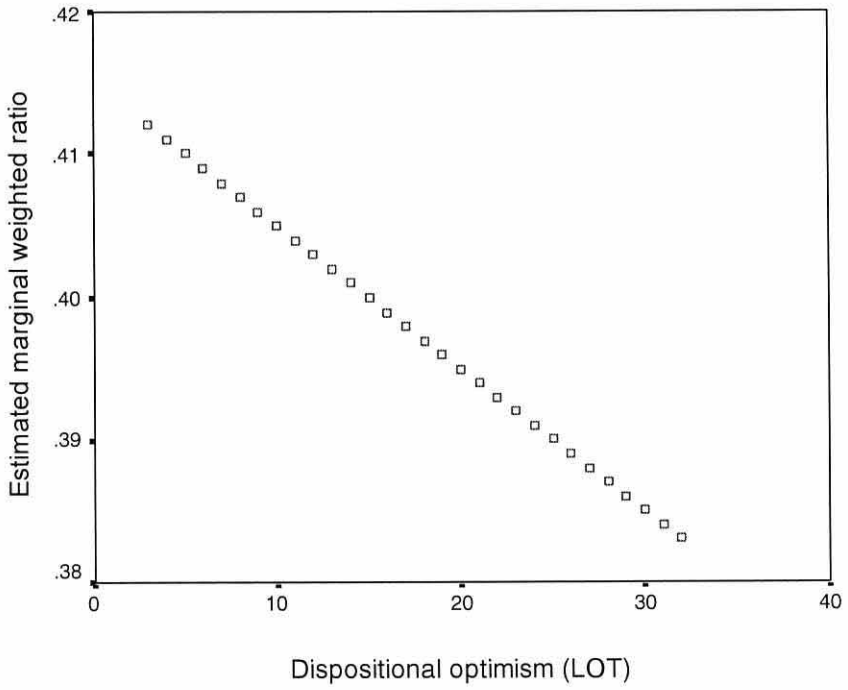


Figure 5.1. Baseline estimated weighted ratio scores predicted from dispositional optimism scores.

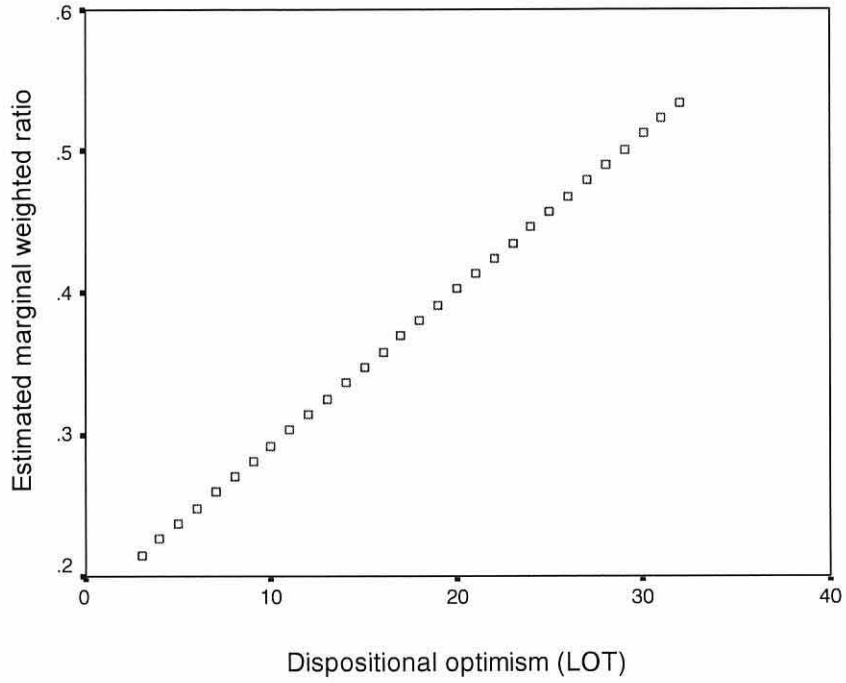


Figure 5.2. Follow-up estimated weighted ratio scores predicted from dispositional optimism scores.

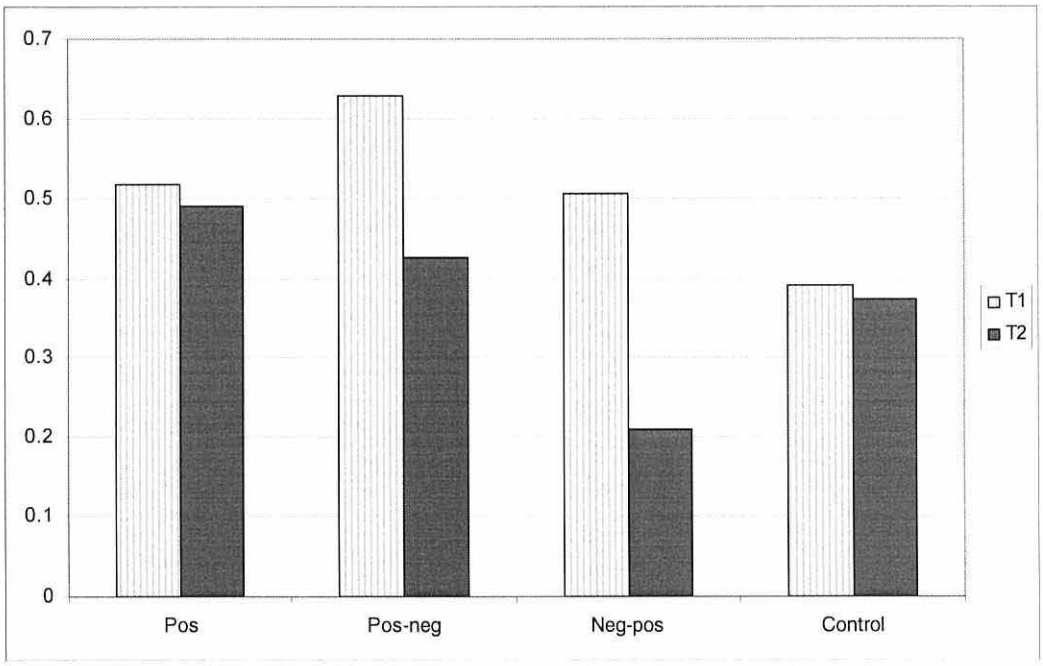
The estimated marginal means associated with dispositional optimism (independent of the other significant variable, information group) declined between assessments (see Table 5.5). The decline signifies a reduction in the importance of benefits (pros) relative to barriers (cons) as a proportion of all the benefits and barriers reported by each respondent. In Figures 5.1 and 5.2 baseline and follow-up estimated marginal weighted ratio scores are displayed against optimism scores as scatterplots. As the estimated marginal weighted ratio scores were estimated from the optimism scores it is no surprise to find perfect linear relationships in Figures 5.1 and 5.2. However, they reveal a complete reversal in the relationship of the variables from baseline to follow-up. At baseline a negative relationship was seen with greater optimism being associated with a lower weighted ratio score and the converse at follow-up with greater optimism being associated with a higher weighted ratio scores (higher = more benefits being cited).

Table 5.6

Estimated Marginal Means of the Weighted Ratio Scores by the Information Groups

Information group	Assessment				95% Confidence interval	
		N	Mean	Std. error	Lower bound	Upper bound
Positive	Baseline	33	.517(a)	.061	.395	.639
	Follow-up	33	.491(a)	.053	.386	.597
Pos-neg	Baseline	32	.628(a)	.063	.504	.753
	Follow-up	32	.426(a)	.055	.317	.534
Neg-pos	Baseline	26	.507(a)	.069	.369	.644
	Follow-up	26	.210(a)	.060	.090	.329
Control	Baseline	32	.391(a)	.062	.267	.514
	Follow-up	32	.375(a)	.054	.268	.483

a = Covariates appearing in the model are evaluated at the following values: Dispositional optimism score = 18.2602.



Key: T1 (vertical hashing) = baseline score, T2 (solid colour) = follow-up score.

Figure 5.3. Estimated marginal means of the weighted ratio scores by information group.

Irrespective of information group, weighted ratio scores declined between the baseline and follow-up assessments (see Table 5.6 and Figure 5.3), mirroring the findings in relation to dispositional optimism. The largest reductions were recorded by the pos-neg and neg-pos groups, the largest change being recorded by the latter information group. Paired post hoc comparisons of the information groups revealed that only the neg-pos and the control groups were statistically

significantly different (LSD test, P=0.025) to each other in terms of change in their weighted ratio scores from baseline to follow-up. The neg-pos group recorded a decline in their weighted ratio scores of -0.297 out of a maximum possible change of ±2.00 whilst the control group only recorded a decline of -0.016.

Behavioural Intention (Intention and Self-prediction) of Having Cancer Genetic Testing and Counselling

Hypothesis 4: Experiment information produces statistically significant change in self-prediction and intention to have testing and counselling.

Hypothesis 5: Positive information will increase self-prediction and intention scores, both pos-neg and neg-pos information will reduce self-prediction and intention scores, and little or no change in scores will occur for the control group.

Hypothesis 6: Behavioural intention (intention and self-prediction) of booking an appointment for genetic testing and counselling for breast cancer will decline between assessments statistically significantly more for the neg-pos than the pos-neg information group (ordering/primacy effect).

The dependent variables change in self-prediction and intention scores from baseline to follow-up (post information) were compared by the same independent variables as were used with the weighted ratio scores: information group, demographic characteristics (gender, age and social class), psychological characteristics (anxiety [HADS], depression [HADS] and dispositional optimism [LOT]) and the potentially biasing factor of failing the utility theory axiom tests (dominance, transitivity and non-satiation).

The repeated measures GLM (repeated measures MANOVA) detected no significant effect upon intention scores by time or time in conjunction with any of the independent variables. A significant interaction effect of time with gender was found upon self-prediction scores [F(1,138)=5.776, P=0.018].

Table 5.7

Estimated Marginal Means for Gender by Time

Measure	Information group	Assessment	N	Mean	Std. Error	95% Confidence Interval	
						Lower Bound	Upper Bound
Self-prediction	Male	Baseline	22	52.500	6.122	40.395	64.605
		Follow-up	22	44.091	5.960	32.307	55.875
	Female	Baseline	118	55.466	2.643	50.239	60.693
		Follow-up	118	58.093	2.573	53.005	63.181
Intention	Male	Baseline	22	5.991	0.693	4.621	7.360
		Follow-up	22	5.836	0.703	4.447	7.225
	Female	Baseline	118	7.307	0.299	6.716	7.899
		Follow-up	118	7.718	0.303	7.118	8.318

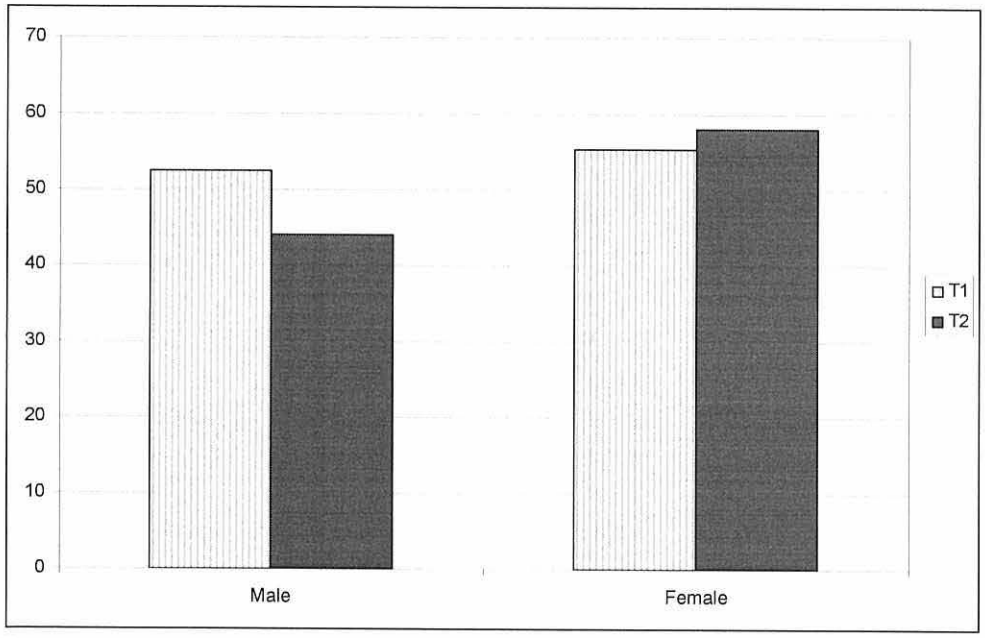


Figure 5.4. Estimated marginal means of self-prediction scores for gender by time.

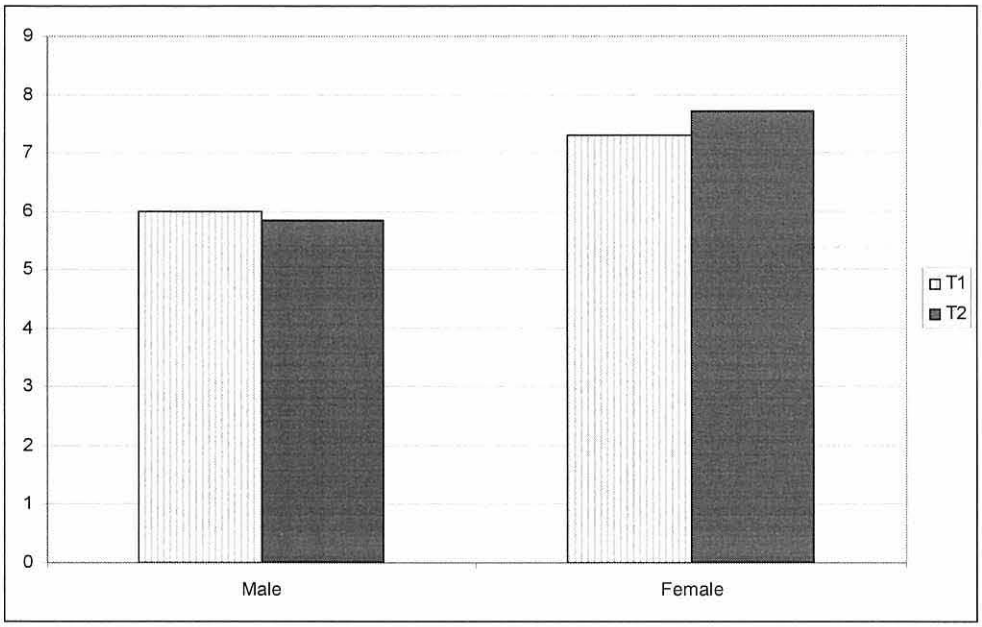


Figure 5.5. Estimated marginal means of intention scores for gender by time.

The descriptive statistics in Table 5.7 and Figure 5.4 reveal that males and females self-prediction of opting to have cancer genetic testing and counselling was completely different. Whilst males were less likely to opt for testing and counselling post information, with self-prediction scores dropping from 52.5 to 44.1, females showed greater self-prediction with scores increasing from 55.5 to 58.1 (scoring range 0-100). Change in mail scores was statistically significant (dependent t test, t=2.422, df=21, p=0.025).

The statistically non-significant results on intention scores mirrored those on self-prediction scores. Intention scores declined for males from 5.99 at baseline to 5.84 at follow-up, whilst female intention scores rose from 7.31 to 7.72 (scoring range 0-14).

Discrete Choices

Baseline Findings

Table 5.8 and 5.9 contain the baseline attribute main effects by information group. A likelihood ratio test comparing the un-segmented baseline model (Table 4.12) with the same baseline model segmented by information group (Tables 5.8 and 5.9) revealed that there was a significant difference (N = 142, χ^2 = 76.744, df

= 45, $p < 0.01$)[1]. There was therefore a significant difference between the preferences of respondents allocated to the information groups before the information was issued to them.

The descriptive statistics in Tables 4.12, 5.8 and 5.9 reveal that the utility functions of the information groups and the resultant coefficients of determination (adjusted R^2) differ substantially with each other and as a result the un-segmented baseline model. For the control group the attribute levels of 80 miles distance to counselling, 1 hour duration of counselling and a cost of £2,500 were statistically non-significant ($p>0.10$). The control group had the lowest explained variance with an adjusted R^2 of 0.28. The neg-pos group had the largest number of statistically non-significant ($p>0.10$) attribute levels at seven; these were: Genetics associate seen for counselling, 20 miles distance to counselling, 80 miles distance to counselling, 1 hour duration of counselling, 1 hour 30 minutes duration of counselling, cost of £2,000 and cost of £2,500. The significant attribute levels explained 34% of the variance (adjusted $R^2=0.34$) in the choices made by the neg-pos group. The pos-neg group had four non-significant attribute levels: 80 miles distance to counselling, 1 hour duration of counselling and both levels of the availability of testing attribute. The pos-neg information group had the largest explained variance in choices at 38% (adjusted $R^2=0.38$). The Positive information group had the smallest number of non-significant attribute levels at three. The attribute levels were: 20 miles distance to counselling, 80 miles distance to counselling and 1 hour 30 minutes duration of counselling. The positive information group had the second largest explained variance in choices between service scenarios at 35% (adjusted $R^2=0.35$). The largest discrepancy in explained variance was between the pos-neg group (adjusted $R^2=0.38$) and the control group (adjusted $R^2=0.28$).

[1] Likelihood ratio test = 2[(-2386.441) - [(-615.9518) + (-569.5630) + (-569.5630) + (-653.2798)]] = -76.744.

Table 5.8
Multinomial Logit Models for Attribute Main Effects by Information Groups at Baseline

		Positive information				Pos-neg information			
Attributes	**Levels**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**
Staff seen	Consultant geneticist	0.840116	0.139688	6.01423	1.80747e-009	1.01314	0.14508	6.98335	2.88236e-012
	Specialist genetics nurse	-0.430664	0.197861	-2.17659	0.0295109	-0.37319	0.170742	-2.1857	0.0288374
	Genetics associate	-0.409452	0.207424	-1.97399	0.0483829	-0.639953	0.198981	-3.21615	0.00129924
Waiting time for letter	1 month	1.82811	0.187527	9.7485	2.88658e-015	1.35703	0.210103	6.45885	1.055e-010
	2 month	0.678813	0.175409	3.86988	0.00010889	1.01185	0.162387	6.23107	4.63245e-010
	4 month	-0.6926	0.193315	-3.58275	0.000339993	-0.926172	0.199302	-4.64708	3.36668e-006
	6 month	-1.81432	0.283566	-6.39823	1.5719e-010	-1.4427	0.305046	-4.72946	2.25113e-006
Distance to counselling	20 miles	0.00309852	0.24534	0.0126295	0.989923	0.694433	0.205543	3.37853	0.000728748
	40 miles	1.09402	0.208232	5.25384	1.48961e-007	0.780316	0.213628	3.65268	0.000259518
	60 miles	-0.92623	0.224768	-4.12083	3.77511e-005	-1.23732	0.216932	-5.70371	1.17227e-008
	80 miles	-0.170884	0.198702	-0.860001	0.389788	-0.237432	0.207125	-1.14632	0.251661
Duration of counselling	30 minutes	-1.19746	0.270667	-4.42412	9.68373e-006	-1.72733	0.270846	-6.37755	1.79944e-010
	1 hour	0.381365	0.210906	1.80822	0.0705719	0.149138	0.201057	0.74177	0.458227
	1 hour 30 minutes	0.265907	0.20707	1.28414	0.199094	0.639699	0.200366	3.19266	0.00140971
	2 hour	0.55019	0.170896	3.21944	0.00128442	0.938495	0.1748	5.36897	7.91891e-008
Availability of testing	High risk	-0.263782	0.126333	-2.08799	0.0367987	0.0486761	0.122269	0.398107	0.690551
	All	0.263782	0.126333	2.08799	0.0367987	-0.0486761	0.122269	-0.398107	0.690551
Cost of service	£1,500	0.780981	0.209408	3.72947	0.000191882	1.38214	0.199216	6.93792	3.97926e-012
	£2,000	0.539751	0.215035	2.51007	0.0120709	0.832429	0.210078	3.96248	7.41755e-005
	£2,500	-0.327142	0.192461	-1.69978	0.0891724	0.422975	0.185379	2.28168	0.0225082
	£3,000	-0.993589	0.246303	-4.03401	5.4834e-005	-2.63755	0.321349	-8.20774	2.88658e-015
	N =38, Number of observations=871, Log likelihood function=-615.9518, Restricted log likelihood (Log-L for Choice model) =-615.9518, R^2=0.35630, Adjusted R^2=0.35071.					N =37, Number of observations=843, Log likelihood function=-569.5630, Restricted log likelihood (Log-L for Choice model) =-569.5630, R^2=0.38501, Adjusted R^2=0.37949.			

Table 5.9

Multinomial Logit Models for Attribute Main Effects by Information Groups at Baseline

		Neg-pos information				Control information			
Attributes	**Levels**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**
Staff seen	Consultant geneticist	1.03594	0.149084	6.94869	3.68705e-012	0.668768	0.131939	5.06876	4.00422e-007
	Specialist genetics nurse	-1.02495	0.208238	-4.922	8.56621e-007	-0.322903	0.152234	-2.12109	0.033914
	Genetics associate	-0.0109886	0.193756	-0.0567136	0.954773	-0.345865	0.168338	-2.05459	0.0399185
Waiting time for letter	1 month	1.27033	0.233931	5.43035	5.62439e-008	1.15478	0.17597	6.56238	5.29548e-011
	2 month	1.1687	0.16543	7.06466	1.61005e-012	0.918429	0.152814	6.01012	1.85382e-009
	4 month	-0.452386	0.208858	-2.166	0.0303115	-0.288871	0.175457	-1.64639	0.0996832
	6 month	-1.98664	0.348772	-5.69611	1.22572e-008	-1.78434	0.269834	-6.61274	3.77276e-011
Distance to counselling	20 miles	0.0311426	0.272019	0.114487	0.908852	0.34726	0.188153	1.84562	0.0649469
	40 miles	1.02021	0.220174	4.63367	3.59236e-006	0.573761	0.18878	3.0393	0.00237126
	60 miles	-0.832653	0.244818	-3.4011	0.000671149	-1.04501	0.207223	-5.04293	4.58451e-007
	80 miles	-0.218703	0.25557	-0.855747	0.392138	0.12399	0.163148	0.759983	0.447265
Duration of counselling	30 minutes	-1.00175	0.316243	-3.16765	0.00153675	-1.31862	0.24757	-5.32625	1.00259e-007
	1 hour	0.116068	0.255898	0.453572	0.650137	0.152366	0.179757	0.847623	0.396648
	1 hour 30 minutes	0.208973	0.232832	0.897525	0.369439	0.73777	0.170944	4.31586	1.5898e-005
	2 hour	0.676708	0.194764	3.47451	0.000511789	0.428486	0.156071	2.74546	0.0060427
Availability of testing	High risk	-0.274452	0.139556	-1.96661	0.0492279	-0.215292	0.11359	-1.89534	0.0580474
	All	0.274452	0.139556	1.96661	0.0492279	0.215292	0.11359	1.89534	0.0580474
Cost of service	£1,500	1.42164	0.204276	6.95939	3.41749e-012	1.01868	0.171363	5.94455	2.77211e-009
	£2,000	0.104856	0.271078	0.38681	0.698897	0.554505	0.190091	2.91705	0.00353356
	£2,500	0.160238	0.231268	0.692868	0.488392	-0.0536122	0.156063	-0.343529	0.731201
	£3,000	-1.68673	0.320436	-5.26387	1.41056e-007	-1.51957	0.232252	-6.54276	6.03952e-011
	N =31, Number of observations=712, Log likelihood function=-509.2744, Restricted log likelihood (Log-L for Choice model) =-509.2744, R^2=0.34893, Adjusted R^2=0.34200.					N =36, Number of observations=828, Log likelihood function=-653.2798, Restricted log likelihood (Log-L for Choice model) =-653.2798, R^2=0.28183, Adjusted R^2=0.27527.			

Repeated Measurement of Discrete Choices

Tables 5.10 to 5.14 show the baseline and follow-up attribute main effects and accompanying statistics for un-segmented and segmented by information group models. There were seven main changes in the *un-segmented model's* utility function from baseline to follow-up (see Table 5.10). Substantial change[2] was seen in the coefficients of the levels, 2 months (approx. Z=2.26), 4 months (approx. Z=2.79) and 6 months (approx. Z=2.57) of the attribute waiting time. At follow-up the coefficient for 2 months waiting time changed from 0.75 to 0.93, whilst the coefficients for 4 months changed from -0.56 to -0.83 and for 6 months from -1.78 to -1.41. These changes show that for the respondents as a whole the utility for a 2 month waiting time increased, dissatisfaction with a 4 month waiting time increased and dissatisfaction with a 6 month waiting time declined. Travelling 80 miles to counselling also changed substantially (approx. Z=2.24), with the coefficient changing from -0.05 to -0.26. The change in the utility of this attribute level to respondents between assessments meant that in the follow-up assessment it was a statistically significant level in the multinomial logit regression; the p value was 0.61 at baseline and 0.006 at follow-up. Two of the levels of the attribute duration of counselling changed at noteworthy levels. The coefficients for 1 hour duration of counselling changed from 0.22 to 0.05; this approached being a substantial change with an approximate Z score of 1.71. The change in the utility of this attribute level (a decline in importance in this case) to respondents between assessments meant that by the follow-up assessment it was a statistically non-significant level in the multinomial logit regression; the p value dropped from 0.03 at baseline and 0.62 at follow-up. A substantial increase in the importance of the level '2 hours duration of counselling' was seen (approx. Z=2.94) with the coefficient increasing from 0.61 to 0.85. Finally, the 'cost of service' attribute level of £1,500 changed substantially; the importance/utility of the lowest charge to respondents declined from 1.10 to 0.85. Explained variance changed very little with an adjusted R^2 of 0.330 at baseline and 0.326 at follow-up.

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[2] Substantial change refers to a change in coefficients yielding an approximate Z score in excess of ±1.96. See Appendix B for a full description of the method.

Table 5.10
Discrete Choice (Multinomial Logit) Model for Attribute Main Effects at Baseline and Follow-up (Un-segmented Model)

		Baseline				Follow-up			
Attributes	**Levels**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**
Staff seen	Consultant geneticist	0.843261	0.0689592	12.2284	2.88658e-015	0.7933	0.0690446	11.4897	2.88658e-015
	Specialist genetics nurse	-0.47997	0.0869375	-5.52086	3.37348e-008	-0.337654	0.0871554	-3.87416	0.000106993
	Genetics associate	-0.363292	0.0934029	-3.88951	0.000100446	-0.455646	0.099984	-4.55719	5.18429e-006
Waiting time for letter	1 month	1.41628	0.0955571	14.8213	2.88658e-015	1.48838	0.0932784	15.9564	2.88658e-015
	2 month	0.927039	0.0801314	11.569	2.88658e-015	0.745886	0.0835103	8.93167	2.88658e-015
	4 month	-0.55868	0.0961259	-5.81196	6.17455e-009	-0.826646	0.0967115	-8.54755	2.88658e-015
	6 month	-1.78464	0.14652	-12.1802	2.88658e-015	-1.40762	0.133291	-10.5605	2.88658e-015
Distance to counselling	20 miles	0.269486	0.108007	2.49508	0.0125928	0.256217	0.106267	2.41106	0.0159061
	40 miles	0.82404	0.10167	8.10502	2.88658e-015	0.937229	0.0977692	9.58614	2.88658e-015
	60 miles	-1.04442	0.109689	-9.5217	2.88658e-015	-0.930949	0.107536	-8.6571	2.88658e-015
	80 miles	-0.0491044	0.0950789	-0.516459	0.605534	-0.262496	0.0949955	-2.76325	0.0057229
Duration of counselling	30 minutes	-1.3567	0.13322	-10.1839	2.88658e-015	-1.45469	0.130877	-11.1149	2.88658e-015
	1 hour	0.224441	0.101384	2.21377	0.0268448	0.050573	0.102962	0.491182	0.623298
	1 hour 30 minutes	0.526961	0.0959232	5.49358	3.93876e-008	0.552979	0.094603	5.84525	5.05793e-009
	2 hour	0.6053	0.0835327	7.24626	4.28546e-013	0.851138	0.0796432	10.6869	2.88658e-015
Availability of testing	High risk	-0.151346	0.0607661	-2.49063	0.0127518	-0.102183	0.0598215	-1.70814	0.0876106
	All	0.151346	0.0607661	2.49063	0.0127518	0.102183	0.0598215	1.70814	0.0876106
Cost of service	£1,500	1.10392	0.0945193	11.6793	2.88658e-015	0.845906	0.102492	8.25338	2.88658e-015
	£2,000	0.525168	0.105662	4.97025	6.68657e-007	0.520194	0.106374	4.89023	1.00718e-006
	£2,500	0.0240341	0.0889284	0.270263	0.786958	0.128568	0.0863989	1.48808	0.13673
	£3,000	-1.65312	0.132718	-12.4559	2.88658e-015	-1.49467	0.133384	-11.2058	2.88658e-015
N = 142, Number of observations=3254, Log likelihood function=-2386.441, Restricted log likelihood (Log-L for Choice model) = -2386.4407, R^2=0.33244, Adjusted R^2=0.33090.						N = 142, Number of observations=3254, Log likelihood function=-2404.581, Restricted log likelihood (Log-L for Choice model) =-2404.5813, R^2=0.32737, Adjusted R^2=0.32581.			

Table 5.11

Multinomial Logit Models for Attribute Main Effects for the Positive Information Group

		Baseline				Follow-up			
Attributes	**Levels**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**
Staff seen	Consultant geneticist	0.840116	0.139688	6.01423	1.80747e-009	0.825921	0.151389	5.45563	4.87988e-008
	Specialist genetics nurse	-0.430664	0.197861	-2.17659	0.0295109	-0.456125	0.210603	-2.1658	0.0303265
	Genetics associate	-0.409452	0.207424	-1.97399	0.0483829	-0.369796	0.230015	-1.6077	0.107901
Waiting time for letter	1 month	1.82811	0.187527	9.7485	2.88658e-015	2.01591	0.191055	10.5515	2.88658e-015
	2 month	0.678813	0.175409	3.86988	0.00010889	0.777445	0.194841	3.99015	6.60314e-005
	4 month	-0.6926	0.193315	-3.58275	0.000339993	-0.91461	0.20067	-4.55777	5.16994e-006
	6 month	-1.81432	0.283566	-6.39823	1.5719e-010	-1.87875	0.294949	-6.36973	1.8936e-010
Distance to counselling	20 miles	0.00309852	0.24534	0.0126295	0.989923	-0.060012	0.261181	-0.229772	0.818269
	40 miles	1.09402	0.208232	5.25384	1.48961e-007	1.1925	0.223242	5.34172	9.20698e-008
	60 miles	-0.92623	0.224768	-4.12083	3.77511e-005	-1.08667	0.242566	-4.47991	7.46735e-006
	80 miles	-0.170884	0.198702	-0.860001	0.389788	-0.0458132	0.212062	-0.216036	0.828959
Duration of counselling	30 minutes	-1.19746	0.270667	-4.42412	9.68373e-006	-2.04736	0.296731	-6.89972	5.21072e-012
	1 hour	0.381365	0.210906	1.80822	0.0705719	0.429302	0.227513	1.88693	0.0591692
	1 hour 30 minutes	0.265907	0.20707	1.28414	0.199094	0.682802	0.199469	3.4231	0.000619107
	2 hour	0.55019	0.170896	3.21944	0.00128442	0.935258	0.170936	5.4714	4.46496e-008
Availability of testing	High risk	-0.263782	0.126333	-2.08799	0.0367987	-0.168772	0.126569	-1.33344	0.182388
	All	0.263782	0.126333	2.08799	0.0367987	0.168772	0.126569	1.33344	0.182388
Cost of service	£1,500	0.780981	0.209408	3.72947	0.000191882	0.678172	0.237486	2.85563	0.0042951
	£2,000	0.539751	0.215035	2.51007	0.0120709	0.416063	0.23809	1.7475	0.0805504
	£2,500	-0.327142	0.192461	-1.69978	0.0891724	-0.195032	0.20163	-0.967277	0.333406
	£3,000	-0.993589	0.246303	-4.03401	5.4834e-005	-0.899202	0.245613	-3.66105	0.000251181
	N =38, Number of observations=871, Log likelihood function=-615.9518, Restricted log likelihood (Log-L for Choice model) =-615.9518, R^2=0.35630, Adjusted R^2=0.35071.					N =38, Number of observations=871, Log likelihood function=-568.2159, Restricted log likelihood (Log-L for Choice model) =-568.2159, R2=0.40619, Adjusted R^2=0.40103.			

Table 5.12

Multinomial Logit Models for Attribute Main Effects for the Pos-neg Information Group

		Baseline				Follow-up			
Attributes	**Levels**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**
Staff seen	Consultant geneticist	1.01314	0.14508	6.98335	2.88236e-012	0.682903	0.134224	5.08778	3.62289e-007
	Specialist genetics nurse	-0.37319	0.170742	-2.1857	0.0288374	-0.407754	0.158287	-2.57604	0.009994
	Genetics associate	-0.639953	0.198981	-3.21615	0.00129924	-0.275149	0.179789	-1.5304	0.125918
Waiting time for letter	1 month	1.35703	0.210103	6.45885	1.055e-010	1.13147	0.195319	5.79296	6.91568e-009
	2 month	1.01185	0.162387	6.23107	4.63245e-010	0.885168	0.151891	5.82766	5.62098e-009
	4 month	-0.926172	0.199302	-4.64708	3.36668e-006	-0.943548	0.191234	-4.934	8.05605e-007
	6 month	-1.4427	0.305046	-4.72946	2.25113e-006	-1.07309	0.239376	-4.48287	7.36449e-006
Distance to counselling	20 miles	0.694433	0.205543	3.37853	0.000728748	0.403633	0.196804	2.05094	0.0402731
	40 miles	0.780316	0.213628	3.65268	0.000259518	1.0074	0.181909	5.53791	3.06097e-008
	60 miles	-1.23732	0.216932	-5.70371	1.17227e-008	-0.700125	0.191588	-3.65433	0.000257857
	80 miles	-0.237432	0.207125	-1.14632	0.251661	-0.710907	0.190813	-3.72567	0.000194801
Duration of counselling	30 minutes	-1.72733	0.270846	-6.37755	1.79944e-010	-1.26003	0.249014	-5.06009	4.19065e-007
	1 hour	0.149138	0.201057	0.74177	0.458227	-0.332877	0.20907	-1.59218	0.111343
	1 hour 30 minutes	0.639699	0.200366	3.19266	0.00140971	0.437456	0.18593	2.3528	0.0186328
	2 hour	0.938495	0.1748	5.36897	7.91891e-008	1.15546	0.154404	7.4833	7.23865e-014
Availability of testing	High risk	0.0486761	0.122269	0.398107	0.690551	-0.0215776	0.116601	-0.185056	0.853185
	All	-0.0486761	0.122269	-0.398107	0.690551	0.0215776	0.116601	0.185056	0.853185
Cost of service	£1,500	1.38214	0.199216	6.93792	3.97926e-012	1.10521	0.202002	5.47126	4.46835e-008
	£2,000	0.832429	0.210078	3.96248	7.41755e-005	0.447777	0.219641	2.03868	0.0414823
	£2,500	0.422975	0.185379	2.28168	0.0225082	0.365936	0.164161	2.22913	0.0258053
	£3,000	-2.63755	0.321349	-8.20774	2.88658e-015	-1.91892	0.296164	-6.47925	9.21809e-011
	N =37, Number of observations=843, Log likelihood function=-569.5630, Restricted log likelihood (Log-L for Choice model) =-569.5630, R^2=0.38501, Adjusted R^2=0.37949.					N =37, Number of observations=843, Log likelihood function=-640.7432, Restricted log likelihood (Log-L for Choice model) =-640.7432, R^2=0.30815, Adjusted R^2=0.30194.			

