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## **DOCTOR OF PHILOSOPHY**

### **Application of the capability approach to health economics research involving informal carers of people with dementia**

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**Application of the capability approach to health economics research  
involving informal carers of people with dementia**

by

**Carys Llywela Jones**

A thesis submitted to Bangor University in partial fulfilment of the  
requirements for the degree of  
Doctor of Philosophy

October, 2013

Centre for Health Economics and Medicines Evaluation,  
Institute of Medical and Social Care Research,  
Bangor University

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Finally, but by no means least, I wish to express heartfelt thanks to my family for their enduring encouragement and support in all my pursuits. I dedicate this thesis to them.

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## Abstract

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### *Background*

In the UK, the number of people with dementia is increasing along with life expectancy. Over half of the £23bn annual cost of dementia is due to informal care time. Therefore, there is an economic argument for identifying clinically-effective and cost-effective ways to maintain/improve carer quality of life (QoL). This thesis explores the suitability of a capability based instrument, the ICECAP-O, for measuring QoL in informal carers of people with dementia.

### *Methods*

Systematic reviews, qualitative interviews, survey data and clinical trial data were used. Construct validity of the ICECAP-O was examined using baseline data from the Challenge FamCare study of people with dementia and challenging behaviours, and an online survey created to collect additional data. A cost-effectiveness analysis was undertaken using a subsample of carers in the REMCARE trial of joint-remembrance therapy (RT).

### *Results*

The current evidence base for interventions to support carers of people with dementia is poor, with few economic evaluations existing. Qualitative work found the major themes affecting carer QoL overlapped well with capability instruments, suggesting this framework is suitable for carer research. Validity work found the ICECAP-O to be feasible and valid for use with carers. At the end of the 10 month RT trial, the mean difference in ICECAP-O scores between groups was -0.02 (bootstrapped 95% CI of -0.105 to 0.066) and the mean difference in costs was £1,464 (bootstrapped 95% CI of £758 to £2,313), RT was dominated by usual care. The cost-effectiveness acceptability curve indicated a 2% probability that RT was cost-effective at a willingness to pay threshold of £20,000 per point improvement and an 8% probability that RT was cost-effective at a threshold of £30,000 per point improvement on the ICECAP-O.

### *Conclusion*

To date, the ICECAP-O has not been used with carers of people with dementia; this thesis offers a unique insight into its suitability from theoretical validity through to applied research. When assessing the cost-effectiveness of interventions targeting outcomes other than physical functioning, I encourage researchers to include capability based instruments in an attempt to bridge the gap between measuring physical functioning and measuring a more encompassing quality of life.

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## Definitions

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**Adult Social Care Outcomes Toolkit (ASCOT):** An instrument to measure social care-related quality of life.

**Capability:** Refers to what individual's are able to do, as opposed to what they actually do (functioning).

**Carer Experience Scale (CES):** A six item instrument used to measure care-related quality of life.

**Challenge FamCare:** An NIHR funded cohort study (ISRCTN 58876649), involving family carers supporting people with dementia and challenging behaviours living in the community.

**Confidence Interval:** Refers to how frequently, from repeated independent samples from the target population, the population parameter would be expected to fall between the limits of the interval.

**Construct validity:** Refers to whether an instrument measures what it is supposed to measure.

**Content validity:** Refers to whether an instrument contains all domains required to measure what it is trying to measure.

**Convergent construct validity:** Refers to whether correlations occur where they would be expected to.

**Cost-benefit analysis:** An analysis which compares the costs and consequences of an intervention in monetary terms. Cross-programme comparison is possible between interventions in different sectors (health, transport, education) however; it is often difficult to assign a monetary value to benefits.

**Cost-consequence analysis:** An analysis where the incremental costs and consequences of an intervention are displayed without aggregating results into a single ratio.

**Cost-effectiveness analysis:** An analysis where the costs of an intervention are measured in monetary units and the consequences in natural units.

**Cost-minimisation analysis:** An analysis in which all outcomes are expected to be the same and the aim is to identify the least costly inputs required to achieve the common outcome.

**Cost-utility analysis:** An analysis where the costs of an intervention are measured in monetary terms and the consequences are measured in utility.