Table 5.13

Multinomial Logit Models for Attribute Main Effects for the Neg-pos Information Group

		Baseline				Follow-up			
Attributes	**Levels**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**
Staff seen	Consultant geneticist	1.03594	0.149084	6.94869	3.68705e-012	0.918515	0.145952	6.29327	3.10849e-010
	Specialist genetics nurse	-1.02495	0.208238	-4.922	8.56621e-007	-0.376984	0.188423	-2.00073	0.0454215
	Genetics associate	-0.0109886	0.193756	-0.0567136	0.954773	-0.54153	0.214005	-2.53046	0.0113914
Waiting time for letter	1 month	1.27033	0.233931	5.43035	5.62439e-008	1.55636	0.193455	8.04509	2.88658e-015
	2 month	1.1687	0.16543	7.06466	1.61005e-012	0.662238	0.184183	3.59555	0.000323704
	4 month	-0.452386	0.208858	-2.166	0.0303115	-0.685301	0.19279	-3.55465	0.000378488
	6 month	-1.98664	0.348772	-5.69611	1.22572e-008	-1.5333	0.275719	-5.5611	2.68073e-008
Distance to counselling	20 miles	0.0311426	0.272019	0.114487	0.908852	0.0862516	0.229889	0.375188	0.707521
	40 miles	1.02021	0.220174	4.63367	3.59236e-006	1.02196	0.204883	4.98801	6.10037e-007
	60 miles	-0.832653	0.244818	-3.4011	0.000671149	-1.20552	0.23438	-5.14346	2.69727e-007
	80 miles	-0.218703	0.25557	-0.855747	0.392138	0.0973113	0.194146	0.501227	0.616211
Duration of counselling	30 minutes	-1.00175	0.316243	-3.16765	0.00153675	-1.83131	0.278022	-6.58693	4.49025e-011
	1 hour	0.116068	0.255898	0.453572	0.650137	0.268378	0.20885	1.28503	0.198781
	1 hour 30 minutes	0.208973	0.232832	0.897525	0.369439	0.683492	0.193134	3.53895	0.000401729
	2 hour	0.676708	0.194764	3.47451	0.000511789	0.879441	0.166215	5.29099	1.21659e-007
Availability of testing	High risk	-0.274452	0.139556	-1.96661	0.0492279	-0.0648957	0.120631	-0.537971	0.590597
	All	0.274452	0.139556	1.96661	0.0492279	0.0648957	0.120631	0.537971	0.590597
Cost of service	£1,500	1.42164	0.204276	6.95939	3.41749e-012	0.753304	0.216869	3.47354	0.000513634
	£2,000	0.104856	0.271078	0.38681	0.698897	0.720972	0.205116	3.51494	0.000439846
	£2,500	0.160238	0.231268	0.692868	0.488392	0.0219884	0.181164	0.121373	0.903395
	£3,000	-1.68673	0.320436	-5.26387	1.41056e-007	-1.49626	0.262507	-5.6999	1.19877e-008
	N =31, Number of observations=712, Log likelihood function=-509.2744, Restricted log likelihood (Log-L for Choice model) =-509.2744, R^2=0.34893, Adjusted R^2=0.34200.					N =31, Number of observations=712, Log likelihood function=-531.9509, Restricted log likelihood (Log-L for Choice model) =-531.9509, R^2=0.31994, Adjusted R^2=0.31270.			

Table 5.14

Multinomial Logit Models for Attribute Main Effects for the Control Information Group

		Baseline				Follow-up			
Attributes	**Levels**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**	**Coefficients (B)**	**Standard error**	**t-ratio**	**P value**
Staff seen	Consultant geneticist	0.668768	0.131939	5.06876	4.00422e-007	0.894875	0.134741	6.64143	3.10649e-011
	Specialist genetics nurse	-0.322903	0.152234	-2.12109	0.033914	-0.314931	0.166977	-1.88607	0.0592855
	Genetics associate	-0.345865	0.168338	-2.05459	0.0399185	-0.579944	0.187449	-3.09387	0.00197561
Waiting time for letter	1 month	1.15478	0.17597	6.56238	5.29548e-011	1.27712	0.183781	6.94914	3.67506e-012
	2 month	0.918429	0.152814	6.01012	1.85382e-009	0.783769	0.155538	5.03908	4.67781e-007
	4 month	-0.288871	0.175457	-1.64639	0.0996832	-0.810784	0.195437	-4.14857	3.34565e-005
	6 month	-1.78434	0.269834	-6.61274	3.77276e-011	-1.25011	0.263215	-4.74938	2.04039e-006
Distance to counselling	20 miles	0.34726	0.188153	1.84562	0.0649469	0.572102	0.188671	3.03227	0.00242724
	40 miles	0.573761	0.18878	3.0393	0.00237126	0.649184	0.190097	3.41501	0.000637792
	60 miles	-1.04501	0.207223	-5.04293	4.58451e-007	-0.819361	0.21062	-3.89024	0.000100145
	80 miles	0.12399	0.163148	0.759983	0.447265	-0.401925	0.188592	-2.13119	0.0330733
Duration of counselling	30 minutes	-1.31862	0.24757	-5.32625	1.00259e-007	-0.818403	0.230866	-3.54493	0.000392715
	1 hour	0.152366	0.179757	0.847623	0.396648	-0.0783525	0.189508	-0.413453	0.679275
	1 hour 30 minutes	0.73777	0.170944	4.31586	1.5898e-005	0.345515	0.19565	1.76599	0.0773979
	2 hour	0.428486	0.156071	2.74546	0.0060427	0.551241	0.164966	3.34154	0.000833157
Availability of testing	High risk	-0.215292	0.11359	-1.89534	0.0580474	-0.195219	0.120782	-1.61629	0.106031
	All	0.215292	0.11359	1.89534	0.0580474	0.195219	0.120782	1.61629	0.106031
Cost of service	£1,500	1.01868	0.171363	5.94455	2.77211e-009	0.988328	0.190515	5.18766	2.12952e-007
	£2,000	0.554505	0.190091	2.91705	0.00353356	0.60131	0.210502	2.85656	0.00428263
	£2,500	-0.0536122	0.156063	-0.343529	0.731201	0.304402	0.168912	1.80213	0.0715254
	£3,000	-1.51957	0.232252	-6.54276	6.03952e-011	-1.89404	0.28948	-6.54291	6.03331e-011
	N =36, Number of observations=828, Log likelihood function=-653.2798, Restricted log likelihood (Log-L for Choice model) =-653.2798, R^2=0.28183, Adjusted R^2=0.27527.					N =36, Number of observations=828, Log likelihood function=-627.4951, Restricted log likelihood (Log-L for Choice model) =-627.4951, R^2=0.31018, Adjusted R^2=0.30387.			

There were six main changes from baseline to follow-up in the utility function of the *positive information group* (see Table 5.11). On the attribute staff seen, for the level 'Genetics Associate' a small change in the utility coefficient was seen, a change from -0.41 to -0.37 (approx. Z=-0.19). This change resulted in the attribute changing from being a significant (p=0.048) variable in the baseline utility function (statistically significant variable in the baseline multinomial logit regression p<0.1) to approaching significance (p=0.11) in the follow-up assessment. Substantial change was seen in the coefficients of three of the duration of counselling levels. The coefficient for 30 minutes duration of counselling changed from -1.2 to -2.05 (approx. Z=3.14). The change in the attribute levels 1 hour 30 minutes and 2 hours duration of counselling were consistent, with a substantial increase in the coefficients of both levels, signifying increased utility from the two longest counselling sessions. The changes in coefficients were respectively 0.27 to 0.68 (approx. Z=2.01) and 0.55 to 0.94 (approx. Z=2.25). In addition, the change in the utility of the attribute level '1 hour 30 minutes duration of counselling' resulted in the level being a significant level in the respondents' utility function in the follow-up assessment; p=0.199 at baseline and p=0.0006 at follow-up. Relatively small changes were seen in the dichotomous levels of the attribute 'availability of testing' (±0.26 to ±0.17, approx. Z=0.75); however, this resulted in the attribute being non-significant in the utility function of respondents at follow-up (baseline p=0.04, follow-up p=0.18). Explained variance in choices changed sharply with the adjusted R^2 increasing from 0.35 to 0.40.

Five key changes were seen over time for the *pos-neg group* (see Table 5.12). Whilst there was little to no change in the coefficient for the attribute level of seeing a Specialist Genetics Nurse for counselling, the remaining two levels did show change. A substantial reduction was seen in the utility coefficient for seeing a Consultant (approx. Z=2.27), changing from 1.01 to 0.68. The change in the dissatisfaction with seeing a Genetics Associate declined from -0.64 to -0.28. This change approached a substantial change with an approximate Z value of 1.83. The change in the coefficient was accompanied by the level changing from being a significant level (p=0.001) in the baseline utility function to being non-significant/approaching significance (p=0.13) in the follow-up. Substantial

change was seen in the coefficients of the two largest levels of the attribute 'distance to counselling'. The dissatisfaction with travelling 60 miles to counselling declined, the coefficient declined from -1.24 to -0.70 (approx. Z=2.48). The dissatisfaction with travelling 80 miles increased from -0.24 to -0.71 (approx. Z=2.29), making it less desirable than travelling 60 miles. The change resulted in this level being a significant level in the follow-up utility function (p=0.0002). Finally, the dissatisfaction with the most expensive charge of £3,000 declined from -2.64 to -1.92 (approx. Z=2.24). Explained variance dropped dramatically from baseline to follow-up, with the adjusted R^2 dropping from 0.38 to 0.30.

Nine major changes were seen in the discrete choices made by the *neg-pos group* (see Table 5.13). A substantial reduction in the dissatisfaction of seeing a Genetics Nurse was seen, coefficients declined from -1.02 to -0.38 (approx. Z=3.11). The dissatisfaction with seeing a Genetics Associate increased from -0.01 to -0.54 (approx. Z=2.74). Genetics Associate became a significant attribute in the utility function of the neg-pos group in the follow-up assessment (p=0.011). A substantial reduction was seen in the utility of having a 2 month wait (approx. Z=3.06), with the coefficient for this level declining from 1.17 to 0.66. Dissatisfaction with a 30 minute consultation increased substantially between assessments (approx. Z=2.62), the coefficient changed from -1.00 to -1.83. The utility of the second highest consultation time, 1 hour 30 minutes, increased substantially (approx. Z=2.04) from 0.21 to 0.68. This increase in the preference/utility for this level was accompanied by it becoming a significant level in the neg-pos group's utility function at follow-up (p=0.0004). Although change in the dichotomous levels of the attribute 'availability of testing' were not substantial, changing from (±0.27 to ±0.06, approx. Z=1. 50) this change resulted in the attribute changing to non-significant (p=0.59) from significant (p=0.049). A substantial reduction in the utility associated with the lowest charge of £1,500 (approx. Z=3.27) was seen as the coefficient changed from 1.42 to 0.75. Finally, a substantial increase (Z=2.27) was seen in the utility associated with the second lowest charge of £2,000; the coefficient changed from 0.10 to 0.72. The increased utility associated with the £2,000 charge was reflected in it being a

significant level in the follow-up utility function (p=0.0004). Explained variance declined from 0.34 to 0.31 (adjusted R^2).

Eight major changes were seen in the utility function of the *control group* from baseline to follow-up (see Table 5.14). Dissatisfaction with waiting 4 months for a letter confirming risk status increased substantially (approx. Z=2.97) from baseline to follow-up; the coefficient changing from -0.29 to -0.81. Dissatisfaction declined substantially (approx. Z=1.98) between assessments with having to wait 6 months for a letter (baseline coefficient=-1.78, follow- up coefficient=-1.25). The coefficient associated with the level 'travelling 80 miles for counselling' changes from being a positive non-significant (p=0.45) value in the baseline to a significant coefficient (p=0.033) in the follow-up with a negative sign (baseline coefficient=0.12, follow- up coefficient=-0.40, approx. Z=3.22). Dissatisfaction with a counselling session of only 30 minutes declined substantially (approx. Z=2.02) between assessments, changing from -1.31 to -0.82. The positive utility or satisfaction associated with a counselling session of 1 hour 30 minutes declined (approx. Z=2.29), with the coefficient declining from 0.74 to 0.35. Change in the dichotomous levels of the attribute 'availability of testing' were small, changing from (±0.22 to ±0.20, approx. Z=0.18), however, this change resulted in the attribute changing from significant (p=0.058) to approaching significance (p=0.11). Finally, the attribute level of 'a charge of £2,500' changed substantially (approx. Z=2.29), the utility associated with this level increased from -0.05 to 0.30. The level '£2,500' emerged as a statistically significant (p=0.07) level in the follow-up assessment. Explained variance rose by 2%, with adjusted R^2 rising from 0.28 to 0.30.

The Relationship between Benefits and Barriers and Behavioural Intention to Have Testing

Hypothesis 7: A large degree of positive correlation (0.5 – 1.0) will exist between weighted ratio scores and self-prediction and intention to have testing scores.

The relationship between weighted ratio scores and the self-prediction and intention scores at baseline were weak with small correlation coefficients (see Tables 5.15 and 5.16 and Figures 5.6 and 5.8). By the follow-up assessment the degree of agreement between these measures had improved (see Table 5.16 and Figures 5.7 and 5.9), however, the magnitude of the correlation coefficients were only medium at 0.34 to 0.40.

Table 5.15

Descriptive Statistics for Weighted Ratio, Self-prediction and Intention Scores

	Baseline			Follow-up		
	N	**Mean**	**Std. Deviation**	**N**	**Mean**	**Std. Deviation**
Weighted ratio score	123	.5109	.35760	123	.3844	.32776
Self-prediction rating	123	54.6341	28.88055	123	55.6504	28.50050
Intention score	122	7.0475	3.30286	123	7.3911	3.42759

The two respondents excluded from the analysis of self-prediction and intention as their residuals were identified as extreme have been excluded from this analysis.

Table 5.16

Correlation of Weighted Ratio by Self-prediction and Intention

	Self-prediction		Intention	
	Baseline	**Follow-up**	**Baseline**	**Follow-up**
Pearson Correlation	0.191	0.404	0.242	0.343
Sig. (2-tailed)	0.034	0.000	0.007	0.000
N	123	123	122	123

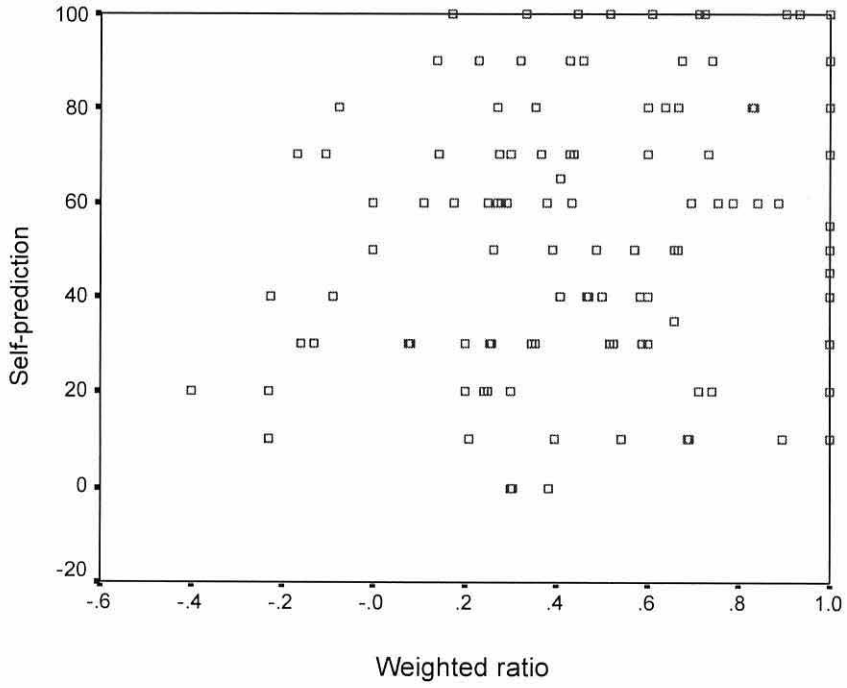


Figure 5.6. Baseline weighted ratios scores by self-prediction ratings.

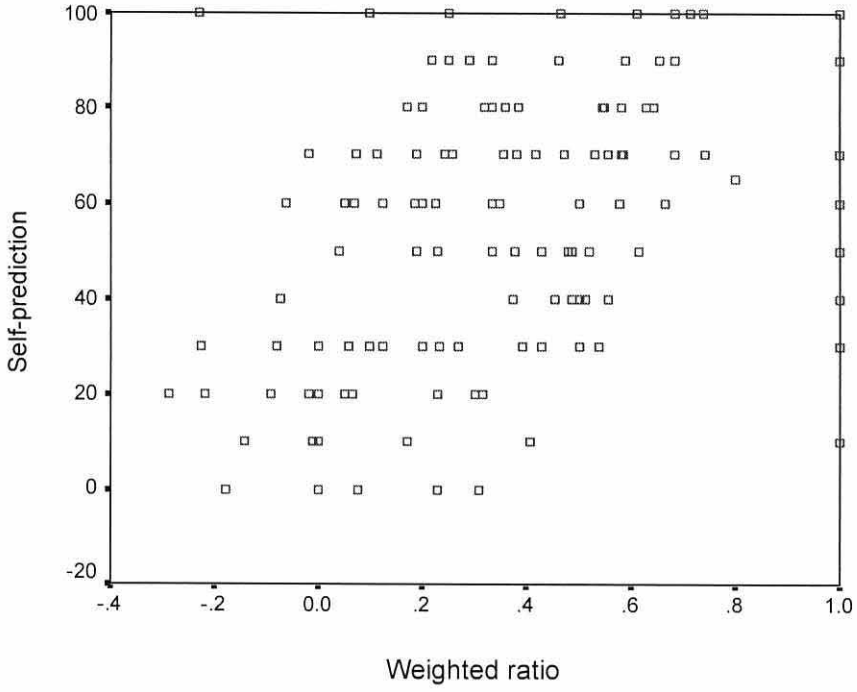


Figure 5.7. Follow-up weighted ratios scores by self-prediction ratings.

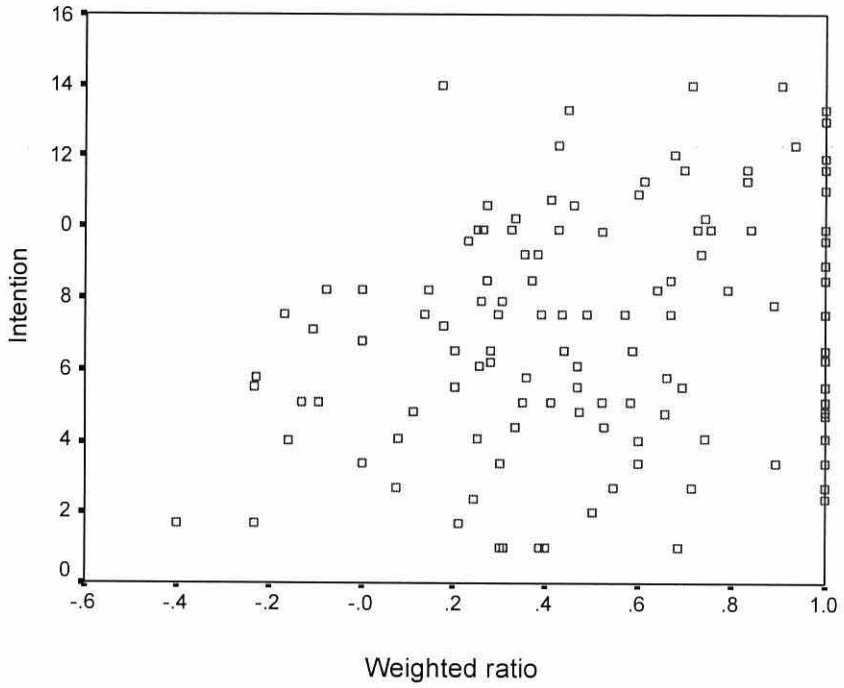


Figure 5.8. Baseline weighted ratios by intention scores.

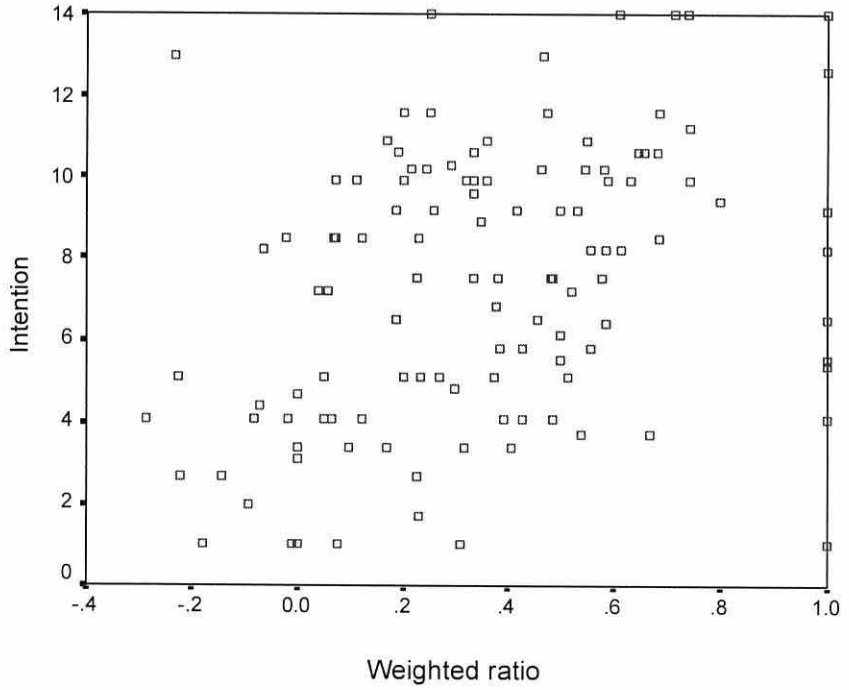


Figure 5.9. Follow-up weighted ratios by intention scores.

Change in Knowledge and Health Perceptions Following the Information Manipulation

To ascertain if the statistically significant change in self-prediction of having genetic testing and counselling was accompanied by changes in health perceptions/cognitions and knowledge following the information manipulation, repeated measures GLM analysis was conducted[3]. The knowledge items were knowledge of; cancer, breast cancer, non-cancer genetic testing, genetic testing for cancer and genetic testing for beast cancer. The health perception/cognition items were cancer worry, coping, anxiety about risk of developing cancer and anxiety about having genetic testing. Social cognition variables utilised in the TPB and HBM were not included in this analysis as the TPB and HBM models are continuum models where specific relationships between variables are assumed to exist. The same set of independent variables as were used to look at changes in self-prediction and intention scores were used with knowledge and health perception/cognition measures.

All statistically significant results are reported in Tables 5.17 to 5.18. As the purpose of this analysis is to attempt to explain how the experiment induced changes in health perceptions/cognitions that lead to significant change in self-prediction scores, the findings of interest are any Knowledge and/or health cognitions explained by the same predictor variables, namely time by gender.

Time by gender was only found to be a statistically significant independent interaction variable for the health cognition item, anxiety about the risk of developing cancer [$F(1,136)=12.631$, $P=0.001$] on Table 5.18. The descriptive statistics in Table 5.19 and Figure 5.10 Show that males and females reacted differently. Whilst females showed a 3 point increase in mean anxiety, males recorded a mean decrease of 19 points.

[3] Wroe and Salkovskis (1999) also utilised this form of testing in their experiment.

Table 5.17

Change in Knowledge Items Between Assessments

		Knowledge of:				
		Cancer	**Breast cancer**	**Non-cancer genetic testing**	**Genetic testing for cancer**	**Genetic testing for beast cancer**
	Type of GLM used	Repeated	#Univariate	Repeated	Repeated	Repeated
	No of excluded respondents due to extreme residuals	1	7	3	3	0
Independent variables						
Time		[F(1,113)=4.891, P=0.029]	-	[F(1,128)=18.305, P=0.000]	[F(1,137)=33.023, P=0.000]	-
Time*information group*sex		-	[F(3,108)=5.147, P=0.002]	[F(3,128)=2.979, P=0.034]	-	-
Time*information group*social class		[F(6,113)=2.4143, P=0.031]	[F(6,108)=4.401, P=0.001]	-	-	-
Time*information group*sex*social class		[F(2,113)=3.337, P=0.039]	[F(2,108)=6.896, P=0.002]	-	-	-

Baseline results found to be statistically significantly different by gender. Baseline scores were subtracted from the follow-up scores and univariate GLM was conducted on the resulting measure. If gender was not a significant independent variable repeated measures GLM was conducted.

Table 5.18

Change in Worry, Ability to Cope and Trait Anxiety Items Between Assessments

		Cancer worry	**Coping**	**Anxiety about risk of developing cancer**	**Anxiety about having genetic testing**
	Type of GLM used	# Repeated	#Univariate	Repeated	#Repeated
	No of excluded respondents due to extreme residuals	0	0	0	0
Independent variables					
Time		-	-	[F(1,136)=6.066, P=0.015]	-
Time*sex		-	-	[F(1,136)=12.631, P=0.001]	-
Time*axiom tests		[F(1, 140)=5.304, P=0.023]	-	[F(1,136)=8.969, P=0.003]	-

Baseline results for found to be statistically significantly different by gender. Baseline scores were subtracted from the follow-up scores and univariate GLM was conducted on the resulting measure. If gender was not a significant independent variable repeated measures GLM was conducted.

Table 5.19

Estimated Marginal Means of Anxiety About Risk of Developing Cancer for Gender by Time

Information group	Assessment	N	Mean	Std. Error	95% Confidence Interval	
					Lower Bound	Upper Bound
Male	Baseline	23	54.667	7.942	38.960	70.373
	Follow-up	23	35.833	7.317	21.364	50.302
Female	Baseline	117	47.737	3.150	41.507	53.967
	Follow-up	117	51.152	2.902	45.413	56.891

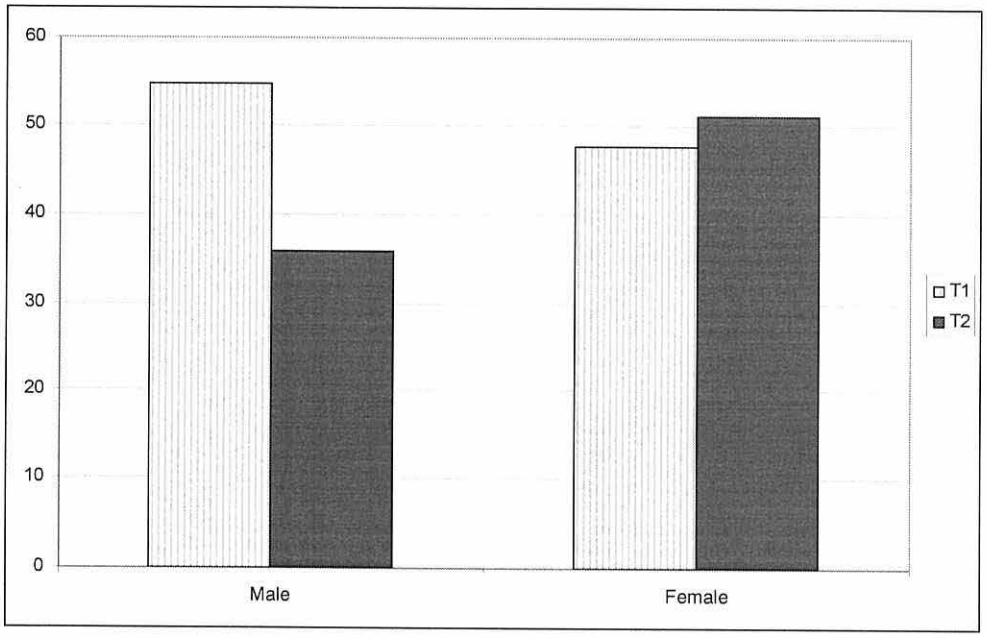
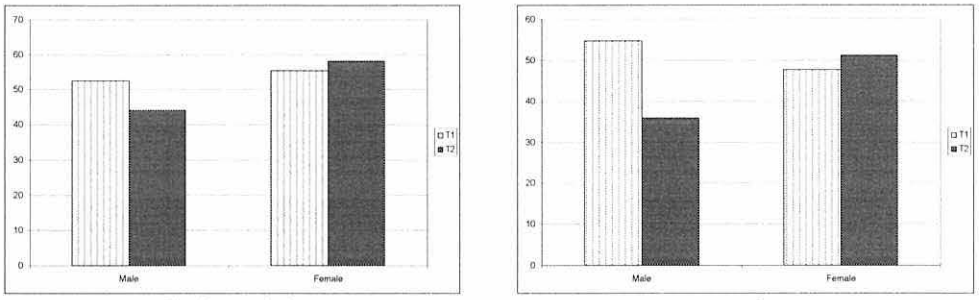


Figure 5.10. Estimated Marginal Means of Anxiety About Risk of Developing Cancer for Gender by Time

Upon comparison of the descriptive statistics for anxiety about the risk of developing cancer with those for self-prediction scores a consistent pattern was found. The results for anxiety (see Table 5.19) mirrored those recorded for the self-prediction of booking a genetic testing and counselling appointment (see Table 5.7); with the females recording an increase in anxiety and males recording a decline in anxiety (see Figure 5.11). Change in female scores was statistically significant (dependent t test, t=-2.597, df=116, p=0.011).



Self-prediction **Anxiety**

Key: T1 (vertical hashing) = baseline score, T2 (solid colour) = follow-up score.

Figure 5.11. Self-prediction of booking a genetic testing appointment and anxiety about having genetic testing by information group.

Alternatives to Random Utility Theory

As hypotheses 2[4] and 7[5] were rejected, and as a result evidence of respondents making decisions in accordance with random utility theory was not fond, objective 6[6] was not assessed. The analyses of components of the TPB and HBM as predictors of behavioural intention were extended to include other cognitions and emotions identified as potentially relevant independent variables. All variables utilised in the analysis as independent variables are listed in Table 5.20.

As the health belief model and theory of planned behaviour are continuum models where specific relationships between independent variables that are believed to influence an individual's health behaviour are assumed to exist, it has been necessary to use regression analysis rather than repeated measures GLM to accommodate this aspect of these theories. Multiple linear regression analysis with sequential (hierarchical) inclusion, and removal of non-significant independent variables by the backwards elimination method was used (see methods section) upon the two dependent variables self-prediction and intention

[4] Hypothesis 2: Positive information will increase the pros relative to the cons of genetic testing and counselling recoded by respondents, both the pos-neg (Wroe & Salkovskis, 1999, called this group the negative information group) and the neg-pos information will increase recorded cons relative to the pros, and little or no change will occur for the control group.

[5] Hypothesis 7: A large degree of positive correlation (0.5 – 1.0) will exist between weighted ratio scores and self-prediction and intention to have testing scores.

[6] *(If respondents are adhering to RUT)*Ascertain which components of the TPB and HBM are significant predictors of behavioural intention.

to have genetic testing and counselling[7]. The entry order of independent variables is displayed in Table 5.20.[8] Backward elimination of the non-significant variables and the change in the explained variance of the dependent variables at each stage is presented in Tables 5.21 and 5.23. Tables 5.22 and 5.24 show the results from the final regression models (parsimonious specification of independent variables).

Table 5.20

Independent Variables and their Block Entry Order

Block 1
Baseline score (self-prediction for self-prediction change and intention for intention change)
Information group (3 dummy coded variables)
Block 2
Sex (1 dummy coded variable)
Social class (2 dummy coded variables)
Age
Block 3
Dispositional optimism (LOT)
Block 4
HADS anxiety
HADS depression
Block 5
Perceived control (TPB)
Subjective norm (TPB)
Attitudes towards behaviour (TPB)
Susceptibility (HBM)
Severity (HBM)
Weighted ratio (Random utility theory & HBM)
Cancer worry
Coping
Anxiety – risk of developing breast cancer
Anxiety – having genetic testing
Block 6
Failing the utility theory axiom tests

[7] Change in likelihood scores (t2 – t1) had 13 extreme values (9 with a reduction of 40 points or more and 4 with an increase of 40 points or more. These extremes were removed as was one extreme residual found in the initial regression analysis. Change in interest scores had 9 extreme scores (6 reduced their score by 4.8 points or more and 3 increased their scores by 5.4 points or more). 1 extremes residual was found in the initial regression analysis. All extreme values were removed from the final regression analyses.

[8] McGregor, Bowen, Ankerst, Andersen, Yasui and McTiernan (2004) have found that perceived risk partially mediated the relationship between optimism and cancer worry. The relationship between optimism, cancer worry and perceived susceptibility were examined in accordance with McGregor et al.'s (2004) findings but no such relationship was found in this study.

Table 5.21

Model Summary for the Change in the Self-prediction of Booking an Appointment

Model	Removed variables	R	R^2	Adjusted R^2	Std. Error of the Estimate	Change Statistics				
						R^2 Change	F Change	df1	df2	Sig. F Change
1 (All variables included)		.641	.411	.257	12.08669	.411	2.664	22	84	.001
2	Pos-neg information	.641	.411	.265	12.01619	.000	.011	1	84	.916
3	Neg-pos information	.640	.410	.273	11.95670	-.001	.151	1	85	.699
4	Intermediate social class	.636	.404	.274	11.94597	-.006	.844	1	86	.361
5	Dispositional optimism (LOT)	.635	.404	.282	11.87976	.000	.027	1	87	.869
6	HADS anxiety	.623	.389	.272	11.96167	-.015	2.231	1	88	.139
7	Perceived control (TPB)	.623	.389	.280	11.89508	.000	.001	1	89	.979
8	Anxiety – having genetic testing	.623	.388	.287	11.83666	-.001	.108	1	90	.743
9	Cancer worry	.622	.387	.294	11.77998	-.001	.121	1	91	.729
10	Weighted ratio (Utility theory & HBM)	.621	.386	.300	11.72813	-.001	.183	1	92	.670
11	Severity (HBM)	.618	.382	.303	11.70133	-.004	.571	1	93	.452
12	Subjective norm (TPB)	.606	.368	.295	11.77462	-.014	2.194	1	94	.142
13	Susceptibility (HBM)	.596	.355	.288	11.82795	-.012	1.872	1	95	.175
14	Coping	.585	.342	.281	11.88796	-.013	1.987	1	96	.162
15	Failed axiom tests	.585	.342	.288	11.82715	.000	.000	1	97	.993

N=107.

Table 5.22

Multiple Linear Regression Coefficients and P Values for the Change in the Self-prediction of Booking an Appointment (Final Model – Model 15, Parsimonious Specification)

	Unstandardised Coefficients		**Standardised Coefficients**	**t**	**Sig.**
	B	Std. Error	Beta		
Constant	-4.770	5.352		-.891	.375
Self-prediction of booking genetic test & counselling at baseline	-.289	.048	-.580	-6.077	.000
Positive information	8.528	2.530	.268	3.371	.001
Sex	-12.304	3.424	-.309	-3.594	.000
Managerial-professional social class	4.753	2.469	.156	1.925	.057
Age	.332	.186	.144	1.787	.077
Depression (HADS)	-.873	.459	-.151	-1.903	.060
Attitudes towards behaviour (TPB)	.155	.060	.243	2.565	.012
Anxious about developing breast cancer	.166	.046	.307	3.608	.000

N = 119, [F (8,110) =7.437, P=0.000], R=0.592, R^2 =0.351, Adjusted R^2 = 0.304

The final model and associated coefficients and statistics for the *change in self-prediction scores* are presented in Table 5.22. In addition to baseline self-prediction score, which is to be expected, seven significant predictor variables of change in the self-prediction of booking a genetic testing and counselling appointment were identified. The seven variables were age, depression ratings, attitudes towards behaviour, anxiety about developing breast cancer, being given positive information, being male and coming from a managerial or professional background (social class). The model explained 30.4% of the variance in the change from baseline to follow-up in the self-prediction scores.

An increase of one unit in age, attitudes towards behaviour and anxiety about developing breast cancer resulted in the following respective positive increases over baseline ratings of the self-prediction of booking an appointment (self-prediction scores ranged between 0 and 100) 0.332, 0.155 and 0.166. An increase of one unit in the depression score of the HADS scale resulted in a decline of -0.873 in baseline self-prediction scores. Being from a managerial and/or

professional background was associated with an increase of 4.753 in self-prediction scores, being given positive information with an increase of 8.528, whilst being male resulted in a decline of -12.204 in baseline self-prediction scores.

The final model and descriptive statistics for the *change in intention scores* are presented in Table 5.24. In addition to baseline intention scores, five predictor variables were identified. The Five variables were positive information, sex, subjective norm, attitudes towards behaviour and cancer worry. Positive information, sex and attitudes towards behaviour were also significant predictors on self-prediction scores. The final model explained 28.0% of the variance in the change in intention scores from baseline to follow-up.

Although the constant, baseline intention and male gender produced negative coefficients, the four remaining predictor variables all had positive coefficients. A one unit increase in subjective norm, attitudes towards behaviour or cancer worry score resulted respectively in an increases of 0.009, 0.184 and 0.136 above baseline intention scores (baseline and follow-up intention scores ranged between 0 and 14). Being issued with positive information resulted in an increase of 0.781 on baseline intention scores. A one unit increase in initial intention to have genetic testing and counselling and being male resulted in a decrease in baseline intention scores of 0.313 and 0.706 respectively.

Table 5.23

Model Summary for the Change in the Intention to Have Genetic Testing

Model	Removed variables	R	R^2	Adjusted R^2	Std. Error of the Estimate	Change Statistics				
						R^2 Change	F Change	df1	df2	Sig. F Change
1(All variables included)		.586	.343	.179	1.64559	.343	2.088	22	88	.008
2	Neg-pos information	.570	.325	.166	1.65822	-.018	2.372	1	88	.127
3	Pos-neg information	.562	.316	.163	1.66082	-.010	1.283	1	89	.260
4	Intermediate social class	.561	.315	.172	1.65199	.000	.035	1	90	.852
5	Age	.560	.314	.180	1.64476	-.001	.196	1	91	.659
6	Managerial social class	.554	.307	.181	1.64379	-.007	.890	1	92	.348
7	Dispositional optimism (LOT)	.547	.299	.180	1.64462	-.008	1.095	1	93	.298
8	HADS depression	.546	.298	.187	1.63693	-.001	.114	1	94	.736
9	HADS anxiety	.545	.297	.195	1.62937	-.001	.115	1	95	.736
10	Susceptibility (HBM)	.545	.297	.203	1.62111	.000	.019	1	96	.891
11	Anxiety – having genetic testing	.545	.297	.211	1.61311	.000	.035	1	97	.851
12	Perceived control (TPB)	.544	.296	.218	1.60588	-.001	.114	1	98	.736
13	Severity (HBM)	.541	.292	.222	1.60211	-.004	.532	1	99	.468
14	Coping	.538	.289	.226	1.59772	-.003	.446	1	100	.506
15	Anxiety – risk of developing breast cancer	.531	.282	.226	1.59745	-.007	.966	1	101	.328
16	Weighted ratio (Utility theory & HBM)	.515	.265	.215	1.60907	-.018	2.504	1	102	.117
17	Failed axiom tests	.514	.265	.222	1.60141	.000	.012	1	103	.912

N=111

Table 5.24

Multiple Linear Regression Coefficients and P Values for the Change in Intention to Have Genetic Testing (Final Model – Model 17, Parsimonious Specification)

	Unstandardised Coefficients		**Standardised Coefficients**	t	Sig.
	B	Std. Error	Beta		
Constant	-.748	.499		-1.499	.136
Intention to have genetic testing at baseline	-.313	.053	-.560	-5.861	.000
Positive information	.781	.302	.198	2.589	.011
Sex	-.706	.397	-.151	-1.779	.078
Subjective norm (TPB)	.009	.002	.329	3.781	.000
Attitudes towards behaviour (TPB)	.018	.007	.220	2.403	.018
Cancer worry	.136	.045	.261	2.997	.003

N = 129, [F (6,122) =9.281, P=0.000], R=0.560, R^2=0.313, Adjusted R^2 = 0.28

Survey of Patients Referred for Cancer Genetics Services

As specific care (the need for counselling and testing, number of counsellors involved and the need for joint counselling with a surgeon) is provided to CGSW patients according to their risk status and the type of cancer they are at risk of developing, patient preferences for the delivery of such services are at their most useful when established in respect of cancer and risk. Due to the relatively small sample size (N=115) analysis of risk status by cancer type would only be possible for the 81 patients at high, moderate and low risk of developing breast cancer. In the interests of brevity only results for high risk breast cancer patients are presented in the thesis.

Table 5.25

Discrete Choice (Multinomial Logit) Model for Attribute Main Effects for High Risk Breast Cancer Patients

Attributes	Levels	Coefficients (B)	Standard error	t-ratio	P value
Staff seen	Consultant geneticist	0.807891	0.141549	5.70752	1.14635e-008
	Specialist genetics nurse	-0.236943	0.168232	-1.40842	0.159005
	Genetics associate	-0.570949	0.19557	-2.9194	0.00350704
Waiting time for letter	1 month	1.11917	0.170467	6.56535	5.19111e-011
	2 month	0.605685	0.173166	3.49771	0.00046928
	4 month	-0.524831	0.18471	-2.84138	0.00449193
	6 month	-1.20003	0.246208	-4.87403	1.09346e-006
Distance to counselling	20 miles	0.281293	0.199913	1.40708	0.159404
	40 miles	0.693696	0.183402	3.78237	0.000155342
	60 miles	-0.996691	0.224472	-4.44015	8.98959e-006
	80 miles	0.0217017	0.175119	0.123926	0.901374
Duration of counselling	30 minutes	-1.48606	0.26215	-5.66874	1.43849e-008
	1 hour	0.365709	0.183933	1.98827	0.0467816
	1 hour 30 minutes	0.470156	0.188568	2.49329	0.0126565
	2 hour	0.650198	0.15015	4.33032	1.48894e-005
Availability of testing	High risk	0.0816467	0.115888	0.70453	0.481103
	All	-0.0816467	0.115888	-0.70453	0.481103
Cost of service	£1,500	0.402371	0.195398	2.05924	0.0394711
	£2,000	0.440799	0.194548	2.26576	0.0234661
	£2,500	0.170999	0.161888	1.05628	0.290842
	£3,000	-1.01417	0.243535	-4.16436	3.12232e-005
N =30, Number of observations=678, Log likelihood function=-552.6894, Restricted log likelihood (Log-L for Choice model) =-552.6894, R^2(McFadden's R^2)=0.25799, Adjusted R^2(Adjusted McFadden's R^2)=0.24969.					

The multinomial logit results for high risk breast cancer patients are presented in Table 5.25. The attribute availability of testing was non-significant for high risk patients. Although patients were willing to accept counselling from a genetics associate they would prefer to receive counselling from a consultant geneticist. Shorter waiting times were favoured as was travelling 40 rather than 60 miles for counselling. Longer counselling durations were preferred to shorter ones with 2 hour providing the greatest utility. Patients stated: "sufficient time should be provided in the counselling session in order that all your questions might be addressed"; "The need to ask many questions of your own – need to see sympathetic staff and have a long appointment". Lower cost of service attribute levels were preferred to higher costs; the highest cost attribute of £3,000 was statistically significant.

Consequences

High risk patients 'preference for' or 'utility of' alternative methods of providing counselling and testing were established based upon the attributes and levels that were found to be statistically significant to them in Table 5.25. Having ascertained willingness to pay results, the cost attribute was not required for the following analysis. With two attributes with four levels and two attributes with two levels it was possible to produce 64 service configuration scenarios. The service configurations, their estimated linear additive utility scores, costs and ranking are presented in Table 5.26.

It is important to note that the estimated utility scores (sum of relevant attribute level coefficients) in Table 5.26 are an ordinal measure that facilitates ranking of utility (preference) and should not be interpreted and used as a cardinal (ratio) measure (Schoemaker, 1982). Table 5.26 reveals that whilst patients would ideally like to travel 40 miles or less, have the longest possible counselling session of 2 hours, wait no more than 1 month from referral to receipt of a letter confirming their risk status and receive their counselling from a consultant geneticist; when this is not possible compensatory decision making takes place. Deficiencies in one attribute can be compensated for by an improvement in one or more of the remaining attributes. Whilst breast cancer patients would prefer to be counselled by a consultant geneticist they were willing to see a genetics associate rather than a consultant if the remaining attributes were more favourable. Four genetics associate led service scenarios were in the top quartile, with the service configuration scenarios ranked 7, 8 and 10 favoured over 26 scenarios where counselling was provided by a consultant. Service configuration scenarios 10 and 11 are good illustrative example, patients would prefer to be counselled by a genetics associate for 1 hour rather than a consultant for 2 hours when the waiting time is the same at 1 month but travelling distance to counselling is 40 miles rather than 60 miles.

Costs

The estimated costs of providing the 64 alternative service configurations from Table 5.26 to high risk presymptomatic breast cancer patients with a living cancer affected relative subsequently found to have a BRCA1/2 mutation are presented in terms of total cost (labour, capital and overhead). Lower costs were characterised by shorter counselling durations, longer waiting times for a letter confirming risk status, the patient travelling 60 miles to counselling (counsellor does not need to travel) rather than 40 miles and counselling sessions with a genetics associate rather than a consultant. The range in costs was substantial, with a range in mean total costs of £1,618 (minimum cost=£2,027, most expensive=£3,645. See Table 5.26).

Sensitivity

Sensitivity of the estimated costs in Table 5.26 to indirect patient contact was assessed by substituting the minimum and maximum reported percentages of indirect patient work time (25% and 75%) into the labour costs. Assuming indirect patient time to be 25% rather than the 33% assumed in the base case resulted in mean costs declining to 92-97% of the mean base case costs. Assuming 75% of work time to be devoted to indirect patient care/referral procedures resulted in costs rising to 141-195% of the mean base case costs. The largest savings (25%) and increased expense (75%) were associated with the service configurations with most labour input and the most expensive staff (consultants). For example, the largest increase in costs 195% was for 2 hours counselling with a consultant, 40 miles travelling distance to counselling and a 1 or 2 month wait for a letter confirming risk status.

Cost-consequences

Table 5.26

Utility (preference) of the Cancer Genetic Services Configurations and their Cost per Patient

Service configuration scenarios for event pathway 15				Utility score	Mean cost	Utility ranking	Cost ranking
Staff	**Wait**	**Distance**	**Duration**				
Consultant geneticist	1 month	40 miles	2 hours	3.270955	£3,645	1	63
Consultant geneticist	1 month	40 miles	1 hour 30	3.090913	£3,357	2	59
Consultant geneticist	1 month	40 miles	1 hour	2.986466	£3,064	3	55
Consultant geneticist	2 month	40 miles	2 hours	2.75747	£3,645	4	63
Consultant geneticist	2 month	40 miles	1 hour 30	2.577428	£3,357	5	59
Consultant geneticist	2 month	40 miles	1 hour	2.472981	£3,064	6	55
Genetics associate	1 month	40 miles	2 hours	1.892115	£2,855	7	49
Genetics associate	1 month	40 miles	1 hour 30	1.712073	£2,692	8	39
Consultant geneticist	4 month	40 miles	2 hours	1.626954	£2,951	9	53
Genetics associate	1 month	40 miles	1 hour	1.607626	£2,525	10	29
Consultant geneticist	1 month	60 miles	2 hours	1.580568	£3,466	11	61
Consultant geneticist	4 month	40 miles	1 hour 30	1.446912	£2,757	12	41
Consultant geneticist	1 month	60 miles	1 hour 30	1.400526	£3,178	13	57
Genetics associate	2 month	40 miles	2 hours	1.37863	£2,855	14	49
Consultant geneticist	4 month	40 miles	1 hour	1.342465	£2,559	15	31
Consultant geneticist	1 month	60 miles	1 hour	1.296079	£2,892	16	51
Genetics associate	2 month	40 miles	1 hour 30	1.198588	£2,692	17	39
Consultant geneticist	1 month	40 miles	30 minutes	1.134697	£2,778	18	45
Genetics associate	2 month	40 miles	1 hour	1.094141	£2,525	19	29
Consultant geneticist	2 month	60 miles	2 hours	1.067083	£3,466	20	61
Consultant geneticist	6 month	40 miles	2 hours	0.951755	£2,951	21	53
Consultant geneticist	2 month	60 miles	1 hour 30	0.887041	£3,178	22	57
Consultant geneticist	2 month	60 miles	1 hour	0.782594	£2,892	23	51
Consultant geneticist	6 month	40 miles	1 hour 30	0.771713	£2,757	24	41
Consultant geneticist	6 month	40 miles	1 hour	0.667266	£2,559	25	31
Consultant geneticist	2 month	40 miles	30 minutes	0.621212	£2,778	26	45
Genetics associate	4 month	40 miles	2 hours	0.248114	£2,424	27	23
Genetics associate	1 month	60 miles	2 hours	0.201728	£2,758	28	43
Genetics associate	4 month	40 miles	1 hour 30	0.068072	£2,313	29	15
Genetics associate	1 month	60 miles	1 hour 30	0.021686	£2,598	30	33
Genetics associate	4 month	40 miles	1 hour	-0.03638	£2,200	31	7
Consultant geneticist	4 month	60 miles	2 hours	-0.06343	£2,821	32	47
Genetics associate	1 month	60 miles	1 hour	-0.08276	£2,438	33	27
Consultant geneticist	4 month	60 miles	1 hour 30	-0.24348	£2,630	34	37
Genetics associate	1 month	40 miles	30 minutes	-0.24414	£2,368	35	19
Genetics associate	2 month	60 miles	2 hours	-0.31176	£2,758	36	43
Consultant geneticist	4 month	60 miles	1 hour	-0.34792	£2,436	37	25
Genetics associate	6 month	40 miles	2 hours	-0.42709	£2,424	38	23
Genetics associate	2 month	60 miles	1 hour 30	-0.4918	£2,598	39	33
Consultant geneticist	4 month	40 miles	30 minutes	-0.5093	£2,369	40	21
Consultant geneticist	1 month	60 miles	30 minutes	-0.55569	£2,605	41	35
Genetics associate	2 month	60 miles	1 hour	-0.59625	£2,438	42	27
Genetics associate	6 month	40 miles	1 hour 30	-0.60713	£2,313	43	15

Genetics associate	6 month	40 miles	1 hour	-0.71157	£2,200	44	7
Consultant geneticist	6 month	60 miles	2 hours	-0.73863	£2,821	45	47
Genetics associate	2 month	40 miles	30 minutes	-0.75763	£2,368	46	19
Consultant geneticist	6 month	60 miles	1 hour 30	-0.91867	£2,630	47	37
Consultant geneticist	6 month	60 miles	1 hour	-1.02312	£2,436	48	25
Consultant geneticist	2 month	60 miles	30 minutes	-1.06918	£2,605	49	35
Consultant geneticist	6 month	40 miles	30 minutes	-1.1845	£2,369	50	21
Genetics associate	4 month	60 miles	2 hours	-1.44227	£2,350	51	17
Genetics associate	4 month	60 miles	1 hour 30	-1.62232	£2,242	52	9
Genetics associate	4 month	60 miles	1 hour	-1.72676	£2,135	53	5
Genetics associate	4 month	40 miles	30 minutes	-1.88814	£2,095	54	3
Genetics associate	1 month	60 miles	30 minutes	-1.93453	£2,278	55	13
Genetics associate	6 month	60 miles	2 hours	-2.11747	£2,350	56	17
Consultant geneticist	4 month	60 miles	30 minutes	-2.19969	£2,245	57	11
Genetics associate	6 month	60 miles	1 hour 30	-2.29751	£2,242	58	9
Genetics associate	6 month	60 miles	1 hour	-2.40196	£2,135	59	5
Genetics associate	2 month	60 miles	30 minutes	-2.44802	£2,278	60	13
Genetics associate	6 month	40 miles	30 minutes	-2.56334	£2,095	61	3
Consultant geneticist	6 month	60 miles	30 minutes	-2.87489	£2,245	62	11
Genetics associate	4 month	60 miles	30 minutes	-3.57853	£2,027	63	1
Genetics associate	6 month	60 miles	30 minutes	-4.25373	£2,027	64	1

Note: ═══ signifies the separation of quartiles. Ranks 1-16=1st quartile, 17-32=2nd quartile, 33-48=3rd quartile, 49-64=4th quartile.

Table 5.26 contains the 64 service configuration scenarios ranked from maximum to minimum estimated utility (preference/desirability) accompanied by means estimated costs. The results show that not only did counselling by a genetics associate accompanied by favourable levels of other attributes provide high utility, e.g. scenarios ranked 7, 8, 10 and 14 were in the top quartile, but they also provide substantial cost savings. For example, the scenarios ranked first and seventh have the most favourable levels of the attributes waiting, distance and duration, but the first has a consultant and the seventh has a genetics associate giving counselling. The scenario ranked seventh was £790 cheaper per presymptomatic patient than the scenario ranked first[9]. From a cost consequences perspective the scenario ranked tenth (in terms of utility/patient preference) emerges as the most desirable scenario configurations in the author's opinion. The scenario (genetics associate, 1 month, 40 miles, 1 hour) achieved one of the highest utility/desirability rankings and at a cost of £2,525; scenario estimated costs ranged between (£2,027 and £3,645).

[9] £3,645 (consultant geneticist, 1 month, 40 miles, 2 hours) - £2,855 (genetics associate, 1 month, 40 miles, 2 hours) = £790.

CHAPTER 6: DISCUSSION

Reliability and Validity (Chapter 4)

Information Manipulation Experiment

Reliability and Validity of Psychosocial Measures

The majority of composite measures used in the experiment had Cronbach alpha coefficients approaching or above 0.7. There was one exception to this, the weighted ratio score, which comprises the completely opposing variables of the sum of the weighted pros and the sum of the weighted cons of genetic testing and counselling. As we would not expect a strong alpha coefficient between such variables; the internal consistency of all the measures used in this experiment are acceptable.

The degree of agreement between the post-coding of open ended responses in this thesis with a test set (inter rater reliability test) was acceptable.

Both the new and established measures administered to respondents were found to be valid based upon correlation coefficients revealing acceptable levels of concurrent validity (see Tables 4.7 to 4.10).

Validity of Discrete Choice Measures

Acceptability and ease of completion.

Only seven of the cohort of 142 participants provided missing data (one at baseline and six at follow-up) on the discrete choice questions, and only one of these provided missing data on more than one choice question. One respondent provided missing data for choices 13 to 18 at follow-up. Given that these six choices were the entire content of two adjoining pages it is most likely that the two pages were turned simultaneously by mistake rather than any reluctance on the part of the respondent to answer these questions at follow-up. Hundley and Ryan (2004) also reported a very high completion rate for their DCM questionnaire.

Three respondents reported indifference between all of the scenarios presented to them; two respondents at follow-up and one at baseline and follow-up. No constant selection of response option A or B was found. These results in conjunction with all the attributes (not all levels) being statistically significant at baseline (see Table 4.12) indicated that the majority of respondents found the attributes and levels to be realistic and trade-offs (compensatory decision making) between attributes to be possible.

Respondents mean ratings of the difficulty of completing the discrete choice exercise indicated that they found the questions moderate to moderately easy to answer at baseline. At follow-up, respondents reported a significant increase in the ease of answering these choice questions; which no doubt reflects their increased familiarity with the question format by follow-up.

Clearly the small number of missing values, lack of a response choice bias (selecting one option constantly) and the ratings on the difficulty of answering the discrete choice questions indicate that respondents found these items acceptable, realistic and easy to answer. These results confirm the face and content validity found when the discrete choice question format was piloted.

Construct validity.

Concurrent validity.

Comparison of baseline preference on the 'availability of genetic testing' attribute ('available to all' or 'available to high risk only') with the subsequently collected and calculated multinomial logit coefficients concurred; providing concurrent validity for the model results on 'availability of testing'. Comparison of preferences for 'staff seen' with the model coefficients did not concur. The discrepancy is most likely due to the large number of respondents not answering the ranking exercise questions; at baseline 46-48 respondents did not rank their preferences for the members of staff they could be counselled by.

Theoretical/internal validity.

The 'distance to counselling' attribute did not comply with expectations; utility coefficients did not increase as distance declined. The largest distance to

counselling attribute level of 80 miles was statistically non-significant; indicating that it made no noteworthy contribution to respondents utility. This may be a result of 80 miles being considered too far or that they did not find travelling 80 miles substantially worse than travelling 60 miles which was a significant level with the largest negative coefficient.

Although travelling 20 miles and travelling 40 miles produced greater utility than having to travel 60 miles, greater utility was associated with travelling 40 miles than 20 miles. The greater utility of travelling 40 rather than 20 miles may be a result of a threshold value or a desire for anonymity. Once travelling distance reached the threshold of 40 miles or less, respondents may be satisfied with this attribute and feel that a further reduction in travel distance did not merit making substantial trade-offs in other attribute levels. Alternatively respondents may desire anonymity. Barriers to genetic testing for breast cancer cited by the respondents included: life insurance problems, implications for the rest of the family, feel uncomfortable/ not feel oneself, rejection and discrimination. Some respondents may well feel more comfortable at a clinic 40 miles away rather than 20 miles, where they are less likely to meet family, friends and acquaintances.

The level £2,500 on the cost attribute was statistically non-significant ($p=0.79$), whilst all the remaining levels were significant ($p<0.0000007$). The level £2,500 made no substantive contribution to respondents' utility, with a coefficient of almost zero (0.024). This suggests that the level £2,500 was taken as a norm; which is probably a result of £2,500 being the level of the cost of service attribute used in the constant comparator. The size and sign of the attribute level coefficients (B) relative to the baseline values coded in the multinomial logit model for the continuous attributes of waiting time, duration of counselling and cost of service complied with expectations, providing evidence of theoretical validity. If, as is the case here, attribute preference directionality is known a priori, this ensures high levels of explained variance (Dawes & Corrigan, 1974; Anderson & Shanteau, 1977).

Six respondents had discontinuous preferences, two with a dominant preference for waiting time and four for availability of testing. They ranked the respective

attributes as the most important to them and always chose a scenario with a favourable level of the dominant attribute no matter how poor the levels of the remaining attributes (Ratcliffe, 2000a; Ratcliffe & Buxton, 1999). It is important to note that dominance was only proven for the set of fractional factorial scenario designs presented to respondents. Had the full factorial design been presented dominance may not have prevailed (Scott, 2002).

The majority of respondents (89.4% - 95.8%) complied with expectations and selected the dominant scenarios in the non-satiation test and three transitivity tests at baseline. In the past respondents selecting dominated scenarios were considered to be irrational (Ratcliffe & Buxton, 1999; Ryan, 1999b; Ryan & Hughes, 1997, Wilson et al., 1999; Ryan & Farrar, 2000; San Miguel, Ryan & Amaya-Amaya, 2005). In this instance the term irrational is misleading; respondents may select scenarios that appear to be inferior or report no preference between a dominant and dominated scenario for a variety of reasons.

Firstly, they may be using a simplifying decision heuristic (Cairns, van der Pol & Lloyd, 2002; Manstead & Hewstone, 1995; Maddala, Phillips & Johnson, 2003), dominance, target setting or lexicographic preference. As has been noted above some respondents had a dominant preference for certain attributes. Target setting[1] may be in operation (San Miguel et al., 2000; Scott, 2002); only opting for a scenario that equals or exceeds a specific value on the dominant attribute and then comparing (trading-off) the remaining attribute levels. Alternatively, respondents may have a lexicographic or hierarchical decision preference (Cave et al., 1994; Lloyd, 2003; Parker & Srinivasan, 1976; Ratcliffe, 2000a; Ratcliffe & Buxton, 1999); where there is an absolute preference order for attributes and no substitution between them.

Although the multinomial logit regression coefficients suggest that there may be some target setting in the form of threshold values for distance, it was not possible to tell in this study if target setting and lexicographic decision heuristics

[1] Target setting is more commonly known in the field of psychology as the conjunctive/disjunctive decision rule.

were in use. It is difficult to identify target setting and lexicographic decision heuristics with self administered questionnaires (Scott, 2002).

Secondly, respondents may be ignoring questionnaire instructions and reinterpreting the attribute levels and scenarios presented to them (Hanson et al., 2005; San Miguel et al., 2005). In a qualitative and quantitative study of seemingly irrational preferences on a DCM questionnaire San Miguel et al. (2005) found six explanations in addition to dominance (consistent underlying preference). 44% had genuinely contradictory preferences. The remaining 56% employed a rational decision process with flawed assumptions:

- 8% - Information gained from other choices led to the rejection of some choices completely.
- 9% - Additional information, characteristics or assumptions were made. (For example, Monroe (1971) amongst others has found that consumers sometimes use price as a measure of quality).
- 4% - Protest responses and ignoring choices that differ to their experience of the area under study.
- 10% - Indifference to certain choice questions.
- 13% - Random error.
- 12% - Dominance.

Thirdly, it may be the case that the intervals between attribute levels are not sufficiently large to entice some respondents to trade between attributes (Cairns et al., 2002; Viney, Lanczar & Louviere, 2002). In a test of dominance Cairns and van der Pol (2001) and Cairns et al. (2002) found that increasing interval levels revealed that almost all respondents (99.5%) did not have a dominant preference. As attribute levels are selected to be realistic and as a result provide valuable data for service development and policy, artificially inflating these values for any purpose other than checking reliability and validity would be inadvisable (Ratcliffe, Buxton, McGarry, Sheldon & Chancellor, 2004; Scott, 2002).

In total 28 respondents (19.7%) provided choices that did not comply with the random utility axioms of dominance, transitivity and non-satiation. This result is favourable compared to many other DCM studies, for example, 74% of respondents were recorded as having dominant and/or intransient preferences by McIntosh and Ryan (2002).

There was a significant difference between those respondents passing and those failing the dominance and transitivity tests in terms of their preferences for the manner in which cancer genetic services are delivered. The utility function for the respondents that *failed* the dominance and transitivity tests had the lowest number of significant attributes ($p<0.10$) at thirteen; explaining 15% (adjusted $R^2=0.15221$) of the variance in the discrete choices made. The utility function for the respondents *passing* the dominance and transitivity tests had sixteen significant predictors attribute levels ($p<0.10$) and explained 40% (adjusted $R^2=0.39860$) of the variance in the discrete choices made. Excluding the respondents that failed the tests therefore increased the explained variance in choices by 7% (un-segmented model's adjusted $R^2=0.331$). Similar discrepancies were found by Bryan et al. (1998), San Miguel et al. (2000) and van der Pol and Cairns (2001) when they found respondents with dominant preferences and McIntosh and Ryan (2002) when they found discontinuous preferences.

No significant differences were found between those passing and failing the utility theory axiom tests (non-satiation, dominance and transitivity) on the weighted ratio, self-prediction or intention variables. The axiom variable remained a non-significant predictor of the three dependent variables (weighted ratio, self-prediction or intention) when it was included with the other potential predictor variables in the repeated measures GLM and the multiple linear regression analysis. Respondents failing the dominance and transitivity tests were characterised by comprising significantly more mature students, parents and having higher dispositional optimism. San Miguel et al (2005) also found a relationship between age and results on axiom tests; finding younger and older people more likely to fail the tests.

Presently there is a disagreement in the field of discrete choice modelling relating to how to deal with respondents that violate theoretical axioms such as dominance, transitivity and continuity (tested with dominance, transitivity and non-satiation tests). This disagreement stems from decision theory. DCM was developed upon the assumption that individuals make choices in accordance with random utility theory (RUT) (Ben-Akiva & Lerman, 1985; Louviere et al., 2000; Ryan, 1996a; Ryan et al., 1996). The axioms of the forerunners of random utility theory, subjective expected utility theory and expected utility clearly state that utility theory should only be applied to individuals that comply with the axioms of utility theory such as transitive and continuous preference. Within the field of Health Economics it has become customary for some economists to examine responses and exclude respondents that do not comply with the transitivity and continuous axioms (Hanson et al., 2005; McIntosh & Ryan, 2002; Ratcliffe & Buxton, 1999; Ratcliffe & Longworth, 2002; Ryan, 1999b; Wilson et al., 1999; Ryan & Farrar, 2000). Some health economists advocate presenting the results of their analysis for the sample of respondents as a whole and for those respondents that adhere wholly to the axioms of random utility theory (San Miguel et al., 2000; Scott, 2002; Ratcliffe, van Haselen, Buxton, Hardy, Colehan & Partridge, 2002; Ratcliffe et al., 2004; van der Pol & Cairns, 2001). However, some health economists and other disciplines using discrete choice stated preference elicitation such as transport economics, environmental economics and marketing (Hall, Kenny, King, Louviere, Viney & Yeoh, 2002) do not exclude any respondents (Hall et al., 2002). Three reasons have been cited in support of including all respondents in the analysis. Firstly, it preserves the orthogonality of the questionnaire design (Hall et al., 2002; Lancsar & Savage, 2004b), which allows the parameters of interest in a model to be estimated independently of one another (Louviere et al., 2000). Secondly, including all respondents yields results that are representative of the sample drawn and if the sample has been drawn correctly, the population from which the sample was drawn (Bryan et al., 1998; Bryan & Dolan, 2004; Lancsar & Savage, 2004b; Scott, 2002; Viney, Lancsar & Louviere, 2002). Excluding respondents would result in findings that cannot be generalised to the entire population of interest and as a result should not be used to inform service provision and health policy for that population. Thirdly, Viney et al. (2002) suggest that the random component in random utility theory allows

for the seemingly irrational violations of the theory's axioms e.g. dominant preference, lexicographic preferences, intransient preferences and selecting inferior scenarios (non-satiation test). Including all respondents is clearly the better method of obtaining representative research results and as a result the respondents failing the axiom tests were retained for all the analysis in this experiment.

Clearly there is evidence of construct validity for the DCM questionnaire used and the data gathered from it. Evidence of concurrent validity was found in relation to the attribute availability of testing and theoretical validity was found in relation to the attributes waiting time, duration of counselling and cost of service. Also 80.3% of respondents complied with the transitivity and continuous preference axioms and the mix in the order of the attribute levels (see Table 4.19 and Figure 4.1) confirmed that the respondents as a whole had continuous preferences.

Response options and coding.

This study differs with the majority of applications of discrete choice modelling in health economics as it has a 'no preference' (indifference or non-demand) response option for respondents that are indifferent to the two scenarios presented to them (Ryan, 1999a; Ryan & Skatun, 2004; Viney et al., 2002). As the 'no preference' response option was used by 2.8% to 8.5% of respondents in each of the choices presented to them, it is clearly an improvement over questionnaires that neglected to include this option both in terms of data collection and consequently representative results. Failure to allow for indifference or opting out when this is a realistic option would result in forced choices for respondents that do not opt to leave the choices blank (missing values), result in over estimation of preferences and uptake of health services (Ryan & Skatun, 2004; Viney et al., 2002).

In addition to effects coding, discrete choice data can be coded using dummy codes, ordinal ranking codes and interval/ratio values (Farrar & Ryan, 1999; Johnson et al., 2000; Louviere et al., 2000; Ratcliffe et al., 2002). In the

following text comparisons will be drawn between the coding used in this study and the possible alternatives.

Dummy variables were not used in this study as it would have been impossible to code the attribute levels for the 'no preference' response option in each of the choices presented to respondents. It is worth noting that Bech and Gyrd-Hansen (2005), Louviere et al. (2000) and Viney et al. (2002) advocate effects codes over dummy codes when using fixed constant comparator choices (e.g. Choice A is always the same scenario) as a constant term should be specified in the regression equation and when dummy codes are used there is no unique interpretation of the coefficient. The constant term (intercept) may reflect a preference for or against the constant scenario and/or the utility associated with the omitted attribute levels as all dummy coded coefficients are correlated with the intercept.

Ordinal codes were used by Wilson et al. (1999) in their study assessing user preferences for cancer genetic counselling (Genetics Associate = 1, Genetics Nurse = 2, Consultant = 3). The ordinal codes were determined by means of asking respondents to rank their preference amongst counsellors. The motivation behind utilising this coding system is that if a linear additive model is assumed and the ordinal codes are assumed to have ratio properties, the attribute coefficients can be combined (as numerators and denominators) to determine marginal rates of substitution for the entire attribute.

There are a number of issues with utilising ordinal codes with nominal data. Firstly, in this study a 'no preference' response could not be coded for a nominal attribute. For example, if a respondent was indifferent between a scenario with a consultant and a genetics associate, a mid point code of 2 could not be used as this represents a nurse on the codes (Genetics Associate = 1, Genetics Nurse = 2, Consultant = 3). Secondly, since Von Neumann and Morgenstern's (1944; 1947) expected utility theory, utility theories have implicitly assumed that full cardinal utility (interval to ratio data characteristics) exists otherwise it would be impossible to psychologically determine the certainty equivalence of a risky choice (Schoemaker, 1982). However, preferences are determined by at least two

factors; strength of preference for a choice and attitudes towards risk. The random utility function is a combination of these two factors without the aid of interval comparison and strength of preference measures; thus as a preference measure it is wholly ordinal, providing no more than ordinal ranking of choices (Schoemaker, 1982). Examination of the coefficients for staff seen in Table 4.12 provides support for Schoemaker's interpretation. The coefficients for staff seen do not have the ratio properties required to conduct marginal rates of substitution, the gap between the preferred choice and the second (0.84 and -0.36) and the second and third (-0.36 and -0.48) are not equal increments[2].

The continuous attributes in this study could have been coded using their actual attribute levels (interval/ratio coding values) e.g. 1 hour 30 minutes as 1.5 for the attribute duration of counselling, and the mid point of competing attribute levels for indifference (no preference) responses. However, Schoemaker's (1982) comments are equally valid for ratio coding of continuous variables. A cursory glance at the equally spaced attribute levels of distance to counselling, duration of counselling and cost of service in Table 4.12 reveal that the utility coefficients produced by the multinomial logit model are not equally spaced. This finding indicates that using interval/ratio codes in this study would have been inappropriate, yielding unreliable results in the multinomial logit regression and conducting marginal rates of substitution on the coefficients would have compounded the error. However, it should be noted that Telser and Zweifel (2002) found evidence of linearity in coefficients for ordinal and continuously coded variables. Telser and Zweifel did not provide any of their results. It should be noted that the discrepancy between the results of this experiment and the results of Telser and Zweifel may well stem from the differences in the methods used in both studies. Firstly, there was no indifference response option in Telser and Zweifel's study. Secondly, random effect binary choice probit regression was conducted in their study. Thirdly, the alternative coding used by Telser and Zweifel was dummy coding. Finally, equivalence was established by means of comparing dummy coded models with ordinal and continuously coded models

[2] In contrast to DCM; in the field of conjoint analysis (ranking and rating) it has been proven by Luce and Tukey (1964) that cardinal data can be obtained from ordinal data when preferences obey cardinal axioms (San Miguel et al., 2000).

using a likelihood ratio test. It is possible that the likelihood ratio test was not sensitive enough to detect the type of differences seen in Table 4.12.

Survey of Patients Referred for Cancer Genetics Services

Validity of Discrete Choice Measures

The following discussion of the evidence upon the validity of the DCM questionnaire when administered to patients of cancer genetics services is brief. The results are in keeping with those found in the information manipulation experiment when the same questionnaire was administered to students; that more detailed discussion is presented above.

Acceptability and ease of completion.

The small number of missing values, lack of a response choice bias (selecting one option constantly) and the favourable ratings on the difficulty of answering the DCM questions indicate that respondents found these items acceptable, realistic and easy to answer.

Construct Validity

Concurrent validity.

Comparison of ranked preferences on the 'availability of genetic testing' attribute ('available to all' or 'available to high risk only') and 'staff seen' ('consultant', 'nurse', 'associate') with the subsequently collected and calculated multinomial logit coefficients concurred; providing evidence of concurrent validity for the model results on both attributes.

Theoretical/internal validity.