**Discounting:** A technique used to value the future benefits and costs of an intervention in terms of their present value. Discounting is used to reflect time preference i.e. costs and benefits occurring in the future have less value in the present than costs and benefits occurring today.

**Divergent construct validity:** Refers to whether a lack of correlation appears where no relationship would be expected.

**Dyad:** A pair consisting of a person with dementia and their carer.

**EQ-5D:** A generic, five item instrument used to measure health-related quality of life.

**EQ-VAS:** The visual analogue scale (VAS) element of the EQ-5D.

**Extra-welfarism:** Refers to the rejection of the traditional framework in welfare economics, where the sole goal is to maximise individual utility, in favour of incorporating other attributes such as the process of care or the characteristics of the individuals receiving care. In practice the concept of extra-welfarism has become synonymous with maximising health rather than utility.

**Gold standard:** The instrument or method that is accepted as the best current practice, and against which new instruments or methods should be compared.

**Health-related quality of life (HRQoL):** A measure which goes beyond mortality and morbidity to include domains such as social activity, general life satisfaction and cognition.

**ICECAP-A:** A five item instrument used to measure an individual's capability to achieve an outcome regardless of whether they actually carry out the functioning. Suitable for adults aged 18+.

**ICECAP-O:** A five item instrument used to measure an individual's capability to achieve an outcome regardless of whether they actually carry out the functioning. Suitable for adults aged 65+.



**Incremental cost:** The cost of producing one additional unit of output.

**Incremental cost-effectiveness ratio (ICER):** The cost per unit of benefit gained from moving to an alternative treatment.

**Inter-rater reliability:** Refers to the degree of stability occurring when a measurement is repeated in identical conditions but with different participants.

**Intervention:** A procedure carried out to improve, maintain or assess a particular outcome.

**Mapping:** A technique used to construct an algorithm for the conversion of a particular instrument score onto a different instrument.

**Marginal cost:** The cost of producing one additional unit.

**Modelling:** A technique used to build a pathway of choices and consequences. Modelling can be used to extrapolate from short-term outcomes to long-term outcomes.

**Opportunity cost:** The value of benefits foregone by not choosing the next best use of resources.

**Pharmacological:** Drug related.

**Psychosocial:** An interaction between social and psychological factors.

**Quality-adjusted life year (QALY):** A measure combining the length of time gained by an intervention with the value attached to the health state being experienced.

**Qualitative:** Observational data.

**Quantitative:** Numerical data.

**Randomised controlled trial (RCT):** A clinical trial where participants are randomly allocated between either an intervention or control group before the trial starts.

**REMCARE:** An NIHR HTA funded 8-centre trial of joint reminiscence therapy for people with dementia and their carers (ISRCTN 42430123).

**Sensitivity analysis:** A technique which tests the robustness of results by varying key parameters one at a time to see if the overall result is sensitive to the change.

**Short Form-6D (SF-6D):** A generic, six item instrument used to measure health-related quality of life.

**Social welfare:** The sum of welfare in society as a whole.

**Spillover:** The effect of an individual's illness on other individuals in close physical or emotional proximity.

**Standard gamble:** A valuation technique where the preferences for health states are elicited by asking respondents to choose between living the rest of their life in a particular health state or gambling, where the two possible outcomes are perfect health or death. The probability between winning (perfect health) or losing (death) are varied until the point where the respondent is indifferent between gambling or remaining in their current health state is reached.

**Statistical significance:** refers to the probability that the observed difference was due to chance if the null hypothesis is true.

**Time trade-off:** A valuation technique where the preferences for health states are elicited by asking respondents to choose between scenarios which involve trading length of life for quality of life until the point where they are indifferent is reached.

**Utility:** In health economics, utility refers to the relative preference for a particular health state or process outcome.

**Validity:** Refers to the degree to which a result is likely to be true and free of systematic error.

**Visual analogue scale (VAS):** An instrument where individuals are asked to mark on a scale from best to worst possible health state where they would rate their own health to be on that day.

**Welfare economics:** A branch of economics which analyses the social desirability of a set of outcomes in terms of the utility obtained by individuals, regardless of distribution.