There is evidence of construct validity for the DCM questionnaire used and the data gathered from it. Theoretical validity was found in relation to the attributes waiting time, duration of counselling, distance to counselling and cost of service as they complied with apriori expectations.

The largest and smallest distance to counselling attribute level of 80 miles and 20 miles, and the cost attribute of £2,500 were statistically non-significant. This may

be a result of 80 miles being considered too far or that respondents did not find travelling 80 miles substantially worse than travelling 60 miles which was a significant level with the largest negative coefficient. They would prefer to travel 40 miles rather than 20 miles. The greater utility of travelling 40 rather than 20 miles may be a result of a threshold value or a desire for anonymity. Once travelling distance reached the threshold of 40 miles or less, respondents may be satisfied with this attribute and feel that a further reduction in travel distance did not merit making substantial trade-offs in other attribute levels. Alternatively respondents may desire anonymity. In the experiment, students cited anonymity barriers to genetic testing for breast cancer such as life insurance problems, implications for the rest of the family, feel uncomfortable/ not feel oneself, rejection and discrimination. Some patients may well feel more comfortable at a clinic 40 miles away rather than 20 miles, where they are less likely to meet family, friends and acquaintances.

The level £2,500 on the cost attribute was statistically non-significant ($p=0.29$), whilst all the remaining levels were significant ($p<0.04$). The level £2,500 made no substantive contribution to respondents' utility, with a coefficient of 0.17. This suggests that the level £2,500 was taken as a norm; which is probably a result of £2,500 being the level of the cost of service attribute used in the constant comparator.

Results of Aims Objectives and Hypothesis Testing (Chapter 5)

Information Manipulation Experiment

Benefits and Barriers

By far the most frequently cited benefits of cancer genetic testing and counselling at baseline were "prevention/early detection" and "promoting greater understanding of breast cancer" with 49.9% of all the benefits listed at baseline falling into these two categories. The third most popular benefit was "to discover susceptibility status". The fourth to sixth most popular categories ("social support/counselling", "come to terms with the possibility of breast cancer", "help the family come to terms with the possibility of breast cancer") all related to

coming to terms with the realisation that they and their family had a gene predisposing them to have a high risk of developing breast cancer. The most frequently cited barrier to genetic testing and counselling was "increased worry/anxiety/distress/depression", which accounted for 31.1% of all the disadvantages reported. The remaining barriers were far less frequently cited with the second most popular category "life insurance problems/financial issues/test expensive/time/travel" only accounting for 10.2% of the barriers cited.

The benefits and barriers reported by experiment participants showed strong agreement with preceding studies which had used open ended questions to elicit the perceived benefits and barriers of genetic testing and counselling from students and the general public (Wroe et al., 1998), women awaiting mammography and gynaecology appointments (Chaliki et al., 1995) and families with a history of breast and ovarian cancer (Lerman, Seay Balshem & Audrain, 1995; Struewing, Lerman, Kase, Giambarresi & Tucker, 1995) (see Appendix R for a full list of the reasons/response categories reported in these studies). Some discrepancies were found with the benefits and barriers reported by Wroe et al. (1998), however these appear to be the result of the manner in which the questions were posed to respondents.

Weighted Ratio of Pros to Cons

Hypothesis 1[3] was positively confirmed with the experiment's information inducing statistically significant change in the weighted ratio of pros to cons. Hypothesis 2[4] was rejected, despite the control and negative groups (pos-neg and neg-pos) reacting as predicted, the positive group did not. Whilst little change was recorded for the control group, the negative groups (pos-neg and neg-pos) recorded a decline in mean weighted ratio scores and the positive group also showed a small decline in weighted ratio scores.

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[3] Hypothesis 1: Experiment information will produce a statistically significant change in the weighted ratio of pros to cons of testing and counselling.

[4] Hypothesis 2: Positive information will increase the pros relative to the cons of genetic testing and counselling recoded by respondents, both the pos-neg (Wroe & Salkovskis, 1999, called this group the negative information group) and the neg-pos information will increase recorded cons relative to the pros, and little or no change will occur for the control group.

The greater decline in weighted ratio scores for the neg-pos and pos-neg groups relative to the remaining information groups (Figure 5.3) indicate that including negative information along with the positive information substantially impacted upon the ratio of important benefits to barriers in the weighted ratio score, with the importance of barriers increasing. Surprisingly positive information alone had little impact, and the effect was to reduce the mean score of the weighted ratio of pros to cons. The small decline for both the positive and control group suggest that this decline may be, in part for the positive group and completely for the control group, due to the questionnaire. The weighted ratio questions were located at the beginning of the questionnaire; it may well be that by the time participants provided weighted ratio scores at follow-up that answering the questionnaire caused them to consider the consequences of developing genetic cancer. There were also a number of questions that could result in participants concentrating on very negative issues, for example, they were asked to consider how anxious they would be if they found they had a genetic mutation, how they would cope and how serious an illness they thought cancer was.

Hypothesis 3[5] was also rejected. The larger decline in weighted ratio scores for the neg-pos group than the pos-neg group suggests that there was an ordering effect. However, as was noted previously in the pair-wise comparison of these two information groups in the 'continuity within information groups' section, there was not a statistically significant difference in the decline in weighted ratio scores between these two groups. Cameron and Diefenbach (2001) also found no statistically significant ordering/primacy effect in their experiment. A significant ordering effect would clearly have implications for genetic counselling as providing negative information first or second would impact upon how positively they viewed genetic testing and counselling.

The significant relationship between dispositional optimism and the weighted ratio of pros to cons indicates that, at baseline more pessimistic individuals had a higher ratio of the benefits to barriers of testing and counselling than the more

[5] *Hypothesis 3*: Neg-pos information will result in a statistically significantly greater decline in the ratio of pros to cons between assessments (baseline to follow-up) than the pos-neg information (ordering/primacy effect).

optimistic respondents. However, once respondents had considered genetic testing and counselling in light of the questionnaire, the ratio of the benefits to barriers declined (benefits still outweighed the barriers but to a lesser extent) and the more pessimistic respondents had higher weighted ratio scores (recording more important benefits of testing and counselling) than the more optimistic respondents. As dispositional optimism was measured using the Life Orientation Test which gives a continuous score it was not possible to ascertain if there was an interaction between optimism and the information provided to respondents.

Hypothetical Intention and Self-prediction of Having Genetic Testing and Counselling

Hypotheses 4[6] and 5[7] were rejected as information did not statistically significantly changed participants' behavioural intention to have genetic testing and counselling for breast cancer on either the self-prediction or intention measures. Only self-prediction changed from baseline to follow-up at a statistically significant level. Irrespective of the information issued to respondents, the change in self-prediction of having genetic testing and counselling for breast cancer from baseline to follow-up only differed significantly by gender.

Hypothesis 6[8] was rejected as behavioural intention (self-prediction and intention) did not change significantly by information group and results did not comply with theoretical expectations as the pos-neg group recorded an increase in self-prediction and intention scores.

Analysis of knowledge and health perceptions revealed that the change in the self-prediction of booking a genetic testing appointment which was mediated by gender was mirrored in the change in the ratings of anxiety about having genetic

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[6] Hypothesis 4: Experiment information produces statistically significant change in self-prediction and intention to have testing and counselling.

[7] Hypothesis 5: Positive information will increase self-prediction and intention scores, both pos-neg and neg-pos information will reduce self-prediction and intention scores, and little or no change in scores will occur for the control group.

[8] Hypothesis 6: Behavioural intention (intention and self-prediction) of booking an appointment for genetic testing and counselling for breast cancer will decline between assessments statistically significantly more for the neg-pos than the pos-neg information group (ordering/primacy effect).

testing. It is possible that the latter finding is a result of the self-prediction of opting for testing dictating the relevance of testing to respondents, which in turn affects how anxious they feel about genetic testing and counselling for breast cancer. Men were less likely to have testing and counselling and as a result were less likely to feel anxious about having genetic testing; the contrary was found for women.

In contrast to the findings of the current experiment, Wroe and Salkovskis' (1999) experiment showed that the information manipulation and the subsequent focussing manipulation independently affected the self-prediction of taking a predictive genetics test for breast cancer. They found participants in the positive group stated that they were more likely to opt for testing after each manipulation, the converse was stated by the negative group and no change was reported by the control group. Post-manipulation the negative (pos-neg) group ratings were found to be significantly different to those of the control and the positive group in terms of anxiety and perceived severity of developing cancer. Whilst the control and positive group had provided relatively constant ratings on both items (increased anxiety for controls), the group receiving negative information showed a statistically significant decline in anxiety and perceived severity. Wroe and Salkovskis concluded that respondents receiving negative information coped by minimising the perceived severity of the threat which in turn reduced their anxiety. In contrast the positive group concentrated on what they could and should do to minimize morbidity and mortality which led to increased anxiety.

There is convergence between the results of the current experiment and the findings of Wroe and Salkovskis (1999) in terms of self-prediction and anxiety scores mirroring each other. However, unlike the findings of the current experiment, Wroe and Salkovskis found that anxiety levels rose for the control group.

Discrete Choices and Utility

Although the explained variance for the un-segmented model had hardly changed at all between assessments (adjusted R^2 declined from 0.330 to 0.326), substantial change was seen when information group was accounted for.

Explained variance rose by 2% for the control group (baseline adj. R^2=0.28, follow-up= 0.30), declined by 3% for the neg-pos group (baseline adj. R^2=0.34, follow-up= 0.31), declined by 8% for the pos-neg group (baseline adj. R^2=0.38, follow-up= 0.30), and rose by 5% for the positive group (baseline adj. R^2=0.35, follow-up= 0.40). The only constant pattern of change seen in the utility function of the information groups was in relation to the attribute 'availability of testing'. At baseline both levels of the attribute ('available to all' and 'available to high risk patients only') were significant for all information groups except for the pos-neg group. By the follow-up assessment the attribute was statistically non-significant ($p>0.10$) for all information groups.

The Relationship between Benefits and Barriers and Behavioural Intention to Have Testing

Hypothesis 7[9] was rejected with the analysis revealing only small to medium correlation between weighted ratio scores and the self-prediction and intention scores. Theory dictates that if individuals are using random utility theory that they weigh up the pros and cons of their options and choose the option with the maximum subjective expected utility (Wroe et al., 1998). In contrast to these findings Wroe et al. (1998) in their descriptive study found a large degree of correlation between weighted ratio scores and the self-prediction of opting for genetic testing ($r=0.75$, $p<0.0001$). The substantial discrepancy between these results may be a result of the current experiment looking at a hypothetical decision with students and Wroe et al. conducting their correlation on data obtained from 62 members of the general public that had considered going for genetic testing (they were asked their self-prediction of going for the genetic test they had been considering having).

Alternatives to Random Utility Theory

Multiple linear regression upon change in respondents' intention to have genetic testing identified six significant predictor variables; baseline intention score, positive information, sex, and the health cognition variables subjective norm (TPB), attitudes towards behaviour (TPB) and cancer worry. Higher cancer

[9] Hypothesis 7: A large degree of positive correlation (0.5 – 1.0) will exist between weighted ratio scores and self-prediction and intention to have testing scores.

worry, higher desire to comply with subjective norm, a positive attitude towards health behaviour (attitudes towards behaviour) and being issued with positive information all increased the respondents' intention to have genetic testing. Males were less interested in genetic testing than females. The regression analysis upon self-prediction scores identified eight significant predictor variables. As was the case for intention scores, the baseline score for the dependent change variable (baseline self-prediction), positive information, sex and attitudes towards behaviour (how valuable genetic testing is perceived to be based upon the perceived most likely outcome of testing) were significant predictors. The beta coefficients (standardized B coefficients) and t values in Tables 5.22 and 5.24 indicate that all the common variables exerted more influence upon the change in self-prediction scores than they did upon intention scores. The remaining significant predictors were the demographic variables (age and managerial-professional social class) and emotions (anxiety about developing cancer and depression). The sign of the regression coefficients revealed that a positive attitude towards health behaviour, being older, higher anxiety about their risk of developing breast cancer, coming from a managerial or professional social class and being issued with positive information, all increased respondents self-prediction of booking an appointment for genetic testing and counselling. Males and respondents with higher depression ratings were less likely to book an appointment at the follow-up assessment.

The finding of gender being a significant predictor of change over time in self-prediction scores is in agreement with the earlier repeated measures GLM analysis testing for utility theory where gender interacting with time was the only significant independent variable.

Of the dummy coded nominal variables representing information group and social class included in the regression, only managerial-professional social class and positive information were significant. Being from an intermediate social class background was not significantly different to being from a working class background. In terms of information, neither the neg-pos nor pos-neg groups were significantly different to the control group, suggesting that balanced information had no significant impact upon self-prediction or intention scores.

Both the pos-neg and neg-pos variables being non-significant in the regression analysis also concurs with the GLM finding of no statistically significant ordering/primacy effect.

There were two other non-significant findings worthy of note. Firstly, not one of the items from the health belief model was significant predictors of change in either self-prediction or intention scores. Secondly, the weighted ratio variable was non-significant. This latter finding confirms the findings from the GLM analysis in hypotheses 4[10] and 5[11] and the correlation analysis in hypothesis 7[12], that utility theory did not predict the hypothetical uptake of genetic testing and counselling in this experiment.

Excluding the 15 respondents (not including the 2 residual outliers) that had extreme change scores compared to the remainder of the respondents on the self-prediction and/or intention scores from the regression analysis was the correct thing to do statistically. Despite the fact that exclusions were necessary to satisfy statistical analysis it remains that 15 respondents changed their mind substantially more than the remaining respondents following the information manipulation. These respondents did not differ significantly on any of the demographic or psychological characteristics from the remainder of the respondents.

In their experiment with a sample of students Cameron and Diefenbach (2001) explained 18% (r^2=0.18) of the variance in the hypothetical intention to obtain genetic testing with the independent variables, cancer worry, positive information, negative information and comprehensive information. Increased worry resulted in greater intention whilst the information categories (positive, negative and comprehensive) had negative coefficient signs indicating reduced intention. Of the four information groups (standard, positive, negative and

[10] Hypothesis 4: Experiment information produces statistically significant change in self-prediction and intention to have testing and counselling.
[11] Hypothesis 5: Positive information will increase self-prediction and intention scores, both pos-neg and neg-pos information will reduce self-prediction and intention scores, and little or no change in scores will occur for the control group.
[12] Hypothesis 7: A large degree of positive correlation (0.5 – 1.0) will exist between weighted ratio scores and self-prediction and intention to have testing scores.

comprehensive), intention to obtain genetic testing was highest amongst the respondents that received the brief standard information. Cameron and Diefenbach proposed that information about the consequences of testing, regardless of its nature (positive or negative), may dampen initially high intention levels. This may be a result of the information leading to careful consideration of the potential implications of testing. Women with higher cancer worry scores appeared to process information relating to cancer genetic testing for breast cancer in a manner that highlighted the utility of testing and its benefits. Cameron and Diefenbach (2001) found a negative relationship between risk perceptions and testing distress beliefs, which was interpreted as a sign that cognitions relating to personal susceptibility may lead to de-emphasising or minimising of emotional reasons against testing.

Clearly there are parallels between the results of Cameron and Diefenbach (2001) and the findings of the current experiment. In both studies risk perception was not a significant predictor of intention, whilst cancer worry and positive information were. Cancer worry also emerged as a significant independent variable in several screening studies including Diefenbach, Miller and Daily (1999), Miller & Miller (1995), and Codori, Peterson, Miglioretti, Larkin, Bushey, Brensinger, Johnson, Bacon and Booker (1999) who found it to be a determinant of genetic testing for colon cancer. There were two major discrepancies between the results of both studies. Firstly, although positive information was a significant independent predictor in both the current and Cameron and Diefenbach's (2001) experiment, its impact upon intention to have genetic testing was completely different in both studies. In the current experiment positive information resulted in increased intention, whilst it reduced intention in Cameron and Diefenbach's experiment. Secondly, in the current experiment neither the pos-neg or neg-pos information significantly affected intention to have genetic testing, whilst Cameron and Diefenbach found their comprehensive (combined neg-pos and pos-neg) and their negative information significantly affected intention. In addition, the current experiment explained 28% of the variance in intention (adjusted R^2=0.280), whilst Cameron and Diefenbach only explained 18%.

The discrepancies between the regression results of the current experiment and those of Cameron and Diefenbach (2001) appear to stem from variations in the design and implementation of the two experimental studies. Firstly, Cameron and Diefenbach did not have a *naïve* control group, all of their participants received standard information upon genetic testing for breast cancer. Secondly, different information was used in both experiments and Cameron and Diefenbach appear to have provided more information; whilst no group in the current experiment received more than two A4 information sheets, Cameron and Diefenbach provided their participants with three to seven pages. Thirdly, Cameron and Diefenbach used a cross-sectional design looking at intention, whilst the current experiment used a repeated measure design looking at change in intention. Finally, the current experiment had a more comprehensive set of potential predictor variables identified from the theoretical and empirical literature in the field.

Self-prediction and Intention Scores

The two behavioural intention measures used in this study, self-prediction and intention showed strong agreement, providing a Pearson correlation coefficient of 0.864 (N=141, p=0.000) at baseline and 0.878 (N=142, p=0.000) at follow-up. However, results were inconsistent upon these two items; self-prediction was more sensitive than intention, identifying a significant difference between male and female respondents between assessments. In the regression analysis six significant predictors were found to be associated with only one of these two items (4 with self-prediction and 2 with intention). The discrepancies in the results comply with the findings of Shepperd et al. (1988) who found that the constructs of self-prediction and intention differed. The findings of Shepperd et al.'s (1988) meta-analysis propose that self-prediction measures are a better predictor of behaviour than intention measures. Differences in the wording of the questions relating to service provision are also likely to have influence discrepancies in results. Whilst Cameron and Diefenbach's (2001) intention items only mention genetic testing, the self-prediction item also offered genetic counselling and made it explicit that the services would be free of charge.

Limitations

As has been noted previously in the methods sections there are some potential limitations to this experiment. Firstly, the experiment was conducted during breast awareness month (October, 2002). Local and national newspapers and national magazines such as The Daily Post, The Welsh Daily Mirror, the Associated Newspapers Group (Femail.co.uk, Daily Mail, Mail on Sunday and Metro Newspapers) and Cosmopolitan magazine were publishing articles and supplements with information upon and references to further information on genetic breast cancer. In order to prevent media coverage and discussion of the issues with others between assessments taking place, both the pre and post information measurement were conducted within one sitting and with all the information groups at the same time. In order to prevent baseline knowledge (and experience) of genetics and/or breast cancer from biasing the experiment a series of screening questions were posed and used to exclude participants with experience and substantial knowledge.

Secondly, conducting both assessments of the experiment in one sitting to combat the effects of the media and discussion between participants raised the potential problem of recall bias; respondents recalling their original responses and answering the second set of questions in light of this. This issue was addressed by preventing respondents from looking at their previous answers and asking them to ignore their original ratings and responses when completing the follow-up assessment. However, this solution could have resulted in the complete opposite effect to that intended. A request to forget baseline responses may have made the responses more prominent in respondents' minds or encouraged them to recall their baseline responses. In addition respondents may have inferred (demand characteristics/ experimental effects) from the request to forget baseline responses that they were supposed to identically reproduce their answers at the follow-up assessment or alternatively provide different responses.

Thirdly, from the outset (see methods, Chapter 3), it has been apparent that the discrete choice questionnaire had some limitations. Due to the total number of scenarios required to conduct a full factorial design and the limited number of respondents available it was necessary to design one questionnaire and not

multiple blocks of questionnaires (blocking) that could be issued to sub-samples. This necessitated the use of a fractional factorial design. The 24 scenarios generated by the Speed 2.1 software were based upon the assumption that there were only main effects and no higher order interactions[13]; a wide spread practice amongst researchers conducting discrete choice studies (Louviere et al., 2000; Viney et al., 2002; Viney, Savage & Louviere, 2005). Subtracting the main effects degrees of freedom[14] from the total degrees of freedom[15] reveals that the potential number of interaction effects (second order and above) equalled 1521. It would be astounding if all of the potential interactions were non-significant. Fortunately, two way and higher order interactions rarely account for a great deal of variance (Louviere et al., 2000). Main effects account for 70-90% of explained variance, two way interactions for 5-15% and higher order interactions account for the remainder (Dawes and Corrigan, 1974).

Fourthly, due to the length of the questionnaire (25 choices including tests) there was a risk of respondents succumbing to boredom or fatigue (San Miguel et al., 2005; Verhoef et al., 1991). Given the low number of missing values and consistent no preference responses in the latter half of the DCM questionnaires it is unlikely that boredom or fatigue had set in and were affected responses.

Fifth, manipulation checks were omitted from the experiment and as a result it is not possible to confirm that research participants interpreted the experiment information in the intended manner e.g. did participants think the positive information was positive etc. However, the information used in this study has been successfully used by Wroe and Salkovskis (1999). In addition, in the event that the information did not have a significant affect on the outcome measures, the design of the experiment would still identify decision making that complied with utility theory. If utility theory were in use hypothesis 7 (a large degree of positive correlation (0.5 – 1.0) will exist between weighted ratio scores and self-prediction and intention to have testing scores) would be confirmed at baseline.

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[13] Main effects are the attributes, whilst higher order effects are interactions between two or more attributes and/or any demographic or experimental variables.
[14] 4(4-1)+(3-1)+(2-1)=15.
[15] $4^4*2*3=1536$.

Sixth, a full experimental design was not used in this experiment as there was not a group issued solely with negative information. A group was not issued with only negative information as the authors of the information (Wroe and Salkovskis, 1999) designed the negative information to be issued with the positive information and negate the positive information.

Seventh, the TPB items used in the experiment were designed by the GenQuest research team and had been successfully used with patients referred to cancer genetic services. The GenQuest team opted to directly measure perceived behavioural control using controllability only. Indirect measurement of perceived behavioural control would have been preferable where both self-prediction of occurrence and perceived facilitating/inhibiting power measures were developed and used. It would also have been preferable to have more questions measuring each of the constructs making up the TPB. However it should be noted that the internal consistency results suggest that the TPB as it was operationalised in this experiment was adequate.

The remaining limitations all relate to the sample of respondents used in this experiment. A sample of students was used in this experiment rather than a sample of individuals with a family history of cancer. A cohort of individuals with a family history of cancer could not be used for two reasons; their knowledge of cancer and genetic cancer could negate the effects of the information manipulation, and it is ethically questionable to manipulate the behavioural intention of an individual to have genetic testing and counselling when they are at increased risk of developing genetic cancer (debriefing has been found to be ineffective in a similar study [Wroe et al., 2000]). However, as the focus of the study was to examine how behavioural intention to have genetic testing for breast cancer and preference (utility) for the service configurations presented were affected by information rather than estimating service uptake by actual cancer genetic patients, conducting the study with students was not as limiting as it may appear at first. Cameron and Diefenbach (2001) point out that laboratory studies looking at the implications of health information and psychosocial factors on behaviour perceptions e.g. Croyle & Williams (1991),

Gintner, Rectanus, Achord and Parker (1987), are based on the principal that information processing effects will be similar across socio-demographic groups.

As the sample comprised undergraduate students, the experiment participants were highly educated (A levels or equivalent qualifications), were predominantly female, young and from managerial and professional backgrounds. However, there were sufficient mature students to detect an effect of age in the regression analysis, and over representation by well educated women from professional backgrounds at familial breast cancer centres is common (Steel et al., 1999). Finally, the sample size was adequate to find a moderate effect size with an α of 0.05 or less with 80% power. Clearly, small effect sizes could not be detected and as a result there may be significant relationships between dependent and independent variables that have been missed in this experiment.

Survey of Patients Referred for Cancer Genetics Services

Patient Preferences

High risk breast cancer patients ignored the attribute availability of testing (non-significant) as they are eligible for testing. They were reminded of their eligibility for testing in the attribute descriptions provided in the questionnaire. Respondents would prefer to receive counselling from a consultant geneticist, but they were willing to accept counselling from a genetics associate. These findings support the increasing use of clinical genetics nurse specialists and genetics associates in genetic counselling (ACOGT, 1998a; Harper & Clarke, 1997) in response to the scarcity of qualified consultant clinical geneticists (Royal College of Physicians, 1991; GRAG, 1995).

Willingness to Pay (WTP)

The multinomial logit regression results for the breast cancer patients have significant coefficients for the cost of service attribute level of £3,000. This signifies that the respondents placed this value upon cancer genetics services. Patients were willing to pay in the form of a single payment to the NHS or a private provider, a one-off tax bill or national insurance contributions. The value of £3,000 actually exceeds the mean estimated cost to the NHS of £2,510. As

£3,000 was the largest attribute level presented in this study it is possible that patients place a higher WTP value on cancer genetics services than £3,000. These results add to the body of evidence accumulating upon the acceptability and value of adding WTP attributes (cost/out of pocket expenses) in appropriate DCM studies (Hall, Viney, Haas & Louviere, 2004; Hanson, Barbara, McPake, Nakamba & Archard, 2005; Maddala et al., 2003; Ratcliffe, 2000b; Ryan 1999b; Ryan, 2004b; Ryan & Hughes, 1997; Ryan & Skatun, 2004; San Miguel et al., 2000; Sculpher, Bryan, Fry, Winter, Payne & Emberton, 2005; Skjoldborg & Gyrd-Hansen, 2003).

Cost-Consequences Analysis

High risk breast cancer patients engaged in compensatory decision making i.e. whilst they would prefer to be counselled by a consultant geneticist they were willing to see a genetics associate rather than a consultant, particularly if the remaining attributes were more favourable. Four genetics associate led service scenarios were in the top quartile, with the service configuration scenarios ranked 7, 8 and 10 favoured over 26 scenarios where counselling was provided by a consultant. For example, scenarios 10 and 11; patients would prefer to be counselled by a genetics associate for 1 hour rather than a consultant for 2 hours when the waiting time was the same at 1 month but travelling distance to counselling was 40 miles rather than 60 miles.

The results show that not only did counselling by a genetics associate accompanied by favourable levels of other attributes provide high utility, but also substantial cost savings. For example, the scenarios ranked first and seventh have the most favourable levels of the attributes waiting, distance and duration, but the first has a consultant and the seventh has an associate giving counselling. The scenario ranked seventh was £790 cheaper per presymptomatic patient than the scenario ranked first. In terms of cost-consequences the scenario ranked tenth (in terms of utility/patient preference) emerges as the most desirable scenario configurations in the author's opinion. The scenario (genetics associate, 1 month, 40 miles, 1 hour) achieved one of the highest utility/desirability rankings and at a cost of £2,525; scenario estimated costs ranged between (£2,027 and £3,645). Clearly there is substantial advantage to be gained in using genetics associates to

provide counselling under appropriate circumstances rather than consultants as they provide substantial cost saving.

The overall results of this study are similar to those of Wilson et al. (1999) who found that utility increased as duration of counselling appointments increased, and distance to appointment and waiting time declined. Additionally they found genetics nurse and genetics associate led counselling to be cost saving.

Given the way in which Wilson et al. (1999) implemented their study it is surprising that the overall results comply with those reported here. Respondents in Wilson et al.'s study were not offered the opportunity of stating that they had no preference for any of the paired scenarios presented to them. Interval and ordinal codes were used rather than the effects codes used in the current study. As a result Wilson et al. cannot be sure that all of the levels of statistically significant attributes are actually significant to patients.

Wilson et al. (1999) explained substantially less variance in patients' preferences/utility function (adjusted McFadden's R^2=0.0004 to 0.002) than the current study (adjusted McFadden's R^2's of 0.25799). This is a result of the current study including two additional attributes (cost of service and availability of testing – only the former was significant to high risk patients) and a no preference response option.

There are substantial differences in the costs reported by Wilson et al. (1999) and those reported here. This is predominantly a result of Wilson et al. looking at a single counselling session and the current study looking at a more intensive protocol offering multiple counselling sessions and genetic testing to high risk patients.

No detail was given by Wilson et al. (1999) on the assumptions made when producing cost estimates for the comparison of service provision based upon patient preferences for significant service attribute levels. Based upon the results presented it would appear that little to no provision was made in the costs to allow for the attributes of distance to counselling and waiting time. For example,

for the Aberdeen clinic a counselling session with a doctor for 45 minutes with a distance travelled to counselling of 1 mile and a waiting time of 4 months incurs a cost per patient of £79.79, whilst a counselling session with a doctor for 45 minutes with a distance travelled to counselling of 5 mile and a waiting time of 8 months incurs a cost of £79.72. A £0.07 additional cost for a reduction of 4 miles in travel and 4 month waiting time suggests that travel costs, capital, labour and overheads costs were underestimated.

Finally, Wilson et al. (1999) established that there was financial benefit in using genetics nurses and associates rather than consultants by conducting cost utility analysis. To do this a cardinal (interval value) utility measure must be divided into the cost of the event pathway for which the utility value is derived. As has been pointed out in Chapters 2 and earlier in this Chapter, random utility theory (DCM) is a strength of preference measure; a preference measure that is wholly ordinal, providing no more than ordinal ranking of choices (Schoemaker, 1982). The multinomial logit regression coefficients calculated in the current study provide support for Schoemaker's assertions. Bases upon the current evidence it appears that Wilson et al. should not have conducted cost-utility analysis as it is wholly inappropriate to do so.

Limitations

In interpreting the results of the survey of CGSW patients, readers should bear in mind the following limitations. Firstly, the measurement of labour for clinical and administrative tasks was based upon the stated responses of seven members of staff to an administered questionnaire. Secondly, the costs in this study are based upon a service working at full capacity, in the unlikely event that demand for cancer genetic services declines (Ponder, 1999) the cost per patient would obviously rise in accordance with the decline in demand. Thirdly, all mutation screening in this study was conducted using medium throughput technology. Smaller laboratories that do not have access to such labour saving devises would incur greater labour and capital costs per test and the converse would be the case for larger laboratories with more throughput capacity. Despite the limitations noted above, the cost estimates derived in this study are representative of the protocol used and are generalisable to similar services and settings. It should be

noted that Lerman (1997) has questioned the appropriateness of counselling protocols such as the one in this study as they are derived from Huntington's disease protocols. Protocols with extensive pre-test assessment; initially designed to identify depression and suicidal potential.

Fourthly, as the same DCM questionnaire was administered in both the patients' survey and information manipulation experiment, the limitations noted above in relation to the DCM questionnaire are equally applicable to the patient survey.

Fifth, the information manipulation experiment found that random utility theory was not being used. If preferences are not grounded in RUT then the validity of the DCM results are in question.

Sixth, the DCM questionnaire was issued to patients post their clinical risk assessment results. This is an early stage in the care process for high risk patients (Griffith et al., 2005). Clarke et al. (1996) recommend elicitation of patient views on completion of the care process or once they are sufficiently informed to make a judgement. However, Hundley and Ryan (2004) and Ryan et al. (1998) found that experience and knowledge of the availability of certain aspects of care influence patient preferences and patients report preferences for current/available service provision.

Finally, although the results of this study do provide some interesting insight into patient preferences (bearing in mind that the validity of DCM is in doubt) and their potential use in conjunction with cost estimates for clinicians, service providers and policy makers; this study has the limitation of a small sample of patients from which choices were elicited. Given this limitation, caution must be exercised in interpreting these results.

CHAPTER 7: CONCLUSIONS

A systematic review of the health economics literature revealed that economic evaluations of cancer genetic services have concentrated upon health outcomes and mutation identification, resulting in a paucity of research upon patient preferences. There is a need to account for patients' views in health care provision, extending the range of outcome measures considered beyond health outcomes to non-health outcomes and aspects of process when they form part of patients' preferences (utility function). Given the finite resources available to service providers and planners it is necessary to ascertain preference data, as is advocated by the NHS and DOH, within an opportunity cost framework, allowing policy makers and service providers to tailor services to deliver maximum utility (benefit) to patients within budgetary constraints. In addition, the Cancer Genetics Service in Wales (CGSW), one of only two national NHS funded services in the UK, which unlike its Scottish counterpart offers a substantially more resource intensive service (including genetic testing to high risk patients), had not undergone a preference based economic evaluation. In light of this evidence the first research question was set for patients of the CGSW. Research question 1, "What are the attributes of cancer genetics services that are important to high risk patients (the patients spending the most time in contact with the service and receiving most services i.e. genetic testing and counselling)? and what would be the cost of providing the service to comply with patient preferences?" was operationalised into empirical aim 1 below.

Empirical Aim 1: Ascertain the aspects of cancer genetics services that are important to patients, and present service configurations prioritised in terms of preferences accompanied by their costs (cost-consequences analysis) for high risk patients.

Discrete choice modelling emerged in the 1990s (Verhoef et al., 1991; Cave et al., 1994; Ryan, 1996a) as potentially the most appropriate technique to apply to

the issue of ascertaining patient preferences. DCM has been applied to numerous health care issues and shown considerable potential as a preference elicitation technique to inform health policy and service provision. This technique offers advantages over alternative methods, incorporates realistic service attributes and levels (health, non-health, process [including charges/WTP]), continuous or nominal/qualitative in nature and presents them in a straight forward choice format which is easy to understand. The choice format also allows opportunity cost to be incorporated into the decision process. The analytical results provide information on whether or not attributes and levels are significant to individuals, provides information upon the strength of preference for attribute levels and complete health care scenarios, and the relative importance of attribute levels and complete scenarios.

Despite the favourable findings in the published literature, there is limited evidence in relation to the reliability and validity of DCM as a research technique in the field of health care. In particular no one had experimentally tested for evidence of utility maximisation decision making amongst respondents of DCM questionnaires. This necessitated research question 2 to be set. Research question 2, "Do respondents of DCM questionnaires make choices in accordance with Random Utility Theory?" was operationalised into empirical aim 2 below.

Empirical Aim 2: Experimentally examine respondents of a DCM exercise by means of an information manipulation to see if they are adhering to DCM's underlying decision theory principals of Random Utility Theory.

Aim 2 was addressed first in the thesis. Although the multinomial logit regression coefficients conformed to expectations, suggesting that utility maximisation was taking place in respondents' decision making, the primary conclusion of the experiment was that respondents *were not* adhering to DCM's underlying decision theory principals of random utility theory. Utility maximisation and as such random utility theory *did not* explain respondents' weighted ratio of pros to cons, hypothetical intention to have genetic testing or

self-prediction of having testing and counselling. A finding that is in keeping with the findings of the 'Utility: Theories, Measurement and Applications' conference (Edwards, 1992); where the leading academics working in the field of normative, descriptive and prescriptive utility theory concluded that utility maximisation the primary axiom of utility theory in its various forms (EU, SEU, random utility and other generalised utility theories) was indefensible as a descriptive model of decision making (Edwards, 1992).

Despite the agreement between the experiment findings and Edwards (1992) findings, it is not possible to conclude that RUT is not being used by individuals in their decision making. A number of potential limitations of the information manipulation experiment cannot be categorically rejected. In particular the preponderance of questions concentrating on negative issues may have affected responses. The repeated success of Wroe and colleagues (Salkovskis et al., 1999; Wroe & Salkovskis, 1999, 2000; Wroe, Salkovskis & Rimes, 1998; 2000) in finding results that confirm and comply with utility theory with a range of diseases including genetic breast cancer, under survey and experimental conditions, with a range of participants (students, the general public, individual contemplating accessing genetic services and individuals eligible for bone density screening) provide strong contradictory evidence of the findings of the experiment outlined in this thesis.

Although RUT cannot be rejected the results of the current experiment do cast further doubt upon its merits as a descriptive model. Given that utility theory underpins many of the techniques used in economics and health economics such as time trade off, standard gamble and in particular discrete choice modelling, there is clearly a need to conduct further research into RUT as soon as possible. Until the uncertainty relating to the descriptive validity of RUT is resolved, all analytical techniques based upon it are potentially invalid. All the DCM results reported in this thesis, including the results from the survey of high risk patients referred to the CGSW (research question 1/aim 1), are therefore potentially invalid and should be treated with caution. "Generally speaking there can be no

valid measurement without an underlying theory of the behaviour of the numbers which result from measurement. Thus, ... measurement in the absence of theory is at best uninterpretable and at worst meaningless" (Louviere et al., 2000, p25).

Empirical Aim 1: Ascertain the aspects of cancer genetics services that are important to patients, and present service configurations prioritised in terms of preferences accompanied by their costs (cost-consequences analysis) for high risk patients.

All the attributes selected from the literature and tested in this study were found to be significant to high risk patients of the CGSW apart from availability of testing which was included for low and moderate risk patients (reported elsewhere). The six attributes tested were: 'staff seen for counselling', 'waiting time for a letter about risk of cancer', 'distance to counselling', 'duration of counselling', 'availability of testing' and 'cost of service'. Although high risk breast cancer patients would prefer to be counselled by consultant geneticists, genetics associates were acceptable as counsellors. Patients preferred shorter waiting times, longer consultations and shorter travelling distances. Patients were willing to pay £3,000 for genetic serviced, which exceeds the current estimated cost of providing testing and counselling.

The results of the cost consequences analysis (see table 5.26) show that not only did counselling by a genetics associate accompanied by favourable levels of other attributes provide high utility but also provide substantial cost savings. In terms of cost-consequences the scenario ranked tenth (in terms of utility/patient preference) emerges as the most desirable scenario configurations in the author's opinion. The scenario (Genetics associate, 1 month, 40 miles, 1 hour) achieved one of the highest utility/desirability rankings (ranked 10th) and at a cost of £2,525; scenario estimated costs ranged between (£2,027 and £3,645). Clearly there is substantial advantage in using genetics associates under appropriate circumstances rather than consultants as they provide substantial cost savings. These findings support the use of genetics associates and other suitably trained

clinical staff for genetic counselling in response to the scarcity of qualified consultant clinical geneticists. The savings obtained from such a service configuration could be used to fund improvements in the service such as more staff (clinical and administrative) to reduce the waiting time between receipt of referrals and issuing patients with a risk assessment or be used in relation to other attributes or completely different health services.

In addition to the main study aims, the thesis has raised other service development and policy issues and methodological research issues. There was an under representation of ovarian, breast ovarian, colorectal patients, men, patients with lower educational attainment and from lower socio economic groups presenting to the CGSW. This situation needs to be rectified and one or more health promotion programs may be in order to inform patients and referrers that this service is available for individuals with a family history of cancer.

Reluctance upon the part of males to attend cancer genetics services was also found in the information manipulation experiment. Regression analysis upon change in self-prediction scores identified eight significant predictor variables; baseline self-prediction score, positive information, attitudes towards behaviour, three demographic variables (age, sex and managerial-professional social class) and two emotions (anxiety about developing cancer and depression). The sign of the regression coefficients revealed that a positive attitude towards health behaviour, being older, increased anxiety about their risk of developing breast cancer, coming from a managerial or professional social class and being issued with positive information, all increased respondents self-prediction of booking an appointment for genetic testing and counselling. Males and respondents with increased depression ratings were less likely to book an appointment at the follow-up assessment. Multiple linear regression upon change in respondents' intention to have genetic testing scores identified six significant predictor variables; baseline intention score, positive information, sex, and the health cognition variables of subjective norm, attitudes towards behaviour and cancer worry. Increased cancer worry, increased desire to comply with subjective norm,

a positive attitude towards health behaviour and being issued with positive information all increased the respondents' intention to have genetic testing. Males were less likely to book an appointment at the follow-up assessment.

As all information and counselling is delivered strictly within the edict of nondirective counselling it would be ethically unacceptable to use the results of the regression analysis to influence the uptake of genetic cancer services. However, these results can be used as a checklist against which cancer genetics services can compare the information they issue to potential service users. For example, the results of this experiment show that males and younger adults are less likely to access services. This may be a result of the information being issued in the experiment being perceived as more relevant to females and to older adults. However, the risks (prevalence and penetrance) are equally high for both genders and sufficiently high to merit careful consideration from young adults (Struewing et al., 1997). Genetics centres may wish to check their information and if possible uptake rates to see if they have such an imbalance in their information. Research may be needed before any modifications can be included with confidence by genetics centres.

Several methodological research issues emerged from the thesis. In both the information manipulation experiment and the survey of patients of the cancer genetics service in Wales strong evidence of construct validity was found in the form of concurrent and theoretical validity for the discrete choice questionnaire used. The DCM questionnaire was also found to be acceptable, realistic and relatively easy to answer.

Although there were significant differences between the responses and characteristics of respondents that passed and those that failed the axiom tests of random utility theory in the experiment[1], current evidence suggests that they must be included in the analysis. Failure to include all respondents will result in

[1] Similar findings were found for in the patient study but omitted from the thesis in the interests of brevity.

research findings that cannot be generalised to the population of interest, and as a result service provision and policy making will be based upon inaccurate evidence.

In both the experiment and the patient survey, including an indifference response option was found to be a valuable addition to the response options offered in the DCM questionnaire. Effects coding was found to be superior to dummy, ordinal and interval coding. Multinomial logit regression results complied with Schoemaker's (1982) interpretation of utility theory, namely that the results of utility theory and as a result DCM provide ordinal ranking of respondents' utility. This casts doubt upon the validity of the practices of calculating marginal rates of substitution and using regression coefficients to calculate cost-utility ratios.

Merits & Originality

Despite the limitations noted in Chapter 6 and the potential improvements proposed below, there are a number of original features to this research. The systematic review conducted in Chapter 2 to ascertain what is currently known about cancer genetic services in the field of health economics is the first systematic review in the field and was published in the peer reviewed journal the British Journal of Cancer in 2004 (Griffith et al., 2004). The micro costing conducted is the first full micro costing of a commonly used cancer genetic counselling and testing protocol and was published in the British Journal of Cancer in 2005 (Griffith et al., 2005).

The discrete choice analysis conducted with patients of the Cancer Genetics Service in Wales (Chapter 5) is the first with cancer genetics patients outside of Scotland, the first to look beyond a single counselling session and the first study to employ DCM in a cost-consequences study. It has identified health policy and health promotion issues in relation to the need to address the inequity of which members of society are accessing cancer genetic services in Wales. Additionally, the cost-consequences analysis has provided valuable data for service providers,

clinicians and policy makers on high risk genetic breast cancer patients' preferences (bearing in mind the doubts raised about the theoretical validity of DCM in the information manipulation experiment) and the accompanying costs for a service similar to the Cancer Genetics Service in Wales. This data will allow service providers to gage the cost and acceptability of introducing alterations to an existing service and the cost and acceptability of setting up a new cancer genetics service.

Both the patient study and the information manipulation experiment have highlighted methodological issues in the field of DCM. A strong case is made for including indifference response options, using effects coding and including all respondents in the analysis of DCM studies. Of particular importance is the evidence suggesting that the common practice of conducting marginal rates of substitution is invalid as is using DCM coefficients to conduct cost-utility analysis.

Finally, and perhaps most importantly, the information manipulation experiment conducted incorporates contemporary work in health psychology with health economics to strengthen the theory underlying discrete choice modelling as it is currently used in health economics. It is the first experiment testing the implicit assumption applied in discrete choice modelling that respondents are using random utility theory to make their decisions. Additionally, in order to go beyond the weighted benefits and barriers used in RUT/DCM and assess in more detail the determinants of choice; the experiment incorporated the social/health psychology theories of the theory of planned behaviour (TPB) and components of the health belief model (HBM).

Future Research

In terms of improving upon the research looking at patient preferences for the delivery of cancer genetic services there are a number of potential improvements. Firstly, a larger sample of patients resulting in increased statistical power would

be essential. To account for the debate relating to when preferences should be elicited from patients a cohort design should be used with assessments conducted as patients progress through the cancer genetics service. The discrete choice questionnaire used could be designed to allow for interaction effects and by pairing scenarios in terms of minimum overlap and utility balance it would be possible to present respondents with fewer paired choice questions. In terms of estimating maximum willingness to pay using a DCM questionnaire, the attribute levels should be increased to see how much more than £3,000 respondents are willing to pay. Alternatively, as the WTP attribute has been proven to be a practical attribute in a DCM study with UK patients (who do not pay for such NHS services) in this study, it could be excluded in favour of another attribute.

In this study all the attributes tested were identified from the literature. The attributes identified but excluded from this study should be considered for use in future DCM research with patients of cancer genetic services. In addition alternative methods of attribute identification should be employed such as focus groups with staff and patients of cancer genetics services.

In terms of establishing the costs of cancer genetic services, ideally the service under consideration will have multiple sites using the same clinical and laboratory protocols, allowing differences by cancer genetic centre to be allowed for. Labour input for the costing should be measured by means of a time and motion study.

In terms of improving upon the information manipulation experiment there are five main ways in which this research could be improved upon. Firstly, to account for the potential bias from media coverage and to account for different types of choices, a series of experiments should be conducted based upon different health decisions. Ideally a mix of minor to very serious health care choices should be used. Respondents should be randomised and different DCM questionnaires issued to them. Secondly, at least a month should be left between administration of the pre and post information questionnaires. Thirdly, care

should be taken to ensure that the psychosocial questions and wording of the DCM questionnaire do not influence respondents i.e. lead them to concentre on negative or positive issues, which will invalidate the information manipulation. Fourthly, a representative sample of the general public should be used. This would enable the results of the experiment to be generalized to a wider population; the current experiment can only be generalized with confidence to young well educated females from higher socioeconomic backgrounds. Finally, the sample size should be increased so that small effect sizes can be detected.

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APPENDICES

Appendix A. Methods of obtaining user preferences

Opinion Polls

In this context an opinion poll requires respondents to rank attributes in order of preference to them.

Patient Satisfaction Surveys

These studies take a variety of formats with the specific intention of eliciting satisfaction with a service as a whole and its attributes.

Willingness to Pay

Willingness to pay or contingent valuation directly asks for the maximum amount of money that individuals are willing to pay for an item or service, deriving a utility value in monetary terms (Donaldson et al., 1995). WTP provides a complete benefit measure, a valuation that encompasses all the characteristics or preferences that are important to the individual in relation to the service (Ryan, 1996b; Phillips, Homan, Luft, Hiatt, Olson, Kearney & Heard, 1997).

The following three methods are used to establish Quality Adjusted Life Years (QALYs). A measurement of health status incorporating mortality and morbidity on a single continuum and weighting life years gained from an intervention by quality of life (Bowling & Jones, 1997; Drummond, 1995).

Visual Analogue

Conventionally this technique has consisted of presenting respondents with health states and requesting that respondents mark the visual analogue scale (range 0 to 1), indicating the order and preference that respondents have for the health states.

Standard Gamble

Respondents are asked to choose between a certain health state e.g. remaining in their current health state, and being offered an intervention that can improve their health but with a possibility of deteriorated health or death. Levels of probability for a successful outcome (p) and an unsuccessful outcome (1-p) are varied until respondents indicate that the benefit is no longer worth the risk (Bowling & Jones, 1997; Ryan, 1996a).

Time-trade Off

Respondents are asked to consider being in a certain state of health for a defined period of time. They are offered a health intervention that will give normal or improved health for a shorter time period and followed by severe disability or death. Time and health are varied until respondents feel that there is no longer a gain in reducing time in good health in exchange for that quality of health. A variant of this method includes asking respondents to consider the number of individuals in a given health state (A) that need to have improved health to be equivalent to improved health for one person in health state (B) (Bowling & Jones, 1997).

Appendix B. Statistical analysis

Examination of data prior to analysis

All data was visually examined by means of frequency counts. For continuous (ordinal, interval or ratio) data stem and leaf plots, normality plots and box plots were also examined. When assessing the normality of a distribution of scores on a continuous variable three procedures were used. Firstly visual examination of the output listed above, secondly conducting Kolmogorov-Smirnov and Shapiro-Wilks tests, and finally examination of Z scores for Fisher's skewness coefficient and Fisher's coefficient of Kurtosis (Pett, 1997) [Skewness coefficient = skewness/ standard error of skewness, Coefficient of Kurtosis = kurtosis/ standard error of kurtosis]. The final procedure was only used when the Kolmogorov-Smirnov and Shapiro-Wilks tests reject normality. It is common for goodness of fit tests such as the Kolmogorov-Smirnov and Shapiro-Wilk tests to reject the null hypothesis when the sample size is relatively large (Norusis, 1994). If the Fisher's skewness coefficient and Fisher's coefficient of Kurtosis are between ±1.96 (critical value of two tailed z statistic at α=0.5) the variable is considered to be normally distributed.

Serious violation of normality required a nonparametric inferential test to be used rather than a parametric test (inferential tests are discussed below) when a bivariate test was appropriate or for multivariate tests the variable would need to be transformed. In the case of a dependent variable the appropriate power for transforming the data would be determined by subtracting the slope of the least-squares line from 1, which is provided by a Spread-Versus-Level Plot of the data (log of the median against the log of the inter quartile range).

Equality of variance was assessed for the t tests, one-way ANOVA, and Univariate GLM with Leven's test of homogeneity of variance. Box's M was used for the multivariate GLM.

Cronbach Alpha

The internal consistencies (reliability) of the items in an index or composite score were assessed using Cronbach's Alpha; the best measure of internal consistency (Kline,

1993). Alpha coefficients range between 0 and 1. A score of 1 indicates that all items in the composite score are measuring exactly the same construct.

Correlation

The degree of the linear relationship between two variables measured on a continuous scale was assessed using the Pearson's product moment correlation coefficient. In the event that one or both variables were skewed Spearman rank order correlation would have been used (this was not necessary). As in essence the later technique is merely conducting a Pearson correlation upon the rank order of data (Hinton, 1995) the results of both tests are extremely close if not identical. Both tests provide a correlation coefficient between -1 and +1 which represents the strength of the linear association between the variables. Cohen (1988) suggests that correlation coefficients of 0.1 to 0.29 are small, 0.30 to 0.49 are medium and 0.50 to 1.00 are large. A positive sign indicates a positive relationship with both variables increasing in relation to each other, whilst a negative sign indicated that an increase in one variable is associated with a decline in the other variable.

One sample t-test

To compare the mean response of a single sample with a population value e.g. an established population norm, a one-sample t test was used. The dependent variable under consideration must be recorded at least at an ordinal (semi-interval) level and be normally distributed.

Independent Samples

Chi-square.

To ascertain if a statistically significant difference existed between two or more independent samples or groups upon nominal level data a Chi-square test was used. Results were only considered reliable when there was more than one degree of freedom if less than 20% of cells had an expected value less than 5 (Pett, 1997).

Tests for two independent samples (continuous data.)

Independent samples t test, Wilcoxon-Mann-Whitney U test, Medians test.

The Independent-samples t test was used to establish independence between two mutually exclusive samples (dichotomous independent variable) if the dependent variable under consideration was recorded at least at an ordinal (semi-interval) level and was normally distributed. In the event that the dependent variable was skewed for the independent samples but the distributions were of a similar shape the Wilcoxon-Mann-Whitney U test (Mann & Whitney, 1947; Wilcoxon, 1945) was used. Similarity of distribution for two samples with data at ordinal to interval level was examined visually and using the Kolmogorov-Smirnov two-sample test (Daniel, 1990). In the event that the underlying assumptions of the Wilcoxon-Mann-Whitney U test could not be fulfilled the Median test (Conover, 1980) was used. The assumptions of the Median test are that the dependent variable is at least measured at the ordinal level and the categories of the independent variable are mutually exclusive.

Tests for K independent samples (3 or more).

One-way analysis of variance, Kruskal-Wallis one-way ANOVA by ranks test and the Median test.

One-way analysis of variance (one-way ANOVA) was used to establish independence between three or more independent samples if the dependent variable under consideration was recorded at least at an ordinal level, was normally distributed for the samples and the variance of the samples were equal (SPSS Inc., 1996). If the assumptions of the one-way analysis of variance were not satisfied but the data for each sample was similarly distributed with the possible exception of a difference in measures of central tendency of at least one of the samples, the Kruskal-Wallis one-way ANOVA by ranks test (Hinton, 1995; Pett 1997) was used. In the event that a Kruskal-Wallis test could not be conducted a Median test was used.

GLM univariate.

Univariate GLM was used to look for significant change in a single continuous dependent variable by multiple independent variables, both continuous and nominal. The dependent variable should be normally distributed and have equal variance. As is the case for t tests and one-way analysis of variance GLM is robust to moderate violation of these assumptions (any violations are reported in the analysis section).

Repeated Measures

Two time points and no independent variables.

Dependent t test, Wilcoxon signed ranks test and Sign test.

To compare results from pre to post experiment for a single sample the paired comparison or dependent t test was used. The matched dependent variables must be normally distributed. In the event that the data was skewed but difference scores were symmetrical about the median a Wilcoxon signed ranks test (Wilcoxon, 1945) would be used. In the event that difference scores were not symmetrical about the median a Sign test would be used.

Two time points and independent variables.

GLM repeated measures.

Prior to conducting repeated measures GLM baseline differences in the outcome measures for the independent nominal variables were examined using an independent t test or a one-way analysis of variance. In the event of a significant baseline difference, the baseline scores of the dependent variable were subtracted from the follow-up scores to create a new variable that was free of the baseline bias. This variable was then used in a univariate GLM or multivariate GLM as appropriate. Prior to conducting repeated measures GLM (or a multivariate GLM) data was also examined for other examples of covariates e.g. a continuous variable which differed significantly by one or more of the nominal independent variables and was linearly related to the dependent variable or variables. None were found; had there been any, it would have been necessary to

establish that there was homogeneity of regression slopes for the dependent variable upon the covariate for the independent nominal variables.

Repeated measure GLM (previously repeated measure analysis of variance/covariance or repeated measure multiple analysis of variance/covariance) was used to assess change in one or more continuous dependent variable over time. In this case change was examined in relation to multiple independent variables (continuous and nominal). The dependent variable(s) should be normally distributed, although the test is robust to moderate violation of this requirement.

Repeated measures GLM analyses the data and provides separate results using two approaches, univariate and multivariate (SPSS, 1999). In this thesis the univariate results are used. In the majority of cases results are similar if not identical for both approaches. The univariate approach (split-plot or mixed model approach) requires that measurement data should be a sample taken from a multivariate normal distribution, and the variance-covariance matrices are the same for the between-subject effects. The F statistics used in the univariate approach can be assured if the variance-covariance matrix is circular (Huynh & Mandeville, 1979). This assumption is tested by means of Mauchly's test of sphericity and not the Levene test as in the univariate GLM. If the significance value of the test is greater than 0.05, sphericity is assumed. If the significance value of the test is less or equal to 0.05 the numerator and denominator degrees of freedom are multiplied by epsilon to validate the F statistic (done by SPSS 11.5). Three epsilon values and their respective F scores are produced by SPSS, the Greenhouse-Geisser, the Huynh-Feldt and the Lower-bound. In this thesis when sphericity could not be assumed the most conservative of these estimates was used, the 'lower-bound'. Like the univariate approach the multivariate approach requires that measurement data should be a sample taken from a multivariate normal distribution and the variance-covariance matrices are the same across the cells formed by the between-subject effects. This is tested in the case of the multivariate approach by Box's M test. If the significance value of the test is greater than 0.05 homogeneity of variance can be assumed.

Regression of Discrete Choice Data

To establish utility (preference) from discrete choice data, discrete choice regression models were used. Choices must be discrete for each of the multiple sets of scenarios presented to respondents (summing to one). Initially a discrete choice multinomial logit model was specified and a Hausman test of the independence of irrelevant alternatives (IIA) was conducted as IIA is a tenate of the multinomial logit model (Greene, 1997). If this test failed or it could not be calculated as the difference matrix was not positive, the alternatives to the multinomial logit model were run and compared to the original model. The alternatives to the multinomial logit model available on Limdep 7.0/Nlogit 2.0 software are the heteroscedastic extreme value model, the random parameters mixed logit model and the multinomial probit model. Attribute levels with a p value less than 0.1 ($p<0.10$) were retained as significant predictor variables. This is the same criteria as was used by Scott (2001; Scott, Watson & Ross, 2003) and when linear regression with backward elimination or GLM analysis were conducted.

Tests for independent samples.

There are two methods of establishing if the utility function of a discrete choice model (multinomial or bimodal) is significantly different for two or more independent samples; the Wald chi-square (Wilson et al., 1999; Ratcliffe et al., 2002) and the likelihood ratio test (Hanson et al., 2005; Maddala et al., 2003; Ratcliffe et al., 2004 ; Scott, 2001; San Miguel, Ryan and Scott, 2002; San Miguel et al., 2005). For the former method, all attributes are interacted with the independent groups/samples of interest. A Wald Chi-Square test can then be run by group or by attribute for each group e.g. Wilson et al. (1999). The alternative is to calculate discrete choice models for each of the samples or levels of the independent variable and compare them collectively to the un-segmented model using a likelihood ratio test[1]. Like the Weald test the null hypothesis is that both models (segmented and un-segmented) will have the same preference/utility. In this instance the likelihood ratio test value is calculated manually using the formula: 2(log likelihood function [un-segmented model] – Σ log likelihood function [segmented models]). The likelihood ratio test value is then looked up on a Chi-square distribution Table for the appropriate degrees of freedom. The likelihood ratio test has been used in

[1] For a detailed discussion of the likelihood ratio test see Felsenstein (1981), Huelsenbeck and Crandall (1997), Huelsenbeck and Rannala (1997) and Swofford, Olsen, Waddell and Hillis (1996).

this thesis due to the relative ease of calculation and the increased confidence in its results. Howell (1997) noted that as a guide of how well a variable predicted the dependent variable in multiple regression that questions have been raised about the Wald criteria, Howell also pointed out that Hosmer and Lemeshow (1989) favour the likelihood ratio test over the Wald test.

DCM repeated measures.

As there is currently no established inferential statistical test to examine repeated measures DCM results, an analogy of the Fisher's skewness coefficient and Fisher's coefficient of Kurtosis (Pett, 1997) tests has been used as a guide of substantial change in DCM regression coefficients between baseline and follow-up assessments. **Please note the term substantial change is used and not statistically significant change**. An approximate Z score (approx. Z) is calculated by subtracting the baseline coefficient from the follow-up coefficient and dividing the result by the baseline coefficient's standard error. If the resulting approx. Z score is greater than ±1.96 (critical value of two tailed z statistic at α=0.5) the change in the coefficient is considered to be substantial.

Multiple Linear Regression

Scatter plots of the dependent variable by each of the independent variables were used to confirm that a linear model was appropriate and to identify possible outliers. As regression was used to test relationships in accordance with established academic theories e.g. HMB etc., a sequential entry method was used for the independent variables. To establish which independent variables were significant predictors the backward elimination method was used to eliminate irrelevant independent variables; variables with a p value greater or equal to 0.10. Collinearity statistics (tolerance, variance inflation factor and the condition index in particular) were assessed for the final models to ensure that there was not strong overlap between predictor variables leading to an unstable model. A predictor variable with a low tolerance value (close to 0), a high variance inflation factor and a high condition index would be considered for removal. A condition index of 15 or more indicates a possible problem and a score of 30 or more suggests a serious problem (SPSS Inc., 1996). To satisfy the assumptions of

linear regression that errors were normally distributed with a mean of zero, errors had a constant variance and errors were independent of each other the residuals of the final models were checked and any outliers were removed.

Appendix C. Von Neumann and Morgenstern's axioms

Von Neumann and Morgenstern's axioms (Von Neumann & Morgenstern, 1947):

Complete ordering

1. Completeness of the system of individual preference.

$U = V, U > V, U < V$

2. Transitivity of preference.

$U > V, V > W \therefore V > W$

Ordering and combining

3. If V is preferred to U, then even a chance $1 - \alpha$ of V alternative to U is preferable.

$U < V \therefore U < \alpha U + (1 - \alpha)V$

4. The converse of 3 is true.

$U > V \therefore U > \alpha U + (1 - \alpha)V$

5. If $U < W < V$ then if the probability of obtaining U or W are small enough they will not affect W's desirability. For example: Despite the desirability of V its influence can be made as weak as desired by giving it a sufficiently small probability of occurrence.

$\alpha U + (1 - \alpha)V < W$

6. The converse of 5 is true.

$\alpha U + (1 - \alpha)V > W$

Algebra of combining

7. It is irrelevant in which order the constituents of a combination are made.

$\alpha U + (1 - \alpha)V = (1 - \alpha)V + \alpha U.$

8. It is irrelevant whether a combination of two constituents is obtained in two successive steps (firstly the probabilities α and $1 - \alpha$ and secondly β and $1 - \beta$) or in one operation (probabilities γ and $1 - \gamma$, where $\gamma = \alpha\beta$).

$\alpha(\beta U + (1 - \beta)V) + (1 - \alpha)V = \gamma U + (1 - \gamma)V$

Appendix D. Subjective Expected Utility Theories

Table D1

Subjective Expected Utility Theories in Chronological Order

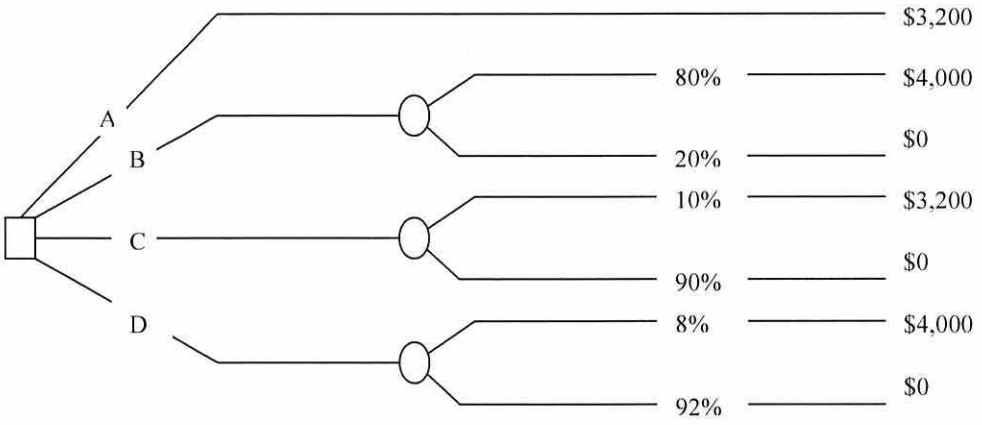
Davidson, D. and Suppes, P. (1956). A finitistic axiomatization of subjective probability and utility. *Economoetrica*, 24, 264-275.
Suppes, P. (1956). The role of subjective probability and utility in decision making. *Proceeds of the Third Berkley Symposium on Mathematical Statistics and Probability, 1954 – 1955*, 5, 61-73.
Debreu, G. (1959). Cardinal utility for even-chance mixtures of pairs of sure prospects. *Review of Economic Studies*, 26, 174-177.
Anscombe, F. J. and Aumann, R. J. (1963). A definition of subjective probability. *Annals of Mathematical Statistics*, 34, 199-205.
Pratt, J. W., Raiffa, H. and Schlaifer, R. (1964). The foundations of decision making under uncertainty: An elementary exposition. *Journal of the American Statistical Association*, 59, 353-375.
Bolker, E. D. (1967). A simultaneous axiomatization of utility and subjective probability. *Philosophy of Science*, 34, 333-340.
Fishburn, P. C. (1967). Preference-based definitions of subjective probability. *Annals of Mathematical Statistics*, 38, 1605-1617.
Pfanzagl, J. (1968). *Theory of measurement*. New York: Wiley.
Fishburn, P. C. (1969). A general theory of subjective probability and expected utilities. *Annals of Mathematical Statistics*, 40, 1419-1429.
Fishburn, P. C. (1970). *Utility theory of decision making*. New York: Wiley.
Luce, R. D. and Krantz, D. H. (1971). Conditional expected utility. *Economoetrica*, 39, 253-271.
Ferreira, P. E. (1972). On subjective probability and expected utilities. *Annals of Mathematical Statistics*, 43, 928-933.
Luce, R. D. (1972). Conditional expected, extensive utility. *Theory and Decision*, 3, 101-106.
Stigum, B. P. (1972). Finite state space and expected utility maximization. *Economoetrica*, 40, 253-259.
Fishburn, P. C. (1973). A mixture-set axiomatization of conbditional subjective expected utilities. *Economoetrica*, 41, 1-25.
Balch, M. (1974). On recent developments in subjective expected utility. In M. Balch, D. McFadden and S. Wu. (Eds.), *Essays on economic behaviour under uncertainty* (pp. 456-54). Amsterdam: North-Holland.
Balch, M. and Fishburn, P. C. (1974). Subjective expected utility for conditional primitives. In M. Balch, D. McFadden and S. Wu. (Eds.), *Essays on economic behaviour under uncertainty* (pp. 57-69). Amsterdam: North-Holland.
Roberts, F. S. (1974). Laws of exchange and their applications. *SIAM Journal of Applied Mathematics*, 26, 260-284.

Fishburn, P. C. (1975). A theory of subjective expected utility with vague preferences. *Theory and Decision*, 6, 287-310.
Richter, M. K. (1975). Rational choice and polynomial measurement models. *Journal of Mathematical Psychology*, 12, 99-113.
Narens, L. (1976). Utility uncertainty trade-off structures. *Journal of Mathematical Psychology*, 13, 296-322.
Domotor, Z. (1978). Axiomatization of Jeffrey utilities. *Syntheses*, 39, 165-210.
Jefferey, R. C. (1978). Axiomatization the logic of decision, In C. A. Hook, J. J. Leach and E. F. McClennen (eds.), *Foundations and Applications of Decision Theory, Vol. I: Theoretical Foundations* (pp. 227-231). Reidel, Dordrecht, Holland.
Myerson, R. B. (1979). An axiomatic derivation of subjective probability, utility, and evaluation functions. *Theory and Decision*, 11, 339-352.

(Source : Fishburn, 1981)

Appendix E. Violations of the Axioms and Properties of EU and SEU

Substitution



(Source: Keller, 1992)

Figure 1E. Substitution property decision tree.

The substitution, common-ratio or independence property of EU and SEU requires that whenever a choice or lottery is preferred or indifferent to another ($A \geq B$) the compound lottery $pA + (1\text{-}p)Z \geq pB + (1\text{-}p)Z$ must hold. According to the substitution property an individual preferring option or lottery A to option B in Figure 1E should also prefer option C to option D. In fact most individuals would opt for A over B and D over C (Keller ,1992). MacCrimmon and Larsen (1979), Kahneman and Tversky (1979) and Keller (1985a) have all found violations of this property.

Sure thing

The prime example of the violation of the sure thing or common consequences properties is Allais paradox (1952). Allais paradox shows that the majority of individuals order uncertain choices (prospects or lotteries) in a manner inconsistent with the axioms of substitution/independence and the independence of irrelevant alternatives. Violations of the sure thing property have been recorded by MacCrimmon and Larsen (1979), Kahneman and Tversky (1979) and Keller (1985a).

Linearity in probabilities

Indifference curves for subjective/expected utility theory should be linear and parallel. However, violation of the substitution and/or sure thing properties, violate linearity and commonly have indifference curves that fan-out or fan-in e.g. MacCrimmon and Larsen (1979), Kahneman and Tversky (1979) and Keller (1985a). Prospect theory can deal with fanning-out.

Betweenness

The betweenness property is a special case of the substitution property. The betweenness property states that if A is preferred to B (A > B) then the compound choice (or lottery) pA + (1-p)B is 'in between' the original choices in preference ordering. Coombs (1969, 1975) and Coombs and Huang (1970) found violations of this property and proposed portfolio theory as a generalised utility theory that could capture betweenness violations.

Ambiguity of indifference

EU/SEU theory requires ambiguity indifference. Ambiguity indifference means that an individual is indifferent between two identical choices where one has a non-vague probability p and the other the same (but ambiguous) subjective probability. Ellsberg's (1961) paradox suggests this is not the cased. Ellsberg found that individuals preferred choices where probabilities were known to choices with unknown or ambiguous probabilities.

Fixed reference levels

In EU/SEU the reference level (status quo) is assumed to remain constant for the period the theory is applied e.g. experiment length. However, people often react asymmetrically to incremental change in perceived gains and losses with respect to the current reference level (Keller, 1992)[2]. This has led to the development of generalised models such as prospect theory (Kahneman & Tversky, 1979) that treats gains and losses differently with a type of rank and sign utility function.

[2] A utility function with a fixed reference level can have different risk attitudes in the gain and loss domains (Keller, 1992) giving an S shaped function. The S shaped function was first proposed by Friedman and Savage (1948).

Risk attitude

EU/SEU does not directly address the issue of the choice of risk attitude. For example a student opting for option X over Y, where X is a 100% chance of a B- grade and Y is a 50% chance of an A grade and a 50% chance of a C grade, would be labelled risk averse based upon the USA standard grading scale. Using the US grading scale the Y option gives the greatest expected value of a B grade, 0.5(4.0) + 0.5(2.0)=3, 3=B. However the individual may not be forgoing a B in favour of a B – to avoid risk. If the individual values the increase from a C to a B- as much as an increase from a B- to an A, then they are risk neutral not risk averse relative to the preference for the outcome (example taken from Dyer and Sarin, 1982; Keller, 1985b). The only generalised utility theory that can currently deal with the strength of preference aspect is Prospect Theory.

Transitivity

Luce (1992) notes that the frequently conducted preference reversal experiment e.g. Lindman (1971), Reilly (1982) and Tversky, Slovic and Kahneman (1990), and the experiments comparing a chain of alternatives differing only slightly in probabilities e.g. Tversky (1969) and Raynard (1977), are frequently offered as evidence of intransitivity. However, Bostic et al. (1990) and Tversky, Slovic and Kahneman (1990) have questioned the equivalence of judged and choice indifference, which casts doubts upon the validity of preference reversal as evidence of intransitivity. Researchers differ in the weight they give to the chain experiments as intransitivity may be the result of inobservance of small differences in choices on the part of respondents (Luce, 1992)[3].

Regret theory questions the axiom of irrelevant alternatives (An irrelevant alternative should not affect the choice made). Bell and Raiffa (1982), Looms and Sugden (1982, 1987) and Sugden (1985) make the case that it is not irrelevant to take potential regret into account e.g. 'what if I had/do?' If regret is an attribute in the individual's decision (utility function) it may result in the violation of the axiom of transitivity (Schoemaker, 1992). However, LaValle (1992) argues that regret, elation, disappointment and similar emotions should not be included in decision models, even when used normatively or prescriptively due to the cognitive burden. Regret Theory (Loomes & Sugden, 1982)

[3] For a summary of transitivity violations under experimental conditions see Luce (1992).

and Nontransitive Measurable Utility (Fishburn, 1982) are both generalised utility theories that have been designed to allow for intransitivity.