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## Chapter 1: Introduction

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## **Statement of the problem**

With an ageing population the number of people affected by dementia is set to increase. Many people with dementia are supported in their own home by unpaid carers in the form of friends and family members. Without this support, the pressure on the health and social care sector would rise considerably. Therefore, there is a need to gather evidence on the most clinically effective and cost-effective way to maintain or improve the quality of life of informal carers. The National Institute for Health and Care Excellence (NICE) assesses the cost-effectiveness of new technologies and interventions. Treatments falling below the widely accepted threshold of £20,000- £30,000 per quality-adjusted life year (QALY) are considered cost-effective (NICE, 2013).

To measure the quality of life component of the QALY, NICE prefer the use of generic preference based utility measures such as the EQ-5D (NICE, 2013) (see Appendices 1 and 2, pages 180-181). Instruments which focus primarily (but not exclusively) on physical functioning aspects of health, such as the EQ-5D (EuroQoL group, 1990), are appropriate for use in interventions which aim to improve physical health; however, for complex psychosocial interventions involving populations such as informal carers of people with dementia, the aim is often to improve a broader aspect of quality of life than physical health alone. In this case, it is necessary to consider using additional instruments which are sensitive enough to detect changes in broader quality of life. The capability approach is a framework which allows outcomes other than health to be considered when evaluating societal utility and welfare.

This thesis explores the following overarching research questions:

- To what extent is the capability approach appropriate for measuring quality of life in informal carers of people with dementia?
- What implications does this have for future economic evaluations of interventions to support family carers of people with dementia?

## **Background and rationale for this thesis**

### *Dementia definition and prevalence*

Dementia is the term used to describe the collection of symptoms caused by damage to nerve cells in the brain. It is a progressive condition and symptoms include memory loss, a

decline in spatial awareness and reasoning, behavioural and mood changes, communication difficulties and a decline in physical ability (Alzheimer's Society, 2012a). While there are many different types of dementia, the four main types are Alzheimer's disease, vascular dementia, dementia with Lewy bodies and frontotemporal dementia (The Royal College of Psychiatrists, 2012).

Alzheimer's disease is believed to be the most common type of dementia (62% of all diagnoses) however a definitive diagnosis can only be achieved by a post-mortem. Alzheimer's disease is caused by excessive build-up of proteins around and inside neurones. The accumulation of excessive protein in the brain causes the death of neurones over time and this leads to deterioration of brain functioning. Vascular dementia is caused by either a single stroke (single-infarct dementia) or multiple strokes in an individual (multi-infarct dementia). The strokes cause damage to the blood vessels in the brain and thus disrupt the transportation of oxygen around the bloodstream. If brain cells become starved of oxygen they will eventually die and this causes the degeneration of brain functioning. Lewy body dementia is caused by a build-up of small proteins inside neurones, leading to the disruption of the chemical transmissions between nerve cells. Frontotemporal dementia occurs when the front lobes of the brain are damaged. This area of the brain is associated with behaviour so the damage can be manifested through changes in personality, behaviour and mood.

The number of people with dementia is estimated to be 35 million worldwide (Wimo & Prince, 2010), with over 800,000 people affected in the UK (Luengo-Fernandez, Leal & Gray, 2010). The prevalence of dementia is expected to increase as life expectancy increases (United Nations, 2009).

Dementia predominantly affects older people and estimates suggest that from the age of 30 onwards, the percentage of people affected by dementia doubles between each 5 year age category (Knapp et al., 2007). The Expert Delphi Consensus method was used to estimate the prevalence of dementia by age group in the UK. In the 65-69 age category, 1.3% of the population were estimated to have dementia. This rose to 32.5% of people in the 95+ age category. Rates were higher for women than men. In those aged over 65, dementia was estimated to account for 10% of male deaths and 15% of female deaths. The experts consulted as part of the Dementia UK panel believed that if the onset of dementia could be

delayed by five years, then the number of deaths attributed to dementia could be reduced from 60,000 to 30,000 per year. Although it is rarer in younger people, dementia is not exclusively an illness of older people; it is estimated that 8% of dementia cases are early onset as they occur in people under the age of 65 (Luengo-Fernandez et al., 2010).

### *Carers*

According to Carers UK, there are six million carers in the UK (Carers UK, 2009). This data is from the 2001 census, and only represents people who self-identify as a carer. Fifty-eight percent of carers were female, and women were more likely to give up working to provide care. The highest proportion of carers appeared in the 50 to 59 age category, with over 1 in 5 people in that category providing care.

Under the Carers (Equal Opportunities) Act, carers are defined as *'persons (who) provide, or intend to provide, a substantial amount of care on a regular basis'* (Department of Health, 2004). No clarification of what is deemed to be substantial or regular is provided, but care can cover a variety of tasks including physical help with self-care and housework, supervision of medication taking and assistance with shopping or financial planning.

In guidelines produced jointly by The National Institute for Health and Care Excellence (NICE) and the Social Care Institute for Excellence (SCIE) (NICE and SCIE, 2007), it is recommended that health and social care managers should ensure that carers of people with dementia receive the assessment of needs that they are entitled to, as outlined in the Carers (Equal Opportunities) Act 2004 (Department of Health, 2004). All SCIE reports include views of both formal and informal carers so the joint nature of the report with NICE has allowed the opportunity for the voice of those involved with the day to day work of caregiving to be heard on a wider platform. While the remit of NICE is very much focused on assessing the clinical-effectiveness and cost-effectiveness of health care technologies, SCIE is an independent charity funded by the Department of Health with the aim of identifying and promoting good clinical and social care practice. The carer's assessment is carried out by Social Services with the purpose of identifying additional services that the carer is entitled to and at what cost these services will be made available. Following the assessment a plan of action to support the carer, which might include breaks from caring commitments, is agreed.

The Carer's Strategy in England (Department of Health, 2010a) and the Carer's Strategy for Wales (Welsh Government, 2013) both provide frameworks for supporting carers. One of the ways that the strategy in England planned to do this was through the provision of additional resources for training GPs in their awareness of carer needs and the available support for carers. The need for reasonable breaks from the caring role, both to enable the continuation of caring in the longer term and allow the carer to have a life beyond caring, is highlighted in the Welsh strategy, and one of the key action points in the report is the need to consider how best to promote recreational breaks for carers.

The demands of caring can have an effect on both the physical and mental health of carers (Schulz & Sherwood, 2008). In the 2001 census, 21% of people who provided over fifty hours of care per week categorised themselves as not being in good health (Carers UK, 2004). In comparison, only 11% of non-carers would describe their health as not being good. Risks to physical health can include back pain from lifting the person being cared for. Common mental health problems for carers include stress, depression and social isolation. Depression has been found to affect up to 30% of carers for people with dementia (Schoenmakers, Buntinx & Delepeleire, 2010). Mental health issues can also affect physical health through increased blood pressure and the associated risks of heart disease and stroke. A longitudinal study conducted by Hirst (2005) followed carers and non-carers over six years. Respondents were asked to complete a questionnaire to assess mental health (the General Health Questionnaire-12), along with demographic information. Carers who had provided care over a long duration had relatively higher levels of mental distress compared to non-carers, and the risk of onset of mental distress became higher as the number of hours of care per week increased. Schulz and Sherwood (2008) used a stress-coping model framework to examine the existing literature of how caring can affect both physical and mental health. They used the stress-coping framework as they determined that the often unpredictable and uncontrollable nature of caring over an extended period of time induces stress in both the carer and the person being cared for. The detrimental effect on physical health was found to be lower than the effect on mental health. The effect on mental health was found to be moderated by older age, lower socioeconomic status and less availability of additional informal support. Predictors of poor mental health in carers were: the behaviour of the care recipient, the cognitive and functional impairment of the care recipient, the amount and duration of care provided, the age and sex of the carer, and the relationship between the carer and care recipient.

Interventions targeting carer health broadly fall into two categories; those aimed at reducing the amount of care provided (through improved efficiency or breaks), and those aimed at improving carer quality of life and coping skills (Sørensen et al., 2002). Sørensen et al. (2002) conducted a meta-analysis of the effectiveness of 78 carer interventions and found that larger effects were found in studies targeting increased carer knowledge than those targeting carer burden and depression. This suggests that improving quality of life for carers is challenging, and further research is required to explore ways to support the mental health of carers.

Financial worries can be a large source of concern for carers because many reduce their working hours or give up work completely to provide care. For those entitled to Carer's Allowance, the 2013-14 rate is £59.75 per week regardless of the number of hours spent caring. Carer's Allowance is also reduced by other benefits received by the carer, such as state pension. A survey of carers found that 66% of respondents were using their own savings or income to pay for care and 49% had made cutbacks to the amount they spent on caring support, such as equipment and breaks, due to financial strain (Carers UK, 2008). Eight-seven percent of carers felt that their financial situation was likely to deteriorate further in the future.