Appendix F. Generalised Expected Utility Theories

Table F1

Representative examples of generalised expected utility theory

Theory	Authors
Prospect theory	Kahneman, D and Tversky, A. (1979). Prospect theory: an analysis of decision under risk. *Econometrica*, 47, 2, 263-291.
Weighted Utility	Chew, S. H. and MacCrimmon, K. R. (1979a). *Alpha-nu choice theory: a generalization of expected utility theory (Working paper #669)*. Faculty of Commerce and Business Administration, University of British Columbia, Vancouver, British Columbia. Chew, S. H. and MacCrimmon, K. R. (1979b). *Alpha utility theory, lottery composition and the Allais paradox (Working paper #686)*. Faculty of Commerce and Business Administration, University of British Columbia, Vancouver, British Columbia. Chew, S. H. (1983). *A generalisation of the quasilinear mean with applications*. Boston: Kluwer Academic Publishers.
Skew-symmetric bilinear utility	Fishburn, P. C. (1983). Transitive measurable utility. *Journal of Economic Theory*, 31, 293-317. Fishburn, P. C. (1984). SSB utility theory: an economic perspective. *Mathematical Social Science,* 8, 63-94.
Regret theory	Bell, D. (1982). Regret in decision making under uncertainty. *Operations Research*, 30, 961-981. Loomes, G. and Sugden R. (1982). Regret theory: an alternative theory of rational choice under uncertainty. *Economic Journal*, 92, 805-824.
Lottery dependent utility	Becker, J. L. (1986). *A new model of decision under risk using the concept of lottery dependent utility function*. Unpublished doctoral dissertation. Graduate School of Management, University of California at Los Angeles. Becker J. L. and Sarin, R. (1987). Decision analysis using lottery dependent utility. *Journal of risk and uncertainty*, 2, 105-117.
Approximate expected utility	Leland, J. (1988). *A theory of 'approximate' expected utility maximization (Working paper)*. Social and Decision Sciences, Carnegie-Mellon University, Pittsburgh, Philadelphia.
Expected utility with rank dependent probabilities (anticipated utility)	Quiggin, J. (1982). A theory of anticipated utility. *Journal of Economic Behaviour and Organization*, 3, 323-343.
Binary rank dependent (or	Yarri, M. E. (1987). The dual theory of choice under risk. *Econometrica*, 55,

dual bilinear) utility	95-115. Luce, R. D. and Narens, L. (1985). Classification of concatenation structures according to scale type. *Journal of Mathematical Psychology*, 29, 1-72.
General quadratic utility	Chew, S. H., Epstein, L. and Segal, U. (1988). Mixture symmetric utility theory (Working paper). University of Toronto. Machina, M. (1982). Expected utility analysis without the independence axiom. *Econometrica*, 50, 277-323.
Implicit expected utility	Chew, S. H. (1985). Implicit-weighted and semi-weighted utility theories, M-estimator, and nondemand revelation of second-price auction for an uncertain auction object *(Working paper #155)*. Department of Political Economy, The John Hopkins University, Baltimore, MD. Dekel, E. (1986). An axiomatic characterization of preference under uncertainty: weakening the independence axiom. *Journal of Economic Theory*, 40, 304-318.
Ordinal independence	Segal, U. (1984). Nonlinear decision weights with the independence axiom (working paper). Economics Department, University of California, Los Angeles. Green, J. and Jullien, B. (1988). Ordinal independence in nonlinear utility theory. *Journal of Risk and Uncertainty*, 1, 4, 355-387.

Appendix G. Experiment Questionnaire

Ysgol seicoleg **Prifysgol Cymru, Bangor** Adeilad Brigantia, Ffordd Penrallt Bangor, Gwynedd LL57 2AS Ffôn (01248) 382211 - Ffacs (02148) 382599 e-bost: hology@bangor.ac.uk www.psychology.bangor.ac.uk	• PRIFYSGOL CYMRU • UNIVERSITY OF WALES BANGOR 1884	**School of Psychology** **University of Wales, Bangor** Adeilad Brigantia, Penrallt Road Bangor, Gwynedd LL57 2AS Tel: (01248) 382211 – Fax: (02148) 382599 e-mail: psychology@bangor.ac.uk www.psychology.bangor.ac.uk

Informed Consent

Influences on Genetic Testing Decisions

I agree to participate as a volunteer in a scientific study as an authorised part of the research undertakings with the School of Psychology at the University of Wales, Bangor under the supervision of Dr. Val Morrison. The study and my part in it have been fully explained to me by either, Caroline White-Gwenin, Abbie Unwin, Nonn a'ch Dafydd or Gethin Griffith and I understand their explanation. The procedures of this study have been explained to me.

I understand that I am free not to answer specific items on the questionnaire. I understand that all data will remain confidential with regards to my identity. I am free to withdraw my consent at any time and terminate my participation at any time without penalty. I understand that I may request a summary of the results from this study.

In the case of any complaints concerning the conduct of research, these should be addressed to Professor C. F. Lowe, Head of School, School of Psychology, University of Wales, Bangor, Gwynedd LL57 2DG or Professor I. Russell, Director, The Institute of Medical and Social Care Research, Wheldon Building, University of Wales, Bangor, Gwynedd LL57 2UW.

Participant's signature ..... Date .....

I the undersigned have fully explained the study to the above individual

Experimenter's signature ..... Date .....

ID						

Influences on genetic testing decisions

(Preferences for Breast Cancer Genetics Services)

If you have any difficulties with any of these questions please ask the researcher that handed you the questionnaire for help.

What this questionnaire is about.

In this questionnaire we would like you to tell us about:

- Yourself and your family.
- Your personal experience (if any) of cancer, genetics and testing.
- Your perceptions of the advantages and disadvantages of genetic testing for breast cancer.
- Your interest in obtaining testing.
- How you feel about the testing process and its implications.
- What type of service/appointment you would prefer to attend.

Have you previously studied psychology on an Access, AS or A level course?

Yes ☐ No ☐

Have you previously studied economics on an Access, AS or A level course?

Yes ☐ No ☐

Please state your date of birth: Day ..... / Month ..... / Year .....

Please state your gender? (Please tick ✓one box only).

Male ☐ **Female** ☐

Do you have any children, if so, how many children do you have? ☐

Would you describe your ethnic origin as:
(Please tick ✓one box only)

Ashkenazi Jewish	☐	White	☐	Black African	☐
Black Caribbean	☐	Indian	☐	Pakistani	☐
Bangladeshi	☐	Chinese	☐	Japanese	☐

Other ☐ If Other, please describe .....

.....

All the questions on this page and the next relate to the main earner in your household. Please answer them to the best of your ability.

The household for the purposes of this questionnaire, is the one you reside in during non-term time such as the summer vacation e.g. with your parents/guardians, spouse or partner and any other individuals permanently residing in the home. Please do not include house or flatmates in this category.

Last week, was the main earner any of the following? (Please circle the number next to the answer you have selected)

Employee	1
Self-employed/freelance, or in your own/family business	2
On a government training scheme	3
Retired	4
In full-time education	5
Looking after home/family	6
Permanently sick/disabled	7
Other	8

If Other, please state .....
.....

	Yes	No
Has the main earner/head of household ever worked?	1	0

If No, please ignore the questions on page 4 and go on to page 5.

Please answer the remaining questions for the main job the 'main earner' was doing last week, or if not working last week, the last main job. The main job is the job in which most hours are usually worked.

Does (did) the main earner work as an:

Employee	1
Self-employed with employees	2
Self-employed/freelance without employees	3

How many people work(ed) for main earner/employer at the place of work?

None	**0**
24 people or less	**1**
25 people or more	**2**

What is (was) the full title of the main earner's 'main job'?
(For example, Primary School Teacher, State Registered Nurse, Car Mechanic, Television Service Engineer, Benefits Assistant. Civil Servant, Local Government Officer - Please give job title, not grade or pay band).

... ...

... ...

Please describe what the main earner does (did) in their main job?

... ...

... ...

	Yes	No
Does (did) the main earner supervise any other employees?	1	0

A supervisor or foreman is responsible for overseeing the work of others employees on a day-to-day basis.

What is (was) the business of the main earner's employer at the place where they work (worked) or if self-employed, what is (was) the nature of the business?
(For example, Making Shoes, Repairing Cars, Secondary Education, Food Wholesale, Clothing Retail, Hospital).

... ...

... ...

Please answer the following question to the nearest half hour. If hours of work are not fixed please give the average for the last four weeks.

How many hours a week does (did) the main earner usually work in their main job?

... ...hours per week.

Have you ever had cancer? Yes ☐ No ☐

If you answered **Yes**, where was the cancer?
(Please tick ✓all relevant boxes)

Breast ☐

Ovary ☐

Colorectal ☐

Lung ☐

Other ☐

If other, where was it? ... ...

Have any of your family ever had cancer? Yes ☐ No ☐ Don't know ☐

If you answered **Yes**, please tell us if any of the following family members have had cancer.

Relative	**Had cancer?** Yes	No	**Where was the cancer?** (e.g. breast)	**Age when cancer found?** (in years)
Your sister(s) & brother (s)	☐	☐	Person 1...................................... Person 2...................................... Person 3...................................... Person 4......................................	1................. 2................. 3................. 4.................
Your mother	☐	☐	....	....
Your mother's sister(s) & brother (s)	☐	☐	Person 1...................................... Person 2...................................... Person 3...................................... Person 4......................................	1................. 2................. 3................. 4.................
Your father	☐	☐	....	....
Your father's sister(s) & brother (s)	☐	☐	Person 1...................................... Person 2...................................... Person 3...................................... Person 4......................................	1................. 2................. 3................. 4.................
Your mother's mother	☐	☐	....	....
Your mother's father	☐	☐	....	....
Your father's mother	☐	☐	....	....
Your father's father	☐	☐	....	....

Do you have a genetic disorder? Yes ☐ No ☐

If you answered Yes, what is it? ... ...
... ...

Have you ever had genetic testing or counselling? Yes ❐ No ❐

If you answered **Yes**, please state the disorder you were tested or counselled for?
... ...
... ...

If you answered **No**, have you ever considered going for genetic testing? Yes ❐ No ❐

If you answered **Yes**, please state the disorder you considered obtaining testing for and the reasons that made you to consider obtaining the test.
... ...
... ...

Do you know any one who has a genetic disorder? Yes ❐ No ❐

Do you know any one who has had counselling or testing? Yes ❐ No ❐

Have you ever read genetic testing literature? Yes ❐ No ❐

Have you ever discussed genetic testing or counselling with anyone? Yes ❐ No ❐

How knowledgeable would you say you were about the following issues?
(Please circle one figure in each row)

Issues	Know nothing										Very knowledgeable
Cancer	0	10	20	30	40	50	60	70	80	90	100
Breast cancer	0	10	20	30	40	50	60	70	80	90	100
Non-cancer genetic testing	0	10	20	30	40	50	60	70	80	90	100
Genetic testing for cancer	0	10	20	30	40	50	60	70	80	90	100
Genetic testing for breast cancer	0	10	20	30	40	50	60	70	80	90	100

Attending cancer genetic testing for breast cancer has been described as each of the terms below, which one do you feel best describes it's purpose
(Please tick ✓one box only)

A preventative step ☐
Confirms that a person is ill ☐
Other ☐

or Don't Know ☐

Please state in the left hand column headed "**Benefits**", all the reasons you can think of **in favour** of genetic testing and counselling for breast cancer. Do not worry about the order in which you state the reasons.

Benefits	*Relevance* (0 = Not at all relevant to 100 = Extremely relevant).										
	0 Not at all relevant	10	20	30	40	50	60	70	80	90	100 Extremely relevant
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100

If genetic testing and counselling for breast cancer was available to you (free of charge) by booking an appointment, how relevant would the "**Benefits**" be to your decision to book or decline genetic testing and counselling? Please, rate the relevance of each of the "**Benefits**" to **you** by circling a number from 0 to 100 in the right hand column headed "**Relevance**".

Please state in the left hand column headed "**Disadvantages**", all the reasons you can think of **against** genetic testing and counselling for breast cancer. Do not worry about the order in which you state the reasons.

Disadvantages	*Relevance* (0 = Not at all relevant to 100 = Extremely relevant).										
	0 Not at all relevant	10	20	30	40	50	60	70	80	90	100 Extremely relevant
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100

If genetic testing and counselling for breast cancer was available to you (free of charge) by booking an appointment, how relevant would the "**Disadvantages**" be to your decision to book or decline genetic testing and counselling? Please, rate the relevance of each of the "**Disadvantages**" to **you** by circling a number from 0 to 100 in the right hand column headed "**Relevance**".

How much control do you believe you have over the following:

	Little /no control		Moderate control			Complete control	
Having a genetic test carried out	1	2	3	4	5	6	7
Getting information about cancer that runs in families	1	2	3	4	5	6	7

How much would your family like you to do the following as a way of becoming aware of cancer in its early stages?

	Not at all		No feelings either way			Very much	
Regular screening procedures (e.g. mammography)	1	2	3	4	5	6	7
Having a genetic test carried out	1	2	3	4	5	6	7
Getting information about cancer that runs in families	1	2	3	4	5	6	7

How much would your GP like you to do the following as a way of becoming aware of cancer in its early stages?

	Not at all		No feelings either way			Very much	
Regular screening procedures (e.g. mammography)	1	2	3	4	5	6	7
Having a genetic test carried out	1	2	3	4	5	6	7
Getting information about cancer that runs in families	1	2	3	4	5	6	7

How much do you want to do any of the following as a way of becoming aware of cancer in its early stages?

	Not at all		No feelings either way			Very much	
Regular screening procedures (e.g. mammography)	1	2	3	4	5	6	7
Having a genetic test carried out	1	2	3	4	5	6	7
Getting information about cancer that runs in families	1	2	3	4	5	6	7

Do you intend to do any of the following as a way of becoming aware of cancer in its early stages?

	Definitely no			Undecided			Definitely yes
Regular screening procedures (e.g. mammography)	1	2	3	4	5	6	7
Having a genetic test carried out	1	2	3	4	5	6	7
Getting information about cancer that runs in families	1	2	3	4	5	6	7

To what extent do you agree with the following statements?

	It will tell me nothing						I will better know my future
If I have genetic testing	1	2	3	4	5	6	7

	Harmful						Beneficial
Genetic testing would be	1	2	3	4	5	6	7

	Not valuable						Extremely valuable
How valuable would it be to know your genetic risk	1	2	3	4	5	6	7

How likely do you think you are of having a gene giving you an increased risk of getting breast cancer? *(Please circle)*

Not at all — *Almost certain or extremely*

1 2 3 4 5 6 7

How likely do you think it is that, at some point in your life, you will get breast cancer? *(Please circle)*

Not at all — *Almost certain or extremely*

1 2 3 4 5 6 7

How vulnerable do you think you are to getting breast cancer at some point in your life? *(Please circle)*

Not at all — *Almost certain or extremely*

1 2 3 4 5 6 7

Compared to others of my age and gender, my chances of developing breast cancer are- (Please circle)

0% 10% 20% 30% 40% 50% 60% 70% 80% 90% 100%

How serious an illness do you think breast cancer is? *(Please circle)*

Not at all serious — *Extremely serious*

0 10 20 30 40 50 60 70 80 90 100

How bad would it be to have breast cancer? *(Please circle)*

Not at all bad — *Extremely bad*

0 10 20 30 40 50 60 70 80 90 100

How bad would it be to find that you have an increased susceptibility of developing breast cancer? *(Please circle)*

Not at all bad — *Extremely bad*

0 10 20 30 40 50 60 70 80 90 100

To what extent are you worried about getting breast cancer? *(Please circle)*

Not at all — *Almost certain or extremely*

1 2 3 4 5 6 7

To what extent are you concerned about getting breast cancer? *(Please circle)*

Not at all — *Almost certain or extremely*

1 2 3 4 5 6 7

How anxious do you feel about your risk of developing cancer? *(Please circle)*

Not at all anxious — *Extremely anxious*

0 10 20 30 40 50 60 70 80 90 100

How anxious would you feel about having genetic testing? *(Please circle)*

Not at all anxious — *Extremely anxious*

0 10 20 30 40 50 60 70 80 90 100

This section looks at how you feel emotionally

Don't take too long over your replies: your immediate reaction to each item will probably be more accurate than a long thought-out response

Tick only one box in each section

I feel tense or wound up:
- Most of the time
- A lot of the time
- From time-to-time, Occasionally
- Not at all

I feel as if I am slowed down:
- Nearly all the time
- Very often
- Sometimes
- Not at all

I still enjoy the things I used to enjoy:
- Definitely as much
- Not quite as much
- Only a little
- Hardly at all

I get a sort of frightened feeling like 'butterflies' in the stomach:
- Not at all
- Occasionally
- Quite often
- Very often

I get a sort of frightened feeling as if something awful is about to happen:
- Very definitely and quite badly
- Yes, but not too badly
- A little, but it doesn't worry me
- Not at all

I have lost interest in my appearance:
- Definitely
- I don't take so much care as I should
- I may not take quite as much care
- I take just as much care as ever

I can laugh and see the funny side of things:
- As much as I always could
- Not quite as much now
- Definitely not so much now
- Not at all

I feel restless as if I have to be on the move:
- Very much indeed
- Quite a lot
- Not very much
- Not at all

Worrying thoughts go through my head:
- A great deal of the time
- A lot of the time
- From time-to-time, but not too often
- Only occasionally

I look forward with enjoyment to things:
- As much as I ever did
- Rather less than I used to
- Definitely less than I used to
- Hardly at all

I feel cheerful:
- Not at all
- Not often
- Sometimes
- Most Of the time

I get sudden feelings of panic:
- Very often
- Quite often
- Not very often
- Not at all

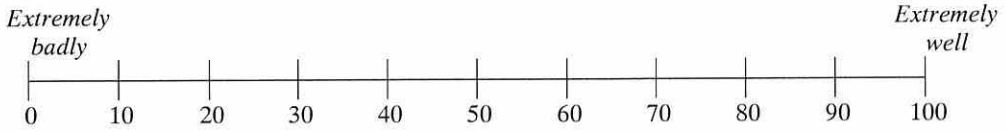
I can sit at ease and feel relaxed:
- Definitely
- Usually
- Not often
- Not at all

I can enjoy a good book or radio or TV programme
- Often
- Sometimes
- Not often
- Very seldom

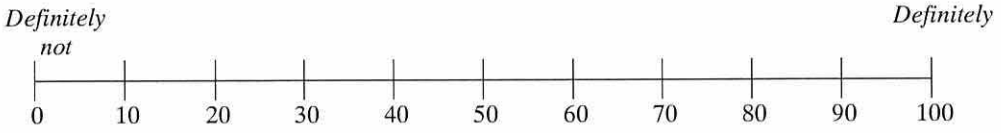
These questions are interested in your general outlook on life. Please be as honest and accurate as you can throughout. Try not to let your response to one statement influence your response to other statements. There are no 'correct' or 'Incorrect' answers. Answer according to your **own feelings,** rather than how you think 'most people' would answer. Please indicate the extent to which you agree or disagree with the questions by placing a tick in the appropriate box below each question.

I agree a lot	I agree a little	I neither agree or disagree	I disagree a little	I disagree a lot
In uncertain times, I usually expect the best.				
It's easy for me to relax.				
If something can go wrong for me, it will.				
I always look on the bright side.				
I'm always optimistic about my future.				
I enjoy my friends a lot.				
It's important for me to keep busy.				
I hardly ever expect things to go my way.				
Things never work out the way I want them to.				
I don't get upset easily.				
I'm a believer in the idea that 'every cloud has a silver lining'.				
I rarely count on good things happening to me.				

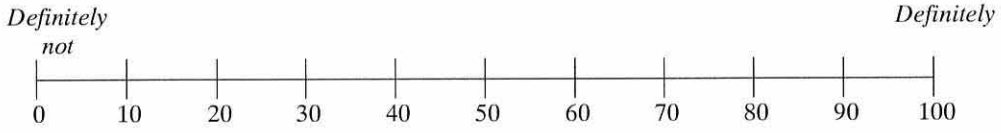
How well do you think you would cope if you had genetic testing and were told that you had an 80% chance of developing breast cancer by the age of 70?
(Please answer by drawing a line on the scale below)



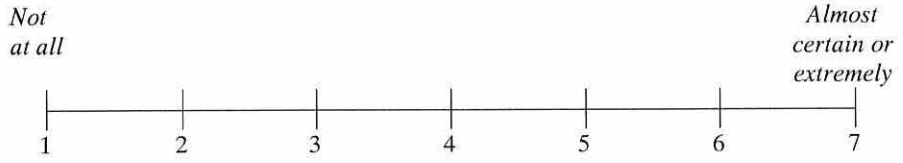
How interested are you in getting a genetic test for breast cancer susceptibility? *(Please circle)*



If genetic testing and counselling for breast cancer was available to you (*free of charge*), how likely would you be to book an appointment to have this service? *(Please circle)*



I plan to have genetic testing for breast cancer susceptibility when it is available? *(Please circle)*



There are many different ways in which parts of the cancer genetics service can be provided. The aim of the following questions is to find out which ways of providing the service you would prefer.

We would firstly like you to familiarise yourself with the different ways in which such a service could be provided.

1. Staff seen for counselling

Bearing in mind that a Consultant Geneticist will always be in overall charge of care; there are three different types of professionals that can provide genetics counselling. The options are:

- ***Specialist genetics nurse*** - a nurse who is trained in genetic counselling.
- ***Consultant geneticist*** - a hospital doctor who is trained in genetic counselling.
- ***Genetics associate*** - a scientist who is trained in genetic counselling.

2. Waiting time for letter

This refers to the time people have to wait between their doctor referring them to the genetics service and receiving a letter telling them their risk of developing familial cancer. The options are:

- **1 month**
- **2 months**
- **4 months**
- **6 months**

3. Distance to counselling

This refers to the distance people have to travel to their appointments with the genetics service. The options are:

- **20 miles**
- **40 miles**
- **60 miles**
- **80 miles**

4. Duration of counselling

This refers to the amount of time people spend with the genetics counsellor on a single appointment (this does not refer to the total time spent in the hospital, which may often be much longer). The options are:

- **30 minutes**
- **1 hour**
- **1 hour 30 minutes**
- **2 hours**

5. Availability of testing

This refers to the fact that currently genetic testing is only available to high risk clients and not to moderate or low risk clients. The options are:

- **High risk** - Testing only available to those at high risk.
- **All** - Testing available to all (whether high, moderate or low risk).

6. Cost of service

Please note that there is no possibility of people actually being asked to pay for the service they receive.

This item refers to the value you would place on cancer genetics services. To ascertain the value you place on cancer genetics services we would like you to consider how much you would be willing to pay a private provider or the NHS in the form of a single payment, one-off tax bill or national insurance contribution. Remember that any money you spend on genetics services would not be available to spend on other things. The options are:

- **£1,500**
- **£2,000**
- **£2,500**
- **£3,000**

Please tell us how important each of the aspects of the cancer genetics services are to you?
(For each aspect of care please circle the figure representing its importance).

	No importance		*Little importance*								*Very important*
Staff seen for counselling	0	1	2	3	4	5	6	7	8	9	10
Waiting time for a letter about risk of cancer	0	1	2	3	4	5	6	7	8	9	10
Distance to counselling	0	1	2	3	4	5	6	7	8	9	10
Duration of counselling	0	1	2	3	4	5	6	7	8	9	10
Availability of testing	0	1	2	3	4	5	6	7	8	9	10
Cost of service	0	1	2	3	4	5	6	7	8	9	10

Please rank these aspects of care in order of importance to you, using a scale of 1 – 6, where 1 = the most important and 6 = the least important, e.g. if staff seen for counselling is the most important place a 1 in the box opposite and if duration of counselling is the second most important place a 2 in the box opposite. Or if you have no preference then please tick ✓the box 'no preference'

	Rank	
• **Staff seen for counselling**	❒	
• **Waiting time for a letter about risk of cancer**	❒	
• **Distance to counselling**	❒	*or*
• **Duration of counselling**	❒	*no preference*
• **Availability of testing**	❒	❒
• **Cost of service**	❒	

Please now rank the following types of genetics counsellor, from 1 = most preferred to 3 = least preferred. Or if you have no preference, please tick ✓the box 'no preference'.

Genetics associate	❒	**Genetics nurse**	❒	*or no preference*
Consultant geneticist	❒			❒

Please now rank availability of genetics testing, from 1 = most preferred to 2 = least preferred. Or if you have no preference, please tick ✓the box 'no preference'.

High risk - Testing only available to those at high risk	❒	*or no preference*
All - Testing available to all (whether high, moderate or low risk)	❒	❒

On the next page you will be asked to choose between options A and B in each of the 25 choices.
For each choice, imagine that your doctor has referred you to a genetics service for the first time.

- You will be asked to look at each choice separately and tick ✓the one you would prefer.
- Everything else about the options, apart from the differences stated, are the same.
- Please answer every choice remembering that there are no right or wrong answers. It is your views that we are interested in.

Here are two examples of completed choice questions.

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics associate
Waiting time for letter	4 months	1 month
Distance to counselling	60 miles	20 miles
Duration of counselling	30 minutes	30 minutes
Availability of testing	All	High risk only
Cost of service	£3,000	£1,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	✓	☐

The person responding to the question in this example prefers option B rather than option A.

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics associate
Waiting time for letter	1 month	1 month
Distance to counselling	40 miles	20 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	High risk only
Cost of service	£3,000	£1,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	✓	☐	☐

The person responding to the question in this example prefers option A rather than option B.

Please turn the page and choose one option from each of the following 25 choices.

Choice 1

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Consultant geneticist
Waiting time for letter	4 months	4 months
Distance to counselling	20 miles	40 miles
Duration of counselling	1 hour	30 minutes
Availability of testing	High	High
Cost of service	£1,500	£2,000

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 2.

Choice 2

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	4 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	2 hours	1 hour
Availability of testing	All	All
Cost of service	£2,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 3.

Choice 3

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	4 months	2 months
Distance to counselling	60 miles	40 miles
Duration of counselling	30 minutes	1 hour
Availability of testing	All	All
Cost of service	£3,000	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 4.

Choice 4

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics associate
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	40 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	High
Cost of service	£2,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 5.

Choice 5

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Consultant geneticist
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	20 miles
Duration of counselling	1 hour	1 hour
Availability of testing	All	High
Cost of service	£2,500	£1,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 6.

Choice 6

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	80 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	High
Cost of service	£2,500	£2,000

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 7.

Choice 7

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics associate	Genetics nurse
Waiting time for letter	2 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	30 minutes	1 hour
Availability of testing	High	All
Cost of service	£1,500	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 8.

Choice 8

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Consultant geneticist
Waiting time for letter	2 months	2 months
Distance to counselling	40 miles	60 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	All
Cost of service	£2,500	£2,000

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 9.

Choice 9

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	2 months	2 months
Distance to counselling	20 miles	40 miles
Duration of counselling	2 hours	1 hour
Availability of testing	High	All
Cost of service	£3,000	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 10.

Choice 10

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	2 hours	1 hour
Availability of testing	High	All
Cost of service	£2,500	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 11.

Choice 11

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	1 month
Distance to counselling	40 miles	80 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	High
Cost of service	£2,500	£3,000

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 12.

Choice 12

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Consultant geneticist
Waiting time for letter	2 months	1 month
Distance to counselling	40 miles	60 miles
Duration of counselling	1 hour	1 hour
Availability of testing	All	High
Cost of service	£2,500	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 13.

Choice 13

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	1 month
Distance to counselling	40 miles	40 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	All
Cost of service	£2,500	£1,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 14.

Choice 14

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics associate	Genetics nurse
Waiting time for letter	1 month	2 months
Distance to counselling	20 miles	40 miles
Duration of counselling	2 hours	1 hour
Availability of testing	All	All
Cost of service	£2,000	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 15.

Choice 15

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	1 month	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	30 minutes	1 hour
Availability of testing	High	All
Cost of service	£2,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 16.

Choice 16

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	6 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	1 hour	1 hour
Availability of testing	High	All
Cost of service	£2,000	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 17.

Choice 17

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics associate
Waiting time for letter	2 months	6 months
Distance to counselling	40 miles	60 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	High
Cost of service	£2,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 18.

Choice 18

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Consultant geneticist
Waiting time for letter	2 months	6 months
Distance to counselling	40 miles	40 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	High
Cost of service	£2,500	£3,000

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 19.

Choice 19

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	6 months
Distance to counselling	40 miles	20 miles
Duration of counselling	1 hour	30 minutes
Availability of testing	All	All
Cost of service	£2,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 20.

Choice 20

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	6 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	1 hour 30 minutes	1 hour
Availability of testing	All	All
Cost of service	£1,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 21.

Choice 21

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	60 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	High
Cost of service	£2,500	£1,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 22.

Choice 22

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	4 months	2 months
Distance to counselling	40 miles	40 miles
Duration of counselling	30 minutes	1 hour
Availability of testing	High	All
Cost of service	£2,000	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 23.

Choice 23

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	20 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	High
Cost of service	£2,500	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 24.

Choice 24

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics associate	Genetics nurse
Waiting time for letter	4 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	1 hour	1 hour
Availability of testing	All	All
Cost of service	£3,000	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 25.

Choice 25

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	4 months	1 month
Distance to counselling	60 miles	40 miles
Duration of counselling	30 minutes	2 hours
Availability of testing	All	All
Cost of service	£3,000	£1,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

How difficult/easy did you find the last 25 questions on choice of options? *(please circle)*

Very Difficult				*Moderate*				*Very easy*	
1	*2*	*3*	*4*	*5*	*6*	*7*	*8*	*9*	*10*

Please Stop Here!

You will be given some information shortly.

Once you have read the information the researcher coordinating your group will ask you to turn to the next page and answer the remaining questions.

Thank you.

How knowledgeable would you say you were about the following issues?
(Please circle one figure in each row)

Issues	Know nothing										Very knowledgeable
Cancer	0	10	20	30	40	50	60	70	80	90	100
Breast cancer	0	10	20	30	40	50	60	70	80	90	100
Non-cancer genetic testing	0	10	20	30	40	50	60	70	80	90	100
Genetic testing for cancer	0	10	20	30	40	50	60	70	80	90	100
Genetic testing for breast cancer	0	10	20	30	40	50	60	70	80	90	100

Attending cancer genetic testing for breast cancer has been described as each of the terms below, which one do you feel best describes it's purpose
(Please tick ✓one box only)

A preventative step ☐
Confirms that a person is ill ☐
Other ☐

or Don't Know ☐

Please state in the left hand column headed "**Benefits**", all the reasons you can think of **in favour** of genetic testing and counselling for breast cancer. Do not worry about the order in which you state the reasons.

Benefits	*Relevance* (0 = Not at all relevant to 100 = Extremely relevant).										
	Not at all relevant 0	10	20	30	40	50	60	70	80	90	Extremely relevant 100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100

If genetic testing and counselling for breast cancer was available to you (free of charge) by booking an appointment, how relevant would the "**Benefits**" be to your decision to book or decline genetic testing and counselling? Please, rate the relevance of each of the "**Benefits**" to **you** by circling a number from 0 to 100 in the right hand column headed "**Relevance**".

Please state in the left hand column headed "**Disadvantages**", all the reasons you can think of **against** genetic testing and counselling for breast cancer. Do not worry about the order in which you state the reasons.

Disadvantages	*Relevance* (0 = Not at all relevant to 100 = Extremely relevant).										
	Not at all relevant										Extremely relevant
	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100
----------	0	10	20	30	40	50	60	70	80	90	100

If genetic testing and counselling for breast cancer was available to you (free of charge) by booking an appointment, how relevant would the "**Disadvantages**" be to your decision to book or decline genetic testing and counselling? Please, rate the relevance of each of the "**Disadvantages**" to **you** by circling a number from 0 to 100 in the right hand column headed "**Relevance**".

How much control do you believe you have over the following:

	Little /no control		Moderate control			Complete control	
Having a genetic test carried out	1	2	3	4	5	6	7
Getting information about cancer that runs in families	1	2	3	4	5	6	7

How much would your family like you to do the following as a way of becoming aware of cancer in its early stages?

	Not at all		No feelings either way			Very much	
Regular screening procedures (e.g. mammography)	1	2	3	4	5	6	7
Having a genetic test carried out	1	2	3	4	5	6	7
Getting information about cancer that runs in families	1	2	3	4	5	6	7

How much would your GP like you to do the following as a way of becoming aware of cancer in its early stages?

	Not at all		No feelings either way			Very much	
Regular screening procedures (e.g. mammography)	1	2	3	4	5	6	7
Having a genetic test carried out	1	2	3	4	5	6	7
Getting information about cancer that runs in families	1	2	3	4	5	6	7

How much do you want to do any of the following as a way of becoming aware of cancer in its early stages?

	Not at all		No feelings either way			Very much	
Regular screening procedures (e.g. mammography)	1	2	3	4	5	6	7
Having a genetic test carried out	1	2	3	4	5	6	7
Getting information about cancer that runs in families	1	2	3	4	5	6	7

Do you intend to do any of the following as a way of becoming aware of cancer in its early stages?

	Definitely no			Undecided			Definitely yes
Regular screening procedures (e.g. mammography)	1	2	3	4	5	6	7
Having a genetic test carried out	1	2	3	4	5	6	7
Getting information about cancer that runs in families	1	2	3	4	5	6	7

To what extent do you agree with the following statements?

	It will tell me nothing						I will better know my future
If I have genetic testing	1	2	3	4	5	6	7

	Harmful						Beneficial
Genetic testing would be	1	2	3	4	5	6	7

	Not valuable						Extremely valuable
How valuable would it be to know your genetic risk	1	2	3	4	5	6	7

How likely do you think you are of having a gene giving you an increased risk of getting breast cancer? *(Please circle)*

Not at all — *Almost certain or extremely*

1 2 3 4 5 6 7

How likely do you think it is that, at some point in your life, you will get breast cancer? *(Please circle)*

Not at all — *Almost certain or extremely*

1 2 3 4 5 6 7

How vulnerable do you think you are to getting breast cancer at some point in your life? *(Please circle)*

Not at all — *Almost certain or extremely*

1 2 3 4 5 6 7

Compared to others of my age and gender, my chances of developing breast cancer are- (Please circle)

0% 10% 20% 30% 40% 50% 60% 70% 80% 90% 100%

How serious an illness do you think breast cancer is? *(Please circle)*

Not at all serious — *Extremely serious*

0 10 20 30 40 50 60 70 80 90 100

How bad would it be to have breast cancer? *(Please circle)*

Not at all bad — *Extremely bad*

0 10 20 30 40 50 60 70 80 90 100

How bad would it be to find that you have an increased susceptibility of developing breast cancer? *(Please circle)*

Not at all bad — *Extremely bad*

0 10 20 30 40 50 60 70 80 90 100

To what extent are you worried about getting breast cancer? *(Please circle)*

Not at all — *Almost certain or extremely*

1 2 3 4 5 6 7

To what extent are you concerned about getting breast cancer? *(Please circle)*

Not at all — *Almost certain or extremely*

1 2 3 4 5 6 7

How anxious do you feel about your risk of developing cancer?*(Please circle)*

Not at all anxious — *Extremely anxious*

0 10 20 30 40 50 60 70 80 90 100

How anxious would you feel about having genetic testing? *(Please circle)*

Not at all anxious — *Extremely anxious*

0 10 20 30 40 50 60 70 80 90 100

How well do you think you would cope if you had genetic testing and were told that you had an 80% chance of developing breast cancer by the age of 70?
(Please answer by drawing a line on the scale below)

Extremely badly — *Extremely well*

0 10 20 30 40 50 60 70 80 90 100

How interested are you in getting a genetic test for breast cancer susceptibility?
(Please circle)

Definitely not — *Definitely*

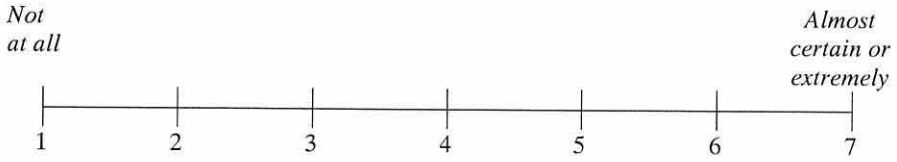
0 10 20 30 40 50 60 70 80 90 100

If genetic testing and counselling for breast cancer was available to you (*free of charge*), how likely would you be to book an appointment to have this service? *(Please circle)*

Definitely not — *Definitely*

0 10 20 30 40 50 60 70 80 90 100

I plan to have genetic testing for breast cancer susceptibility when it is available?
(Please circle)



The aim of the following questions is to find out which ways of providing the service you would prefer.

We would firstly like you to re-familiarise yourself with the different ways in which such a service could be provided.

1. Staff seen for counselling

Bearing in mind that a Consultant Geneticist will always be in overall charge of care; there are three different types of professionals that can provide genetics counselling. The options are:

- ***Specialist genetics nurse*** - a nurse who is trained in genetic counselling.
- ***Consultant geneticist*** - a hospital doctor who is trained in genetic counselling.
- ***Genetics associate*** - a scientist who is trained in genetic counselling.

2. Waiting time for letter

This refers to the time people have to wait between their doctor referring them to the genetics service and receiving a letter telling them their risk of developing familial cancer. The options are:

- **1 month**
- **2 months**
- **4 months**
- **6 months**

3. Distance to counselling

This refers to the distance people have to travel to their appointments with the genetics service. The options are:

- **20 miles**
- **40 miles**
- **60 miles**
- **80 miles**

4. Duration of counselling

This refers to the amount of time people spend with the genetics counsellor on a single appointment (this does not refer to the total time spent in the hospital, which may often be much longer). The options are:

- **30 minutes**
- **1 hour**
- **1 hour 30 minutes**
- **2 hours**

5. Availability of testing

This refers to the fact that currently genetic testing is only available to high risk clients and not to moderate or low risk clients. The options are:

- **High risk** - Testing only available to those at high risk.
- **All** - Testing available to all (whether high, moderate or low risk).

6. Cost of service

Please note that there is no possibility of people actually being asked to pay for the service they receive.

This item refers to the value you would place on cancer genetics services. To ascertain the value you place on cancer genetics services we would like you to consider how much you would be willing to pay a private provider or the NHS in the form of a single payment, one-off tax bill or national insurance contribution. Remember that any money you spend on genetics services would not be available to spend on other things. The options are:

- **£1,500**
- **£2,000**
- **£2,500**
- **£3,000**

Please tell us how important each of the aspects of the cancer genetics services are to you?
(For each aspect of care please circle the figure representing its importance).

	No importance	*Little importance*									*Very important*
Staff seen for counselling	0	1	2	3	4	5	6	7	8	9	10
Waiting time for a letter about risk of cancer	0	1	2	3	4	5	6	7	8	9	10
Distance to counselling	0	1	2	3	4	5	6	7	8	9	10
Duration of counselling	0	1	2	3	4	5	6	7	8	9	10
Availability of testing	0	1	2	3	4	5	6	7	8	9	10
Cost of service	0	1	2	3	4	5	6	7	8	9	10

Please rank these aspects of care in order of importance to you, using a scale of 1 – 6, where 1 = the most important and 6 = the least important, e.g. if staff seen for counselling is the most important place a 1 in the box opposite and if duration of counselling is the second most important place a 2 in the box opposite. Or if you have no preference then please tick ✓the box 'no preference'

	Rank	
• **Staff seen for counselling**	❒	
• **Waiting time for a letter about risk of cancer**	❒	
• **Distance to counselling**	❒	*or*
• **Duration of counselling**	❒	*no preference*
• **Availability of testing**	❒	❒
• **Cost of service**	❒	

Please now rank the following types of genetics counsellor, from 1 = most preferred to 3 = least preferred. Or if you have no preference, please tick ✓the box 'no preference'.

Genetics associate	❒	**Genetics nurse**	❒	*or no preference*
Consultant geneticist	❒			❒

Please now rank availability of genetics testing, from 1 = most preferred to 2 = least preferred. Or if you have no preference, please tick ✓the box 'no preference'.

High risk - Testing only available to those at high risk	❒	*or no preference*
All - Testing available to all (whether high, moderate or low risk)	❒	❒

On the next page you will be asked to choose between options A and B in each of the 25 choices.
For each choice, imagine that your doctor has referred you to a genetics service for the first time.

- You will be asked to look at each choice separately and tick ✓the one you would prefer.
- Everything else about the options, apart from the differences stated, are the same.
- Please answer every choice remembering that there are no right or wrong answers. It is your views that we are interested in.

Here are two examples of completed choice questions.

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics associate
Waiting time for letter	4 months	1 month
Distance to counselling	60 miles	20 miles
Duration of counselling	30 minutes	30 minutes
Availability of testing	All	High risk only
Cost of service	£3,000	£1,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	✓	☐

The person responding to the question in this example prefers option B rather than option A.

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics associate
Waiting time for letter	1 month	1 month
Distance to counselling	40 miles	20 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	High risk only
Cost of service	£3,000	£1,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	✓	☐	☐

The person responding to the question in this example prefers option A rather than option B.

Please turn the page and choose one option from each of the following 25 choices.

Choice 1

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Consultant geneticist
Waiting time for letter	4 months	4 months
Distance to counselling	20 miles	40 miles
Duration of counselling	1 hour	30 minutes
Availability of testing	High	High
Cost of service	£1,500	£2,000

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 2.

Choice 2

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	4 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	2 hours	1 hour
Availability of testing	All	All
Cost of service	£2,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 3.

Choice 3

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	4 months	2 months
Distance to counselling	60 miles	40 miles
Duration of counselling	30 minutes	1 hour
Availability of testing	All	All
Cost of service	£3,000	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 4.

Choice 4

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics associate
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	40 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	High
Cost of service	£2,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 5.

Choice 5

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Consultant geneticist
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	20 miles
Duration of counselling	1 hour	1 hour
Availability of testing	All	High
Cost of service	£2,500	£1,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 6.

Choice 6

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	80 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	High
Cost of service	£2,500	£2,000

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 7.

Choice 7

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics associate	Genetics nurse
Waiting time for letter	2 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	30 minutes	1 hour
Availability of testing	High	All
Cost of service	£1,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 8.

Choice 8

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Consultant geneticist
Waiting time for letter	2 months	2 months
Distance to counselling	40 miles	60 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	All
Cost of service	£2,500	£2,000

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 9.

Choice 9

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	2 months	2 months
Distance to counselling	20 miles	40 miles
Duration of counselling	2 hours	1 hour
Availability of testing	High	All
Cost of service	£3,000	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 10.

Choice 10

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	2 hours	1 hour
Availability of testing	High	All
Cost of service	£2,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 11.

Choice 11

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	1 month
Distance to counselling	40 miles	80 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	High
Cost of service	£2,500	£3,000

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 12.

Choice 12

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Consultant geneticist
Waiting time for letter	2 months	1 month
Distance to counselling	40 miles	60 miles
Duration of counselling	1 hour	1 hour
Availability of testing	All	High
Cost of service	£2,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 13.

Choice 13

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	1 month
Distance to counselling	40 miles	40 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	All
Cost of service	£2,500	£1,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 14.

Choice 14

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics associate	Genetics nurse
Waiting time for letter	1 month	2 months
Distance to counselling	20 miles	40 miles
Duration of counselling	2 hours	1 hour
Availability of testing	All	All
Cost of service	£2,000	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 15.

Choice 15

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	1 month	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	30 minutes	1 hour
Availability of testing	High	All
Cost of service	£2,500	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 16.

Choice 16

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	6 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	1 hour	1 hour
Availability of testing	High	All
Cost of service	£2,000	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 17.

Choice 17

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics associate
Waiting time for letter	2 months	6 months
Distance to counselling	40 miles	60 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	High
Cost of service	£2,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 18.

Choice 18

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Consultant geneticist
Waiting time for letter	2 months	6 months
Distance to counselling	40 miles	40 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	High
Cost of service	£2,500	£3,000

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 19.

Choice 19

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	6 months
Distance to counselling	40 miles	20 miles
Duration of counselling	1 hour	30 minutes
Availability of testing	All	All
Cost of service	£2,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 20.

Choice 20

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	6 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	1 hour 30 minutes	1 hour
Availability of testing	All	All
Cost of service	£1,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 21.

Choice 21

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	60 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	High
Cost of service	£2,500	£1,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 22.

Choice 22

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	4 months	2 months
Distance to counselling	40 miles	40 miles
Duration of counselling	30 minutes	1 hour
Availability of testing	High	All
Cost of service	£2,000	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 23.

Choice 23

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	20 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	High
Cost of service	£2,500	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 24.

Choice 24

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics associate	Genetics nurse
Waiting time for letter	4 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	1 hour	1 hour
Availability of testing	All	All
Cost of service	£3,000	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 25.

Choice 25

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	4 months	1 month
Distance to counselling	60 miles	40 miles
Duration of counselling	30 minutes	2 hours
Availability of testing	All	All
Cost of service	£3,000	£1,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

How difficult/easy did you find the last 25 questions on choice of options? *(please circle)*

Very Difficult				*Moderate*					*Very easy*
1	*2*	*3*	*4*	*5*	*6*	*7*	*8*	*9*	*10*

Thank you for completing the questionnaire.

Please wait for the researcher coordinating your group to ask you to return the completed questionnaire and for them to ensure you receive your credits.

Appendix H. Script of the Oral Invitation to Participate in the 'Influences on Genetic Testing Decisions' Study

1. We would like to invite both male and female students to participate in a study relating to genetic testing for breast cancer susceptibility (Men also get breast cancer).

2. The study is called 'Influences on Genetic Testing Decisions'.

3. The study forms part of a Ph.D. thesis and three undergraduate projects being undertaken with the school of psychology.

4. You will be required to complete a questionnaire relating to your knowledge, beliefs and attitudes towards cancer and genetic testing.

5. In addition, you will hear, and be provided with, some information relating to health issues.

6. Your participation time should be around one and a half hours.

7. Students of the school of psychology will receive 2 course credits and 70 print credits per hour

8. If you would like to participate, please sign up on the sign up sheet which will be outside the Mac Lab of the Wheldon Building and attend Lecture Theatre 1, the Wheldon Building (Science Site) at 11:00 to 11:10 a.m. on Monday the 10th of October and complete the consent form which will be provided at the beginning of the study.

9. Hand out the information sheets.

Appendix I. Information about the Experiment

Information

Influences on Genetic Testing Decisions

We would like to invite both male and female students to participate in a study relating to genetic testing for breast cancer susceptibility (Men also get breast cancer). You will be required to complete a questionnaire relating to your knowledge, beliefs and attitudes towards cancer and genetic testing. In addition, you will hear, and be provided with, some information relating to health issues.

Your participation time should be around one and a half hours. Students of the school of psychology will receive 2 course credits and 70 print credits per hour and non-Psychology students will receive 70 print credits per hour.

If you have had previous experience of cancer and/or genetic testing you may feel unwilling to participate, unable to participate, or unable to answer some of the questions. Please feel no obligation to answer anything you find distressing. You are, of course, able to withdraw from the study at any time. If you would like to participate, please register by entering your name on the 'sign-up sheet' located outside the Mac Lab of the Wheldon Building by 5:00 pm on Wednesday the 9th of October. Then attend Lecture Theatre 1, the Wheldon Building (Science Site) at 11:00 am on Thursday the 10th of October and sign the consent form which will be provided at the beginning of the study.

If you have any questions to ask about the study before you decide whether or not to take part, please phone Gethin Griffith on 07887500272.

If you have any complaints about the way in which the research has been conducted please contact: Professor C. F. Lowe, Head of School, School of Psychology, University of Wales, Bangor, Gwynedd LL57 2DG or Professor I. Russell, Director, The Institute of Medical and Social Care Research, Wheldon Building, University of Wales, Bangor, Gwynedd LL57 2UW.

Appendix J. Patient Questionnaires

Baseline Questionnaire Issued by the GenQuest Team

Prifysgol Cymru · University of Wales
BANGOR

UWCM

GenQuest

A study of the All-Wales Cancer Genetics Service

Dear service user,

Following your referral to the All-Wales Cancer Genetics Service you were asked if you would consent to being approached by the University of Wales college of Medicine to be part of an evaluation study. We would therefore like to ask you if you would like to take part in this study, which will look at the value of the Cancer Genetics Service to those who use it. Your participation will be of great value, but you do not have to take part.

If you decide to take part, but change your mind latter, you can withdraw from the study at any time. If you decide to withdraw, it will not affect the future treatment you receive from either the Cancer Genetics Service or your own doctor. If you decide not to take part in the study, but will allow us to keep in touch with you so that we can make comparisons with those who do take part, this will still be a great help.

More details about the study are enclosed with this letter, along with a number of possible questions and answers designed to help you decide whether you will take part. If you have any other questions please get in touch with us at the address below.

Yours faithfully,

Jim Turner

Jim Turner, University of Wales College of Medicine, Gwenfro Building, Wrexham Technology Park,
Wrexham, LL13 7YP.
Tel. 01978 316241. Fax. 01978 311419

A study of the All-Wales Cancer Genetics Service

PARTICIPANT CONSENT FORM

For those who voluntarily consent to be part of this evaluation, some personal information along with information provided on questionnaires will be required. In addition, it will be necessary for NHS staff to check my medical records as part of the evaluation.

PLEASE DO NOT SIGN THIS CONSENT FORM BEFORE READING THE ENCLODSED INFORMATION SHEET

- I have read and understood the information sheet explaining the study.
- I have had the opportunity to ask questions and discuss the study in detail.
- I have received satisfactory answers to all my questions about the study and I am aware I can ask further questions at any time as these may come to mind.
- I have received enough information about the study to allow me to make an informed decision.
- I understand I do not have to take part in t he study if I do not wish to do so.
- I understand that I can withdraw from the study at any time and that doing so will not affect the future treatment I receive from either the Cancer Genetics Service or my own doctor.
- I understand NHS staff will examine my medical records if I take part in this study.
- I understand that any information I give, or is obtained in the course of the study will be kept completely safe and confidential.

Name in BLOCK LETTERS ... ...

Signed ... ...

Information for participants

The Service
The All-Wales Cancer Genetics Service is the first service of this type in the United Kingdom. This service will make use of what we know about a link between certain types of cancer and genetic factors that are known to increase personal risk. This service is therefore very important.

The Purpose of the Study
There is an increasing need to get the best value from the money used to fund the NHS. The purpose of the study is to find out the value of the Cancer Genetics Service by looking at different aspects such as the service's public acceptability, how effective it is and what its likely cost will be to the NHS and to society. To find the answer we need to speak to the people who use the service and also to those who choose not to use it.

Ethical Approval
Ethical approval has been given both nationally and locally: see below for Local Research Ethics Committee contact details.

Source of Funding for the Study
The National Assembly of Wales and Macmillan Cancer Relief.

The Size and Time Period of the Study
We hope to recruit approximately 200 people from three genetic centres located across Wales over a period of 18 months.

Selecting People to Take Part in the Study
There will be two main groups: people who use the Cancer Genetics Service when offered and those who, for whatever reason, do not. Other groups will be identified from the two main groups by, for example, type of cancer.

Taking Part in the Study
Taking part in the study will mean completing general health, psychological and cancer-specific questionnaires at three separate stages of the study. There will also be questionnaires asking about how much you use the cancer and other health services during the same period: the approximate length of time it will take to fill in the questionnaires is 45 minutes. At the end of the study period a small number of participants will be asked to take part in interviews that will allow people to elaborate their views of the service in more detail. These interviews will also be voluntary and held in a place that is easiest for you (e.g. in your own home).

A Study of the All-Wales Cancer Genetics Service

Some questions you may have about taking part in this study.

Can I take part in this study even if I do not to use the cancer genetics service?

Yes, because this study is completely separate from using the service. It is very important that all those who are referred give their views even if they decide not to use the service.

How many times would I be asked to fill in questionnaires?

You would be asked to fill in questionnaires three times over a period of six months (a fourth time for questions about your use of cancer and other health services).

How long will the study last?

The study will last for two years.

Will medical records be used?

Medical records will be checked, only with your expressed permission, to find out the number of visits you made to your doctor during the period of the study.

How often will medical records be used?

Your records will be checked twice in the study.

Who will see my medical records?

Only NHS staff will see your records.

Will any other records be created?

Yes, the questionnaires you complete will be a paper record and these will also be entered on a computer database.

How safe is the information held in these records?

The information you provide on questionnaires will be combined with similar information from other people in the study and stored on computer disks. This procedure, along with keeping your name and address separate from the information you provide on questionnaires, makes all

the information anonymous. At end of the study period, all the questionnaires will be stored in locked filing cabinets for seven years for the purpose of secondary analysis then

destroyed. Following primary analysis, computer disks will be erased and for those participants who agreed to be interviewed, audiotapes will be destroyed.

Will any of my information be published?

The results of the study will be made public, but your personal details will be kept separate from your answers to questions. Your answers will be added to those of other people and this will make them anonymous.

Will participating cost me anything?

Participation in the study will only cost the time taken to complete the questionnaires. If you have any travel expenses as a result of taking part in the study, then you will be able to claim these back.

What is the first step?

If after reading this letter you decide you want to take part in the study, then sign the consent form. After signing the consent form please complete the first set of questionnaires enclosed with this letter and mail them back in the pre-paid envelope, along with the consent form. Further questionnaires will be sent at approximately one-month, three-months and six-months. If you do not want to take part in the study, please return the first set of questionnaires in the pre-paid envelope.

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GenQuest

Your Views And Feelings About Our Service

Thank you for your help with this important study. The questionnaires should take about 45 minutes to fill in, but please do not feel compelled to complete them all in one session. Please read carefully the instructions for each question.

You will find that some of the questions are repetitive, but we would be grateful if you could complete all of them anyway, as this information is important in helping us to develop our genetics service. The information you give here is confidential and there is no need for you to write your name on the questionnaires.

When you have completed the questionnaires, please check to make sure you have not missed anything out, then return them to us in the stamped addressed envelope provided. We look forward to receiving your completed questionnaires soon.

MANY THANKS FOR AGREEING TO TAKE PART IN THIS EVALUATION

PLEASE NOTE
Some of these questions or issues may not relate to you.

Funded by:

The National Assembly of Wales
and
MacMillan Cancer Relief

Office Use Only: Personal Research No. ……...Date of Issue. ……Qr.1

Demographic Information

1. Please tick highest qualification gained. **(✓)**

Qualification	
None	
GCSE (Grades D to G), CSE (Grades 2 to 5) or equivalent	
O level passes, GCSE (Grades A to C), CSE (Grade 1) or equivalent	
A level, AS level, Advanced Senior Certificate or equivalent	
NVQ/SVQ Level 1, GNVQ Foundation or equivalent	
NVQ/SVQ Level 2, GNVQ Intermediate, City & Guilds Craft, BTEC First Diploma or equivalent	
NVQ/SVQ Level 3, GNVQ Advanced, City & Guilds Advanced Craft, RSA Advanced Diploma or equivalent	
NVQ Level 4, HND, HNC, RSA Higher Diploma or equivalent	
NVQ Level 5	
Degree	
Qualified Teacher Status (for school)	
Qualified Nurse, Midwife or Health visitor	
Qualified Medical Doctor or Dentist	
Postgraduate qualification (MSc etc.)	
Other	

If other, please state qualification:

.....

.....

Please circle the number next to the answer you have selected.

12.

	Yes	No
Are you the main wage earner in your household ?	1	0

Please provide answers for the main wage earner in the household for questions 3 to 4.

3.

	Yes	No
Last week was the main earner doing any work: • as an employee, • self-employed/freelance, or in your own/family business, • on a government sponsored training scheme?	1	0

If you answered yes, please ignore questions 14 to 17.

4.

	Yes	No
Was the main earner actually looking for any kind of paid work during the last 4 weeks?	1	0
If a job had been available last week, could the main earner have started it within 2 weeks?	1	0
Last week was the main earner waiting to start a job already obtained?	1	0

5. Last week, was the main earner any of the following?

Retired	1
In full-time education	2
Looking after home/family	3
Permanently sick/disabled	4
None of the above	5

6.

	Yes	No
Has the main earner ever worked ?	1	0

If No, please go to question 15.

7. Please state the year the main earner last worked: ... ...

Please answer the remaining questions for the main job the 'main earner' was doing last week, or if not working last week, the last main job. Your main job is the job in which you usually work the most hours.

8. Does (did) the main earner work as an:

Employee	1
Self-employed with employees	2
Self-employed/freelance without employees	3

9. How many people work(ed) for main earner/employer at the place of work?

None	0
24 people or less	1
25 people or more	2

10. What is (was) the full title of the main earner's 'main job'?

For example, Primary School Teacher, State Registered Nurse, Car Mechanic, Television Service Engineer, Benefits Assistant.

Civil Servant, Local Government Officer - Please give job title, not grade or pay band.

... ...

... ...

1. Please describe what the main earner does (did) in their main job?

.. ..

.. ..

2.

	Yes	No
)oes (did) the main earner supervise any other employees?	1	0

\ supervisor or foreman is responsible for overseeing the work of others employees on a day-to-day basis.

3. What is (was) the business of the main earner's employer at the place where they work worked) or if self-employed, what is (was) the nature of the business?

For example, Making Shoes, Repairing Cars, Secondary Education, Food Wholesale, Clothing Retail, Hospital.

Civil Servant, Local Government Officer - Please give job title not grade or pay band.

... ...

... ...

Please answer question 24 to the nearest half hour. If your hours of work are not fixed please give the average for the last four weeks.

14. How many hours a week does (did) the main earner usually work in their main job?

... ...hours per week.

Other studies have found that personal and household income have an influence on people's perceptions and reactions to illness. We would like to see if this is the case with inherited cancer and so we would like you to indicate what your approximate total household income (that of you and the remainder of the adults in your household) is. This information is completely confidential, no person's individual responses to any question will be disclosed.

15. What is your total current gross household income from all sources?
Do not deduct tax, national insurance, superannuation or health insurance payments.
Count all income including: earnings, pensions, benefits, interest from savings or investments, rent from property, other (e.g. maintenance payments and grants).

Income			
Nil	**0**	£25,000 to £34,999 per year or £481 to £673 per week	**6**
Less than £4,999 per year or Less than £96 per week	**1**	£35,000 to £44,999 per year or £674 to £865 per week	**7**
£5,000 to £9,999 per year or £97 to £192 per week	**2**	£45,000 to £49,999 per year or £866 to £961 per week	**8**
£10,000 to £14,999 per year or £193 to £288 per week	**3**	£50,000 to £54,999 per year or £962 to £1057 per week	**9**
£15, 000 to £19,999 per year or £299 to £384 per week	**4**	£55,000 or more per year or £1058 or more per week	**10**
£20,000 to £24,999 per year or £385 to £480 per week	**5**		

16. How many adults are there in your household? .....

If you have any comments regarding this questionnaire or matters arising from it please include them here.

.....

.....

.....

The GenQuest team would like to say a really big

THANKYOU

for all the time and effort you have given in completing this questionnaire

If you have any questions about the study, please contact:

Jim Turner
University of Wales College of Medicine
Division of General Practice
Gwenfro Building
Wrexham Technology Park
Wrexham County Borough LL13 7YP

Telephone: 01978 316241/316242

Gethin Griffith
Institute of Medical & Social Care Research,
Wheldon Building
University of Wales, Bangor
Gwynedd
LL57 2UW.

Tel: 01248 383897

Please return the questionnaires in the enclosed pre-paid envelope.

FOLLOW-UP QUESTIONNAIRE

Prifysgol Cymru · University of Wales
BANGOR

UWCM

GenQuest

A study of the All-Wales Cancer Genetics Service

Dear service user,

Thank you for completing the first GenQuest questionnaire that was given to you by the All-Wales Cancer Genetics Service.

It is now time for you to complete the second questionnaire, which is enclosed. **Only fill in this questionnaire after receiving your risk status.**

If you did not return the consent form, to the address below, with the first questionnaire, would you please complete and return it with this questionnaire. I have enclosed another consent form in case the original is not to hand.

Yours faithfully,

Jim Turner

Jim Turner, University of Wales College of Medicine, Gwenfro Building, Wrexham Technology Park, Wrexham, LL13 7YP.
Tel. 01978 316241. Fax. 01978 311419

Prifysgol Cymru · University of Wales
BANGOR

UWCM

GenQuest

Your Views And Feelings About Our Service

Thank you for your help with this important study. The questionnaires should take about 50 minutes to fill in, but please do not feel compelled to complete them all in one session. Please read carefully the instructions for each question.

You will find that some of the questions are repetitive, and many individual questionnaires will be familiar to you from the last time, but we would be grateful if you could complete all of them anyway, as this information is important in helping us to develop our genetics service. The information you give here is confidential and there is no need for you to write your name on the questionnaires.

When you have completed the questionnaires, please check to make sure you have not missed anything out, then return them to us in the stamped addressed envelope provided. We look forward to receiving your completed questionnaires soon.

MANY THANKS FOR AGREEING TO TAKE PART IN THIS EVALUATION

PLEASE NOTE
Some of these questions or issues may not relate to you

Funded by:

The National Assembly of Wales
and
MacMillan Cancer Relief

Office Use Only: Personal Research No. .....Date of Issue. Qr.2

ID					

User Preferences for Cancer Genetics Services

Please state your name, address and contact telephone number.

Name:

Address:

Postcode:

Telephone number:

If you would like to ask any questions about completing this questionnaire please contact:

Gethin Griffith, Institute of Medical and Social Care Research, University of Wales, Bangor.
Telephone 01248 383897

We wish to find out what aspects of the cancer genetics service are important to users of the service. There are many different ways in which parts of the service can be provided. It is important to note that this survey is not trying to evaluate the service you actually had. Its aim is to find out which ways of providing the service you would prefer.

We would firstly like you to familiarise yourself with the different ways in which such a service could be provided.

1. Staff seen for counselling

Bearing in mind that a Consultant Geneticist will always be in overall charge of care; there are three different types of professionals that can provide genetics counselling. The options are:

- ***Specialist genetics nurse*** - a nurse who is trained in genetic counselling.
- ***Consultant geneticist*** - a hospital doctor who is trained in genetic counselling.
- ***Genetics associate*** - a scientist who is trained in genetic counselling.

2. Waiting time for letter

This refers to the time people have to wait between their doctor referring them to the genetics service and receiving a letter telling them their risk of developing familial cancer. The options are:

- **1 month**
- **2 months**
- **4 months**
- **6 months**

3. Distance to counselling

This refers to the distance people have to travel to their appointments with the genetics service. The options are:

- **20 miles**
- **40 miles**
- **60 miles**
- **80 miles**

4. Duration of counselling

This refers to the amount of time people spend with the genetics counsellor on a single appointment (this does not refer to the total time spent in the hospital, which may often be much longer). The options are:

- **30 minutes**
- **1 hour**
- **1 hour 30 minutes**
- **2 hours**

5. Availability of testing

This refers to the fact that currently genetic testing is only available to high risk clients and not to moderate or low risk clients. The options are:

- **High risk** - Testing only available to those at high risk.
- **All** - Testing available to all (whether high, moderate or low risk).

6. Cost of service

Please note that there is no possibility of people actually being asked to pay for the service they receive.

This item refers to the value you would place on cancer genetics services. To ascertain the value you place on cancer genetics services we would like you to consider how much you would be willing to pay a private provider or the NHS in the form of a single payment, one-off tax bill or national insurance contribution. Remember that any money you spend on genetics services would not be available to spend on other things. The options are:

- **£1,500**
- **£2,000**
- **£2,500**
- **£3,000**

Please tell us how important each of the aspects of the cancer genetics services are to you?
(For each aspect of care please circle the figure representing its importance).

	No importance	*Little importance*									*Very important*
Staff seen for counselling	0	1	2	3	4	5	6	7	8	9	10
Waiting time for a letter about risk of cancer	0	1	2	3	4	5	6	7	8	9	10
Distance to counselling	0	1	2	3	4	5	6	7	8	9	10
Duration of counselling	0	1	2	3	4	5	6	7	8	9	10
Availability of testing	0	1	2	3	4	5	6	7	8	9	10
Cost of service	0	1	2	3	4	5	6	7	8	9	10

Please rank these aspects of care in order of importance to you, using a scale of 1 – 6, where 1 = the most important and 6 = the least important, e.g. if staff seen for counselling is the most important place a 1 in the box opposite and if duration of counselling is the second most important place a 2 in the box opposite. Or if you have no preference then please tick ✓the box 'no preference'

	Rank	
• **Staff seen for counselling**	❒	*or*
• **Waiting time for a letter about risk of cancer**	❒	*no preference*
• **Distance to counselling**	❒	❒
• **Duration of counselling**	❒	
• **Availability of testing**	❒	
• **Cost of service**	❒	

Please state the reasons for your ranking .....
.....
.....
.....
.....

Please now rank the following types of genetics counsellor, from 1 = most preferred to 3 = least preferred. Or if you have no preference, please tick ✓the box 'no preference'.

Genetics associate	❒	**Genetics nurse**	❒	*or no preference*
Consultant geneticist	❒			❒

Please state the reasons for your ranking .....
.....
.....
.....
.....

Please now rank availability of genetics testing, from 1 = most preferred to 2 = least preferred. Or if you have no preference, please tick ✓the box 'no preference'.

High risk - Testing only available to those at high risk	❒	*or no preference*
All - Testing available to all (whether high, moderate or low risk)	❒	❒

Please state the reasons for your ranking .....
.....
.....

On the next page you will be asked to choose between options A and B in each of the 25 choices.
For each choice, imagine that your doctor has referred you to a genetics service for the first time.

- You will be asked to look at each choice separately and tick ✓the one you would prefer.
- Everything else about the options, apart from the differences stated, are the same.
- Please answer every choice remembering that there are no right or wrong answers. It is your views that we are interested in.

Here are two examples of completed choice questions.

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics associate
Waiting time for letter	4 months	1 month
Distance to counselling	60 miles	20 miles
Duration of counselling	30 minutes	30 minutes
Availability of testing	All	High risk only
Cost of service	£3,000	£1,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	✓	☐

The person responding to the question in this example prefers option B rather than option A.

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics associate
Waiting time for letter	1 month	1 month
Distance to counselling	40 miles	20 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	High risk only
Cost of service	£3,000	£1,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	✓	☐	☐

The person responding to the question in this example prefers option A rather than option B.

Please turn the page and choose one option from each of the following 25 choices.

Choice 1

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Consultant geneticist
Waiting time for letter	4 months	4 months
Distance to counselling	20 miles	40 miles
Duration of counselling	1 hour	30 minutes
Availability of testing	High	High
Cost of service	£1,500	£2,000

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 2.

Choice 2

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	4 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	2 hours	1 hour
Availability of testing	All	All
Cost of service	£2,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 3.

Choice 3

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	4 months	2 months
Distance to counselling	60 miles	40 miles
Duration of counselling	30 minutes	1 hour
Availability of testing	All	All
Cost of service	£3,000	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 4.

Choice 4

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics associate
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	40 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	High
Cost of service	£2,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 5.

Choice 5

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Consultant geneticist
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	20 miles
Duration of counselling	1 hour	1 hour
Availability of testing	All	High
Cost of service	£2,500	£1,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 6.

Choice 6

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	80 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	High
Cost of service	£2,500	£2,000

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 7.

Choice 7

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics associate	Genetics nurse
Waiting time for letter	2 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	30 minutes	1 hour
Availability of testing	High	All
Cost of service	£1,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 8.

Choice 8

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Consultant geneticist
Waiting time for letter	2 months	2 months
Distance to counselling	40 miles	60 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	All
Cost of service	£2,500	£2,000

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 9.

Choice 9

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	2 months	2 months
Distance to counselling	20 miles	40 miles
Duration of counselling	2 hours	1 hour
Availability of testing	High	All
Cost of service	£3,000	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 10.

Choice 10

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	2 hours	1 hour
Availability of testing	High	All
Cost of service	£2,500	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 11.

Choice 11

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	1 month
Distance to counselling	40 miles	80 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	High
Cost of service	£2,500	£3,000

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 12.

Choice 12

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Consultant geneticist
Waiting time for letter	2 months	1 month
Distance to counselling	40 miles	60 miles
Duration of counselling	1 hour	1 hour
Availability of testing	All	High
Cost of service	£2,500	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 13.

Choice 13

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	1 month
Distance to counselling	40 miles	40 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	All
Cost of service	£2,500	£1,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 14.

Choice 14

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics associate	Genetics nurse
Waiting time for letter	1 month	2 months
Distance to counselling	20 miles	40 miles
Duration of counselling	2 hours	1 hour
Availability of testing	All	All
Cost of service	£2,000	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 15.

Choice 15

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	1 month	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	30 minutes	1 hour
Availability of testing	High	All
Cost of service	£2,500	£2,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

Once you have ticked A, B or no preference, please go to choice 16.

Choice 16

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	6 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	1 hour	1 hour
Availability of testing	High	All
Cost of service	£2,000	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 17.

Choice 17

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics associate
Waiting time for letter	2 months	6 months
Distance to counselling	40 miles	60 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	High
Cost of service	£2,500	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 18.

Choice 18

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Consultant geneticist
Waiting time for letter	2 months	6 months
Distance to counselling	40 miles	40 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	High
Cost of service	£2,500	£3,000

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 19.

Choice 19

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	6 months
Distance to counselling	40 miles	20 miles
Duration of counselling	1 hour	30 minutes
Availability of testing	All	All
Cost of service	£2,500	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 20.

Choice 20

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	6 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	1 hour 30 minutes	1 hour
Availability of testing	All	All
Cost of service	£1,500	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 21.

Choice 21

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	60 miles
Duration of counselling	1 hour	2 hours
Availability of testing	All	High
Cost of service	£2,500	£1,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 22.

Choice 22

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Consultant geneticist	Genetics nurse
Waiting time for letter	4 months	2 months
Distance to counselling	40 miles	40 miles
Duration of counselling	30 minutes	1 hour
Availability of testing	High	All
Cost of service	£2,000	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 23.

Choice 23

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	2 months	4 months
Distance to counselling	40 miles	20 miles
Duration of counselling	1 hour	1 hour 30 minutes
Availability of testing	All	High
Cost of service	£2,500	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 24.

Choice 24

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics associate	Genetics nurse
Waiting time for letter	4 months	2 months
Distance to counselling	80 miles	40 miles
Duration of counselling	1 hour	1 hour
Availability of testing	All	All
Cost of service	£3,000	£2,500

	Prefer option A	Prefer option B	*or no preference*
Which option would you prefer? (*tick one box only*)	☐	☐	☐

Once you have ticked A, B or no preference, please go to choice 25.

Choice 25

Which would you prefer, option A, option B or do you have no preference on this item?

	OPTION A	OPTION B
Staff seen for counselling	Genetics nurse	Genetics nurse
Waiting time for letter	4 months	1 month
Distance to counselling	60 miles	40 miles
Duration of counselling	30 minutes	2 hours
Availability of testing	All	All
Cost of service	£3,000	£1,500

Which option would you prefer? (*tick one box only*)

Prefer option A ☐ Prefer option B ☐ *or no preference* ☐

How difficult/easy did you find the last 25 questions on choice of options? *(please circle)*

Very Difficult				*Moderate*				*Very easy*	
1	*2*	*3*	*4*	*5*	*6*	*7*	*8*	*9*	*10*

If you found the questions difficult please state why .....
.....
.....
.....

Were you aware, prior to contacting the cancer genetics service, that you could obtain cancer genetics testing from a private company? Yes ☐ No ☐

If you were aware that you could obtain cancer genetics testing privately.
Where did you find out about it? (e.g. Newspapers, friends, family GP etc.)
.....
.....
.....

Have you tried to contact a private company providing genetics testing? Yes ☐ No ☐

If you have tried to contacted a private company providing genetics testing, what happened?
.....
.....
.....
.....
.....

Please rank providers of genetics services in order of preference, from 1 = most preferred to 2 = least preferred. Or if you have no preference, please tick ✓the box 'no preference'.

NHS ☐
Private provider ☐
} *or no preference* ☐

Please state the reasons for your ranking
... ...
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If you have any comments regarding this questionnaire or matters arising from it, please include them here.
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Thank you for completing the questionnaire.

Please return the completed questionnaire in the enclosed pre-paid envelope.

Appendix K. Experiment protocol

Experiment protocol

1. All students entering Lecture Theatre 1 (Wheldon Building) are to be given a random number by Gethin. (Gethin will have randomised numbers in advance).

2. Once Gethin has ascertained how many students have turned up, they will be allocated into groups. 3 groups of up to 70 and 2 groups of up to 35.

3. Each group coordinator will be asked to lead their group to their allocated room.

Coordinator	Room	Student numbers
Gethin	Lecture Theatre 1	1 to 70 approx
Val	Teaching Space 1	71 to 140 approx
Caroline	Teaching Space 2	141 to 210 approx
Nonn	Room 114	211 245 approx
Abby	Practical Lab	246 – 280 approx

4. Students will be asked to sit down.

5. They will be asked to switch off their mobile phones, not to talk to each other until the experiment is completed and to remain seated until every one in their group has handed in their questionnaires. At the end of the study they will receive some further information

6. They will be informed that a list will be handed around during the experiment. They are to tick their name off when they receive the list. This will allow us to ensure that they receive their course and print credits.

7. The coordinator will show them page 31 of the questionnaire that asks them to stop. They will be told verbally that they should stop answering questions when they get to this point and that they will be asked to continue when it is appropriate.

8. Hand out questionnaires.

9. Coordinators will tell them that the information on the consent form is the same as that given to them on the information sheet when they were invited to participate in the study. They will then be asked to sign the informed consent form on the front of the questionnaire and then begin answering the questionnaire.

10. Coordinators should then observe and see when everybody has completed the first half of the questionnaire. Once everyone has completed the questionnaire, please hand out the information sheets next to the OHP. Once everyone has a sheet, switch on the OHP and read the contents to the class and then switch off the OHP. Then ask the students to forget about their previous answers and not to look back at them, they can then answer the remainder of the questionnaire.

11. Collect the information sheets.

12. Once all questionnaire have been completed and every one has ticked their names on the list you handed out, please hand out debrief sheets.

If you encounter any problems during the experiment do not leave the students. Please ring Gethin on 07887 500 272 (A member of The Institute of Medical and Social Care Research was available during the course of the experiment to go to any room and assist with any problems that arose e.g. obtain a qualified 'First aid' member of staff in the event of a medical problem.

Appendix L. Informed Consent Form

Ysgol seicoleg
Prifysgol Cymru, Bangor

Adeilad Brigantia, Ffordd Penrallt
Bangor, Gwynedd LL57 2AS

Ffôn (01248) 382211 - Ffacs (02148) 382599
e-bost: hology@bangor.ac.uk
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School of Psychology
University of Wales, Bangor

Adeilad Brigantia, Penrallt Road
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Tel: (01248) 382211 – Fax: (02148) 382599
e-mail: psychology@bangor.ac.uk
www.psychology.bangor.ac.uk

Informed Consent

Influences on Genetic Testing Decisions

I agree to participate as a volunteer in a scientific study as an authorised part of the research undertakings with the School of Psychology at the University of Wales, Bangor under the supervision of Dr. Val Morrison. The study and my part in it have been fully explained to me by either, Caroline White-Gwenin, Abbie Unwin, Nonn a'ch Dafydd, Val Morrison or Gethin Griffith and I understand their explanation. The procedures of this study have been explained to me.

I understand that I am free not to answer specific items on the questionnaire. I understand that all data will remain confidential with regards to my identity. I am free to withdraw my consent at any time and terminate my participation at any time without penalty. I understand that I may request a summary of the results from this study.

In the case of any complaints concerning the conduct of research, these should be addressed to Professor C. F. Lowe, Head of School, School of Psychology, University of Wales, Bangor, Gwynedd LL57 2DG or Professor I. Russell, Director, The Institute of Medical and Social Care Research, Wheldon Building, University of Wales, Bangor, Gwynedd LL57 2UW.

Participant's signature ..... Date .....

I the undersigned have fully explained the study to the above individual

Experimenter's signature ..... Date .....

C Fergus Lowe PhD, FBPsS
Athro a Phenaeth yr Ysgol • Professor and Head of School

Appendix M. Experiment Information

Negative Information

Firstly I would like you to think about the risk levels?
If you find that you do have increased risk for this disease, it is not saying that you will definitely develop the disease, but instead that you would have an 80% chance of developing the disease. Try to imagine what you would feel like if you worried about it then discovered that you never get the disease, or maybe that you suffer from something completely different.

If you found that you do not have increased chance of developing this disease, i.e. you only have an average chance of getting the disease, this would not mean that you will definitely not develop the disease, but that you only had an average chance of developing it. Try to imagine how you would feel if you got a good result and you became lax about your lifestyle, for example breast screenings, taking the pill, or smoking and then still developed the disease.

If you have increased risk of developing breast cancer, you could do regular self examinations to see if you have any small abnormalities. This will not ensure that you catch the cancer early enough or in fact that you catch it at all. Many people do not feel that they trust themselves to find any abnormality. Some people become very worried about detecting changes in their breasts and can end up doing it several times a day.

It is possible for people with high risk of developing cancer to have regular screening. However, a few doctors think that it is not good to have too many x-rays/mammograms as this in itself may actually increase chances of developing cancer.

Another form of screening, to detect small abnormalities, is ultrasound which causes no harm. However, some doctors feel that they are not as accurate as the mammograms.

The final possibility would be to have both breasts removed. This is obviously very traumatic. Many women who find that they have increased risk see this option as the most satisfactory.

Knowledge of increased risk to breast cancer may affect someone's insurance.

Some people become upset and distressed when they find that they have increased risk to breast cancer. An increased risk also has implications for one's family, as it may mean that they also have increased risk to breast cancer.

Positive Information

I would like you to imagine the following scenario.
Imagine that you are told that you are at increased risk of developing this disease.

If you know that you have an 80% chance of developing breast cancer, you have several options. I am going to tell you a bit about what you can do to reduce the likelihood of you developing this disease.

Firstly, you only have an 80% risk of developing breast cancer; it is not definite that you will develop it. It is therefore worth taking all the known actions to reduce your chance of developing any lumps in the first place. It is thought that healthy diet and exercise reduce cancer risks. Also stopping smoking and stopping taking the contraceptive pill could reduce your chances of getting breast cancer.

It would also be a good idea to do regular self examinations, to see if you have any small abnormalities.

It is possible for people with high risk of developing cancer to have regular screening, either mammograms or ultrasound.

The final possibility would be to have both breasts removed. This is most likely to prevent development of breast cancer.

Knowledge of increased risk to breast cancer may enable someone to prepare themselves for the future for example planning oneself financially, or emotionally.

An increased risk also has implications to one's family, as it may mean that they also have increased risk to breast cancer, and therefore they may also be able to take preventative measures.

Neutral/control Information

Now, I would like you to think about a more common illness, the common cold. This is traditionally thought to be spread by coughing and sneezing. It is thought that one can stop the germs by using a handkerchief. Research favours this idea. There is also evidence that infections may also be spread by passage of infected secretions on hands, either by direct contact or by intermediate objects. So, viruses can be spread by fingering the nasal area. Also hand washing is important and effective means of preventing the spread.
There is a popular belief that getting cold or wet causes colds, although it has been shown that significant lowering of the temperature of volunteers by immersion in cold water does not increase their susceptibility to infection.
Passive smoking has been shown to increase the incidence and severity of such infections.

If people know that they are carrying germs of the common cold, it seems advisable that they do as much as they can to try to not pass it on to others. Good precautions to take would be to wash ones hands carefully and to use a handkerchief. Also it might be best of all not to go to areas where there are going to be many people who could catch the cold.

There is no specific treatment for the common cold. Controlled trials of vitamin C have shown no clear-cut benefit. Nasal sprays are effective when given to asymptomatic family members of an affected person, but are ineffective as treatment. However, there are side effects of nasal sprays, such as nasal irritation.

Appendix N. Debriefing sheet

Debriefing sheet

Influences on Genetic Testing Decisions

We would like to thank you for participating in this study. You have just taken part in an experiment where you were in one of four groups. Each group was given different information:
Group 1: received neutral information.
Group 2: received negative information and then positive information.
Group 3: received positive information and then negative information.
Group 4: received positive information.

The neutral information was information on the common cold which had no relevance to the questions you were asked to answer. Both the positive and negative information related to genetic cancer, pre-cancer surveillance and methods of reducing the risk of developing cancer. Both the positive and negative information used in this experiment can be seen on the back of this sheet.

This was done because we are interested in seeing if information type or ordering affected your hypothetical intention to have genetic testing and your preferences for the way genetic services are provided. This will be examined in relation to Utility Theory, the Health Belief Model and the Theory of Planned Behaviour. We are also interested in the role of anxiety, family history and optimism, and how they relate to genetic testing intentions.

Your responses will be treated in the strictest of confidence, only combined and anonymised results will be presented in the final report and any publications. If you wish to withdraw your responses from the study, would like any additional information, or wish to know the results of the study please contact one of the research team on the e-mail addresses provided below. Notices will be placed in the reception of your department, giving you an opportunity to attend an oral presentation of results in 2003.

If any of the issues raised during your participation in the study have caused you distress, we apologise. We can answer any questions we you have, if you want to contact Dr. Val Morrison (e-mail below). Alternatively, if you would like to discuss these issues in confidence, please contact the student counsellor (3rd floor of the student's union, tel: 382024), Nightline (tel: 362121) or the Bangor Community Mental Health Team (26 College Road, tel: 370137).

Thank you once again for participating in this research.

Researcher's contact details:

Caroline White-Gwenin: psu86e@bangor.ac.uk
Abbie Unwin: psu844@bangor.ac.uk
Nonn a'ch Dafydd: psu184@bangor.ac.uk
Gethin Griffith: g.griffith@bangor.ac.uk

Under the academic supervision of Dr. Val Morrison: v.morrison@bangor.ac.uk

Negative information
Firstly I would like you to think about the risk levels?
If you find that you do have increased risk for this disease, it is not saying that you will definitely develop the disease, but instead that you would have an 80% chance of developing the disease. Try to imagine what you would feel like if you worried about it then discovered that you never get the disease, or maybe that you suffer from something completely different.

If you found that you do not have increased chance of developing this disease, i.e. you only have an average chance of getting the disease, this would not mean that you will definitely not develop the disease, but that you only had an average chance of developing it. Try to imagine how you would feel if you got a good result and you became lax about your lifestyle, for example breast screenings, taking the pill, or smoking and then still developed the disease.

If you have increased risk of developing breast cancer, you could do regular self examinations to see if you have any small abnormalities. This will not ensure that you catch the cancer early enough or in fact that you catch it at all. Many people do not feel that they trust themselves to find any abnormality. Some people become very worried about detecting changes in their breasts and can end up doing it several times a day.

It is possible for people with high risk of developing cancer to have regular screening. However, a few doctors think that it is not good to have too many x-rays/mammograms as this itself may actually increase chances of developing cancer.

Another form of screening, to detect small abnormalities, is ultrasound which causes no harm. However, some doctors feel that they are not as accurate as the mammograms.

The final possibility would be to have both breasts removed. This is obviously very traumatic. Many women who find that they have increased risk see this option as the most satisfactory.

Knowledge of increased risk to breast cancer may affect someone's insurance.

Some people become upset and distressed when they find that they have increased risk to breast cancer. An increased risk also has implications to one's family, as it may mean that they also have increased risk to breast cancer.

Positive Information
I would like you to imagine the following scenario.
Imagine that you are told that you are at increased risk of developing this disease.

If you know that you have an 80% chance of developing breast cancer, you have several options. I am going to tell you a bit about what you can do to reduce the likelihood of you developing this disease.
Firstly, you only have an 80% risk of developing breast cancer, it is not definite that you will develop it. It is therefore worth taking all the known actions to reduce your chance of developing any lumps in the first place. It is thought that healthy diet and exercise reduce cancer risks. Also stopping smoking and stopping taking the contraceptive pill could reduce your chances of getting breast cancer.

It would also be a good idea to do regular self examinations, to see if you have any small abnormalities.

It is possible for people with high risk of developing cancer to have regular screening, either mammograms or ultrasound.

The final possibility would be to have both breasts removed. This is most likely to prevent development of breast cancer.

Knowledge of increased risk to breast cancer may enable someone to prepare themselves for the future for example planning oneself financially, or emotionally.

An increased risk also has implications to one's family, as it may mean that they also have increased risk to breast cancer, and therefore they may also be able to take preventative measures.

ID				

All-Wales Cancer Genetic Service Evaluation.

Questionnaire for staff.

The University of Wales College of Medicine & the University of Wales, Bangor are conducting an independent evaluation of the All-Wales Cancer Genetic Service. Your participation in the evaluation by answering our questions is a valuable source of information for this process.

The information requested in this questionnaire.

To produce a unit costing of the All-Wales Cancer Genetics Service it is necessary to identify all care pathways and measure the resources required for each pathway. To minimise the number of questions you have to answer we have grouped cancers and risk status' that have identical pathways e.g. patients at high risk of breast cancer, high risk of ovarian cancer, high risk of colorectal cancer and at high risk of breast ovarian cancer. The questionnaire should not take more than 40 minutes to complete. We would like to ask you which tasks you perform or participate in, the average amount of time you spend on these tasks and on average the type and quantity of resources you use for each of these tasks.

Please tell us about your job details.

Name: ... ...

Job Title: ... ...

Grade: ... ...

Where is your main work location? (Please tick ✓one box only).

Based at all 3 sites ❒ Ysbyty Glan Clwyd ❒

University Hospital of Wales ❒ Singleton Hospital ❒

Do you work full time or part time? (Please tick ✓one box only). Full-time ❒ Part-time ❒

How many hours a week are you contracted to work per week for the NHS? ... ...hours

How many hours per week are you contracted to work for the Cancer Genetics Service? ... ...hours

In the following 6 tables, we would like you to tell us which tasks you perform or participate in, the average amount of time you spend on each task in minutes (do not include the time you are waiting for postal and telephone replies), the resources you use and how much you use.

Table 1: Tasks undertaken when a referral is received and the family history questionnaire is not returned.

	Task	**Do you participate?** (*Please circle one response*)	**Average amount of time you devote to the task in minutes?**	**What resources, and how much of them are required to perform the task?**
1	Record receipt of referral letter	**Yes** **No**	Minutes	…… …… …… …… ……
2	Input client details to database and issue family history questionnaire	**Yes** **No**	Minutes	…… …… …… …… ……
	Family history questionnaire not returned			
3	Issue 2nd family history questionnaire	**Yes** **No**	Minutes	…… …… …… …… ……

Table 1 (continued): Tasks undertaken when a referral is received and the family history questionnaire is not returned.

	Task	Do you participate? (*Please circle one response*)	Average amount of time you devote to the task in minutes?	What resources, and how much of them are required to perform the task?
	2nd family history questionnaire not returned			
4	Letter to patient, referrer and fill in ISCO database	**Yes** **No**	Minutes	…… …… …… …… ……

Table 2: Tasks undertaken for patients at low risk of breast, ovarian, colorectal or breast ovarian cancer.

	Task	**Do you participate?** (*Please circle one response*)	**Average amount of time you devote to the task in minutes?**	**What resources, and how much of them are required to perform the task?**
	Family history questionnaire returned			
1	Input family history into computer	**Yes** **No**	Minutes	…… …… …… …… ……
2	Review family history	**Yes** **No**	Minutes	…… …… …… …… ……
3	Letter and booklet to patient, letter to referrer and fill in ISCO database	**Yes** **No**	Minutes	…… …… …… …… ……

Table 3: Tasks undertaken for patients at moderate risk of breast cancer.

	Task	Do you participate? (*Please circle one response*)	Average amount of time you devote to the task in minutes?	What resources, and how much of them are required to perform the task?
	Family history questionnaire returned			
1	Input family history into computer	**Yes** **No**	Minutes	…… …… …… …… ……
2	Review family history	**Yes** **No**	Minutes	…… …… …… …… ……
3	Letter and booklet to patient and letter to referrer	**Yes** **No**	Minutes	…… …… …… …… ……

Table 3 (continued): Tasks undertaken for patients at moderate risk of breast cancer.

	Task	**Do you participate?** (*Please circle one response*)	**Average amount of time you devote to the task in minutes?**	**What resources, and how much of them are required to perform the task?**
4	Deal with consent form and patient's telephone call for a Breast Test Wales referral	**Yes** **No**	Minutes	…… …… …… …… ……
5	Refer patient	**Yes** **No**	Minutes	…… …… …… …… ……
6	Letter to patient and referrer if patient decides against having a Breast Test Wales referral.	**Yes** **No**	Minutes	…… …… …… …… ……
7	Fill in ISCO database	**Yes** **No**	Minutes	…… …… …… …… ……

Table 4: Tasks undertaken for patients at moderate risk of colorectal cancer.

	Task	Do you participate? (*Please circle one response*)	Average amount of time you devote to the task in minutes?	What resources, and how much of them are required to perform the task?
	Family history questionnaire returned			
1	Input family history into computer	**Yes** **No**	Minutes	…… …… …… …… ……
2	Obtain permission to access relatives medical records	**Yes** **No**	Minutes	…… …… …… …… ……
3	Obtain medical records and check them	**Yes** **No**	Minutes	…… …… …… …… ……

Table 4 (continued): Tasks undertaken for patients at moderate risk of colorectal cancer.

	Task	**Do you participate?** (*Please circle one response*)	**Average amount of time you devote to the task in minutes?**	**What resources, and how much of them are required to perform the task?**
4	Review family history	**Yes** **No**	Minutes	…… …… …… …… ……
5	Letter and booklet to patient and letter to referrer	**Yes** **No**	Minutes	…… …… …… …… ……
6	Discuss risk etc. if patient telephones	**Yes** **No**	Minutes	…… …… …… …… ……
7	Fill in ISCO database	**Yes** **No**	Minutes	…… …… …… …… ……

Table 5: Tasks undertaken for patients at moderate risk of ovarian or breast ovarian cancer.

	Task	Do you participate? (*Please circle one response*)	Average amount of time you devote to the task in minutes?	What resources, and how much of them are required to perform the task?
	Family history questionnaire returned			
1	Input family history into computer	**Yes** **No**	Minutes	…… …… …… …… ……
2	Obtain permission to access relatives medical records	**Yes** **No**	Minutes	…… …… …… …… ……
3	Obtain records and check them	**Yes** **No**	Minutes	…… …… …… …… ……

Table 5 (continued): Tasks undertaken for patients at moderate risk of ovarian or breast ovarian cancer.

	Task	Do you participate? (*Please circle one response*)	Average amount of time you devote to the task in minutes?	What resources, and how much of them are required to perform the task?
4	Review family history	**Yes** **No**	Minutes	…… …… …… …… ……
5	Letter and booklet to patient and letter to referrer	**Yes** **No**	Minutes	…… …… …… …… ……
6	Counselling session. Obtain patient's informed consent.	**Yes** **No**	Minutes	…… …… …… …… ……
7	Refer to Breast Test Wales and gynaecologist.	**Yes** **No**	Minutes	…… …… …… …… ……

Table 5 (continued): Tasks undertaken for patients at moderate risk of ovarian or breast ovarian cancer.

	Task	**Do you participate?** (*Please circle one response*)	**Average amount of time you devote to the task in minutes?**	**What resources, and how much of them are required to perform the task?**
8	Counselling session with Genetics Nurse and Gynaecologist at joint clinic.	**Yes** **No**	Minutes	…… …… …… …… ……
9	Fill in ISCO database.	**Yes** **No**	Minutes	…… …… …… …… ……

Table 6: Tasks undertaken for patients at high risk of breast, ovarian, colorectal or breast ovarian cancer.

	Task	Do you participate? (*Please circle one response*)	Average amount of time you devote to the task in minutes?	What resources, and how much of them are required to perform the task?
	Family history questionnaire returned			
1	Input family history into computer	**Yes** **No**	Minutes	... ...
2	Obtain permission to access relatives medical records	**Yes** **No**	Minutes	... ...
3	Obtain records and check them	**Yes** **No**	Minutes	... ...

Table 6 (continued): Tasks undertaken for patients at high risk of breast, ovarian, colorectal or breast ovarian cancer.

	Task	**Do you participate?** (*Please circle one response*)	**Average amount of time you devote to the task in minutes?**	**What resources, and how much of them are required to perform the task?**
4	Review family history	**Yes** **No**	Minutes	…… …… …… …… ……
5	Letter and booklet to patient and letter to referrer	**Yes** **No**	Minutes	…… …… …… …… ……
6	Preliminary counselling session.	**Yes** **No**	Minutes	…… …… …… …… ……
7	Refer to surgeon/BTW/gynaecologist/geneticist.	**Yes** **No**	Minutes	…… …… …… …… ……

Table 6 (continued): Tasks undertaken for patients at high risk of breast, ovarian, colorectal or breast ovarian cancer.

	Task	**Do you participate?** (*Please circle one response*)	**Average amount of time you devote to the task in minutes?**	**What resources, and how much of them are required to perform the task?**
8	Letter to patient and referrer.	**Yes** **No**	Minutes	…… …… …… …… ……
9	Fill in ISCO database.	**Yes** **No**	Minutes	…… …… …… …… ……
	Patient decides they want to have genetics testing			
10	Send informed consent form to affected relative.	**Yes** **No**	Minutes	…… …… …… …… ……

Table 6 (continued): Tasks undertaken for patients at high risk of breast, ovarian, colorectal or breast ovarian cancer.

	Task	**Do you participate?** (*Please circle one response*)	**Average amount of time you devote to the task in minutes?**	**What resources, and how much of them are required to perform the task?**
11	Record return of informed consent form.	**Yes** **No**	Minutes	…… …… …… …… ……
12	Letter to patient and referrer if affected relative refuses to participate.	**Yes** **No**	Minutes	…… …… …… …… ……
13	Fill in ISCO database.	**Yes** **No**	Minutes	…… …… …… …… ……

Table 6 (continued): Tasks undertaken for patients at high risk of breast, ovarian, colorectal or breast ovarian cancer.

	Task	Do you participate? (*Please circle one response*)	Average amount of time you devote to the task in minutes?	What resources, and how much of them are required to perform the task?
	Consent of affected relatives obtained			
14	Contact cancer affected relative by phone to arrange home visit.	**Yes** **No**	Minutes	…… …… …… …… ……
15	Home visit to relative affected with cancer to counsel and take blood (Please include average journey time).	**Yes** **No**	Minutes	…… …… …… …… ……
16	Send blood to laboratory.	**Yes** **No**	Minutes	…… …… …… …… …… ……

Table 6 (continued): Tasks undertaken for patients at high risk of breast, ovarian, colorectal or breast ovarian cancer.

	Task	**Do you participate?** (*Please circle one response*)	**Average amount of time you devote to the task in minutes?**	**What resources, and how much of them are required to perform the task?**
17	Test for mutation in affected relative's blood sample (Institute of medical genetics laboratory, Cardiff).	**Yes** **No**	Minutes	…… …… …… …… …… …… …… …… …… …… …… …… …… …… …… …… …… …… …… ……
18	Test for mutation in affected relative's blood sample (Private laboratory).	**Yes** **No**	Minutes	…… …… …… …… ……

Table 6 (continued): Tasks undertaken for patients at high risk of breast, ovarian, colorectal or breast ovarian cancer.

	Task	Do you participate? (*Please circle one response*)	Average amount of time you devote to the task in minutes?	What resources, and how much of them are required to perform the task?
19	Check to see if 12 months has elapsed and no mutation found. Send letter to patient and referrer.	**Yes** **No**	Minutes	…… …… …… …… ……
	Mutation found			
20	Contact cancer affected relative by Letter to arrange a clinic appointment.	**Yes** **No**	Minutes	…… …… …… …… ……
21	Counselling session at clinic.	**Yes** **No**	Minutes	…… …… …… …… ……

Table 6 (continued): Tasks undertaken for patients at high risk of breast, ovarian, colorectal or breast ovarian cancer.

	Task	**Do you participate?** (*Please circle one response*)	**Average amount of time you devote to the task in minutes?**	**What resources, and how much of them are required to perform the task?**
22	Letter to cancer affected relative and GP.	**Yes** **No**	Minutes	…… …… …… …… ……
23	Refer for appropriate annual screening for life.	**Yes** **No**	Minutes	…… …… …… …… ……
	Cancer affected relative informs referred patient that a mutation has been found			
24	1st presymptomatic counselling for referred patient. Take 1st blood sample.	**Yes** **No**	Minutes	…… …… …… …… ……

Table 6 (continued): Tasks undertaken for patients at high risk of breast, ovarian, colorectal or breast ovarian cancer.

	Task	Do you participate? (*Please circle one response*)	Average amount of time you devote to the task in minutes?	What resources, and how much of them are required to perform the task?
25	Send blood to laboratory.	Yes No	Minutes	...
26	Co ordinate clinic appointments.	Yes No	Minutes	...
27	2nd presymptomatic counselling for referred patient. Take 2nd blood sample.	Yes No	Minutes	...
28	Send blood to laboratory.	Yes No	Minutes	...

Table 6 (continued): Tasks undertaken for patients at high risk of breast, ovarian, colorectal or breast ovarian cancer.

	Task	**Do you participate?** (*Please circle one response*)	**Average amount of time you devote to the task in minutes?**	**What resources, and how much of them are required to perform the task?**
29	Mutation confirmation search in the Cardiff laboratory.	**Yes** **No**	Minutes	…… …… …… …… ……
	Mutation confirmation search in a private laboratory.	**Yes** **No**	Minutes	…… …… …… …… ……
30	Results session and post test counselling.	**Yes** **No**	Minutes	…… …… …… …… ……
31	Phone call to patient.	**Yes** **No**	Minutes	…… …… …… …… ……

Table 6 (continued): Tasks undertaken for patients at high risk of breast, ovarian, colorectal or breast ovarian cancer.

	Task	Do you participate? (*Please circle one response*)	Average amount of time you devote to the task in minutes?	What resources, and how much of them are required to perform the task?
32	Letter to patient, GP and referrer.	**Yes** **No**	Minutes	…… …… …… …… ……
33	Follow up clinic appointment (counselling).	**Yes** **No**	Minutes	…… …… …… …… ……
34	Refer patient with a mutation for appropriate annual screening for life.	**Yes** **No**	Minutes	…… …… …… …… ……
35	Fill in ISCO database.	**Yes** **No**	Minutes	…… …… …… …… ……

Table 6 (continued): Tasks undertaken for patients at high risk of breast, ovarian, colorectal or breast ovarian cancer.

	Task	**Do you participate?** (*Please circle one response*)	**Average amount of time you devote to the task in minutes?**	**What resources, and how much of them are required to perform the task?**
36	Co ordinate annual appointments.	**Yes** **No**	Minutes	…… …… …… …… ……
37	Annual clinic appointment for counseling.	**Yes** **No**	Minutes	…… …… …… …… ……
38	Fill in ISCO database.	**Yes** **No**	Minutes	…… …… …… …… ……

Do you believe that any tasks have been left out? Yes ☐ No ☐

Risk Status and cancer type	*Event(s)*
1. Referral is received and the family history questionnaire is not returned	... ...
2. Low risk of: breast ovarian colorectal breast ovarian	... ...
3. Moderate risk of: breast cancer	... ...
4. Moderate risk of: colorectal cancer	... ...
5. Moderate risk of: ovarian breast ovarian cancer	... ...
6. High risk of: breast ovarian colorectal breast ovarian	... ...

Notes

Appendix P. Sample Characteristics

Table P1

Sample Characteristics of Experiment Participants Prior to Applying Exclusion Criteria (N=158)

Demographic characteristics		
Age	Mean	20.2
	Median	18.0
	Standard deviation	6.0
Gender	Male	23 (16.3%)
	Female	118 (83.7%)
Parents	Yes	8 (5.6%)
	No	135 (94.4%)
Ethnicity	Armenian	1 (0.6)
	Ashkenazi Jewish	0 (0.0)
	Bangladeshi	1 (0.6)
	Black African	1 (0.6)
	Black Caribbean	1 (0.6)
	Chinese	42.5)
	Indian	1 (0.6)
	Japanese	1 (0.6)
	Latino	1 (0.6)
	Other	3 (1.9)
	Pakistani	0(0.0)
	Persian	1 (0.6)
	White	142 (90.4)
New socio economic classification	Higher managerial	15 (10.1)
	Professionals	50 (33.8)
	Lower managerial & professional	33 (22.3)
	Intermediate	9 (6.1)
	Small employer & own account workers	13 (8.8)
	Supervisors/craft related	12 (8.1)
	Semi-routine occupations	12 (8.1)
	Routine occupations	4 (2.7)

Psychological characteristics		
HADS anxiety scores	Mean	8.8
	Median	8
	Standard deviation	3.6
	Normal (0-7)	61 (38.6%)
	Mild (8-10)	51 (32.3%)
	Moderate (11-14)	34 (21.6%)
	Severe (15-21)	12 (7.6%)
HADS depression scores	Mean	3.9
	Median	4
	Standard deviation	2.6
	Normal (0-7)	143 (90.5%)
	Mild (8-10)	14 (8.9%)
	Moderate (11-14)	1 (0.6%)
	Severe (15-21)	0 (0%)
LOT dispositional optimism score	Mean	18.1
	Median	18
	Standard deviation	6.9

Appendix Q. New Socio Economic Classification

New Socio Economic classification (Rose & O'Reilly, 1998) data for the experiment participants and comparative data from the 2001 Population Census are presented in Tables Q1 and Q2. In order to improve coverage Rose and O'Reilly recommend that when employing the new socio economic classification that those not in employment are allocated according to their last main job. Household heads were allocated according to Rose and O'Reilly's recommendations in Table Q2 and subsequent analyses. Regardless of how non-workers are categorized (see Tables Q1 and Q2) the participants in this experiment are not representative of the national populations of Wales or England in terms of socio economic classification. As can be seen from Table Q1 and Q2, whilst 22% of the Welsh population (32.9% in Table Q1) and 27.3% of the population of England (38.2% in Table 61) are categorized as Higher Managerial (1A), Professional (1B) or Lower Managerial and Professional (2), 60.6% (68.4% in Table Q2) of the experiment participants came from households where the household head was in one of these categories. There is clearly a strong bias towards professional backgrounds amongst the participants in the study.

Table Q1

New Socio Economic Classification by Current Employment Status

New SEC Category	Experiment sample		2001 Census results for Wales		2001 Census results for England	
	N	%	N	%	N	%
Higher managerial	12	8.5	45,288	2.2	1,243,919	3.5
Professionals	47	33.1	77,368	3.7	1,816,039	5.1
Lower managerial & professional	27	19.0	333,165	16.1	6,656,918	18.7
Intermediate	6	4.2	166,135	8.0	3,366,759	9.5
Small employer & own account workers	13	9.2	146,595	7.1	2,479,472	7.0
Supervisors/craft related	10	7.0	161,807	7.8	2,526,120	7.1
Semi-routine occupations	6	4.2	254,268	12.3	4,139,697	11.7
Routine occupations	3	2.1	206,358	9.9	3,203,764	9.0
Never worked	0	0.0	56,822	2.7	964,978	2.7
Long-term unemployed	0	0.0	22,660	1.1	359,728	1.0
Full-time Students	11	7.7	150,263	7.2	2,498,729	7.0
Not classified/ Missing data	7	4.9	454,618	21.9	6,275,968	17.7

Figures for Wales and England were taken from the 2001 Population Census.

Table Q2

New Socio Economic Classification by Current or Last Main Job

New SEC Category	Experiment sample		2001 Census results for Wales		2001 Census results for England	
	N	%	N	%	N	%
Higher managerial	14	10.3	45,288	3.3	1,243,919	4.9
Professionals	48	35.3	77,368	5.6	1,816,039	7.1
Lower managerial & professional	31	22.8	333,165	24.0	6,656,918	26.2
Intermediate	7	5.1	166,135	11.9	3,366,759	13.2
Small employer & own account workers	13	9.6	146,595	10.5	2,479,472	9.7
Supervisors/craft related	10	7.4	161,807	11.6	2,526,120	9.9
Semi-routine occupations	9	6.6	254,268	18.3	4,139,697	16.3
Routine occupations	4	2.9	206,358	14.8	3,203,764	12.6
Not classified/ Missing data	6	-	454,618	-	6,275,968	-

Figures for Wales and England were taken from the 2001 Population Census.

Appendix R. Follow-up Benefits and Barriers

Table R1

Frequency with Which Each Benefit Was Cited at Follow-up

Benefit	**Reasons**								
	1st	**2nd**	**3rd**	**4th**	**5th**	**6th**	**7th**	**Total**	**%**
Prevent, surveillance, early treatment & inc health behaviou	62	38	31	13	3	1	1	149	34.7
Come to terms with the possibility of breast cancer /prepa	13	17	9	11	3	1	0	54	12.6
Help family come to terms with the prospect of breast cancer	5	20	15	8	5	0	0	53	12.4
Promote greater understanding of breast cancer/ awareness	16	17	12	3	3	2	0	53	12.4
Discover breast cancer susceptibility status	20	10	11	3	0	0	0	44	10.3
Social support / counselling	2	4	7	3	0	1	1	18	4.2
Reduce psychological distress/worry	4	5	3	4	1	0	0	17	4.0
Other psychological	5	6	0	0	1	0	0	12	2.8
Other biomedical	2	2	5	1	0	1	0	11	2.6
Family planning/children	1	1	3	1	1	0	0	7	1.6
Save lives	4	2	0	1	0	0	0	7	1.6
Other miscellaneous	1	1	1	1	0	0	0	4	0.9
Sub Total	**135**	**123**	**97**	**49**	**17**	**6**	**2**	**429**	**100.0**
Missing	7	19	45	93	125	136	140		
Total	142	142	142	142	142	142	142		

Table R2

Frequency with which each Barrier was Cited at Follow-up

Disadvantages	**Reasons**						
	1st	**2nd**	**3rd**	**4th**	**5th**	**Total**	**%**
Increased worry/ anxiety/distress/ depression	54	18	7	3	2	84	34.3
Life insurance, problems/ financial issues/ test expensive	16	9	4	2	0	31	12.7
Issues relating to the accuracy of the test/ No guarantee	8	7	3	0	0	18	7.3
Implications for the rest of the family	2	11	3	1	0	17	6.9
May not want to know genetic status	10	3	3	0	0	16	6.5
No longer worth living if susceptible/ suicidal	7	3	3	0	0	13	5.3
Not being able to plan/ waiting for cancer to occur	4	5	0	2	1	12	4.9
Playing god	1	4	3	1	1	10	4.1
Become complacent about lifestyle if not susceptible	3	3	3	0	0	9	3.7
Rejection/ genetic discrimination/ treated differently	4	2	1	1	0	8	3.3
Increased risk through mammography	5	1	1	0	0	7	2.9
Make patient fell uncomfortable/ not feel oneself	2	4	0	0	1	7	2.9
Other psychological	0	4	2	1	0	7	2.9
Other biomedical	2	1	2	0	0	5	2.0
Wary of health professionals/ counselling	0	1	0	0	0	1	0.4
Sub Total	**118**	**76**	**35**	**11**	**5**	**245**	**100.0**
Missing	24	66	107	131	137		
Total	142	142	142	142	142		

Table R3

Benefits and Barriers of Genetic Testing and Counselling Reported in Other Studies

Benefits	Barriers	Study
• To take extra precautions if the risk were high. • For reassurance that the risk was low.		Chaliki et al. (1995)
• To learn about their children's risk. • To increase use of cancer screening tests. • To take better care of oneself.	• Concerns about test accuracy. • Worry about insurance. • Emotional reactions. • Partner's reactions. • Family reactions.	Lerman et al. (1995)
• To increase screening. • To learn children's risk. • To take better care. • Suspecting being a carrier. • Just wanting to know. • To plan for future. • For preventative oophorectomy decision. • For child bearing decisions. • For preventative mastectomy. • For marital decision.		Struewing, Lerman, et al. (1995)
• To take action, e.g. 'So I can reduce my risk of developing the disease' • Because of a positive attitude to health, e.g. 'it is good to know' • Risk factor, e.g. 'I have a family history of the disease' • Just in case • Concern/anxiety (want good news) • Out of interest • Might as well • To emotionally prepare myself • Perceived risk e.g. 'I feel as if I will develop the disease' • Low barriers e.g. 'it is so easy to have the test' • To plan one's life • To increase one's information • The person would prefer to know • Perceived severity of the disease • For one's family • Concern/anxiety (expect bad news) e.g. 'I am worried that I have the disease and I want to check that I am right' • Low perceived risk, e.g. 'I don't feel as if I will develop the disease' • Relief of uncertainty e.g. 'I am worried, I have to know' • Check correct lifestyle.	• Barriers e.g. 'the test would be too much hassle' • Low perceived risk e.g. 'I don't feel as if I will develop the disease • Negative effect on life e.g. 'it would worry me' • No point in having the test • Risk factor e.g. I have no family history • Too young • Test no good. • Don't want to know • Too anxious • Unpleasant procedure • Negative affect about waiting for results • Affects insurance • Waste of time/resources • I don't think about it. • I don't want to think about it • Not anxious • Severity of the disease • Lack of knowledge • Would feel as if I am being hypochondriacal • Not my responsibility Ambiguous reasons.	Wroe et al. (1998)