Although the physical and emotional demands of caring are well documented, the positive aspects are less well so. A longitudinal study of carers of people with dementia found that carers often reported stability with respect to depression and role overload over time (Gaugler, Davey, Pearlin & Zarit, 2000). Carers can feel a closer relationship to the person being cared for than had been experienced before the caring commenced. A sense of fulfilment can develop from feeling appreciated and valued by the person being cared for (Quinn, Clare & Woods, 2009).

#### *Costs of dementia to the economy*

The cost of dementia to the United Kingdom economy is estimated to be £23 billion annually. Of this, £12.4 billion is attributed to informal care provided by friends and family members, social care costs account for £9 billion due mainly to residential care, and £1.2 billion are accounted for by health care costs (Luengo-Fernandez et al., 2010). Following findings that patients with dementia admitted to hospital for a separate issue were found to be staying much longer than patients without dementia being treated for the



same illness (Alzheimer's Society, 2009), there is the potential to reduce the cost of dementia to the National Health Service (NHS) through improved case management of people with dementia admitted to hospital for a non-dementia related issue. The Alzheimer's Society report (2009) estimated that if the average length of stay of dementia patients was reduced by one week it could save the NHS £80 million annually. As well as freeing up bed space, an already stretched resource, there would also be a positive impact on the health of the person with dementia. The report found that there was often a noticeable deterioration in the health of people with dementia during hospital stays, with up to a third deteriorating to the point where they were admitted to a care home on discharge, rather than going home. Increases in confusion, distress and depression were common along with worsening communication skills leading to lower independence. A separate audit of dementia care in hospitals found that only 6% had a care pathway plan in place for people with dementia, with a further 44% of hospitals in the process of developing one (Royal College of Psychiatrists, 2011). Only 5% of hospitals had mandatory dementia awareness training for staff, yet 32% of staff questioned as part of the audit felt that they had sufficient dementia knowledge to carry out their role.

Dementia currently places a huge demand on both health and social care services and with the number of people affected by dementia both in the United Kingdom and internationally projected to increase, the strain on already limited resources will intensify. Dementia is a growing concern to the government (Department of Health, 2012) but historically research has not been as extensive as for other disorders. When combining the amount of research funding provided by governments and charities in the year 2008, cancer received £600 million, coronary heart disease received £170 million but dementia only received £50 million. The respective costs to the health and social care sector for each disease was £4.6 billion, £2.3 billion and £10.3 billion so the level of research funding does not reflect the relative costs of each disease (Luengo-Fernandez et al., 2010).

Two-thirds of people with dementia in the United Kingdom live in private residences in the community and are supported through unpaid care provided by friends and family members. As such a large proportion of the societal costs are borne by the informal care sector, there is an economic as well as a humanitarian argument for the effects of interventions on informal carers being considered in addition to the effects on the persons with dementia. It is desirable that carers are supported in their role and that the person with

dementia is able to live in the community for as long as possible before entering formal residential care.

### *Current policy*

In 2009 the National Dementia Strategy was released in England (Department of Health, 2009a) as a guideline for the best practice to facilitate an improved quality of life for people with dementia and their carers. The report proposed three key ways to improve services: through improved awareness of dementia; earlier diagnosis and intervention; provision of a higher quality of care once a diagnosis has been made. The need for improved awareness was the result of findings that the general population had a lack of knowledge of dementia; many people believe it to be a natural consequence of old age and subsequently do not seek specialist help for the symptoms. A perceived stigma surrounding dementia further contributes to people being unwilling to discuss it openly. Early diagnosis was highlighted as it gives people with dementia the opportunity to make choices for themselves while they are still able to; however, diagnosis needs to be conveyed sensitively with information and sources of further support made readily available. The Department of Health followed up their message for early diagnosis with a campaign to raise awareness of the early signs of dementia and encourage people to seek medical advice if they thought they were affected (Department of Health, 2011). In Wales, the National Dementia Vision for Wales was released in 2011 (Welsh Assembly Government, 2011). The report identified four priority areas for improved dementia care; improved service provision through better integration between health, social care and third sector agencies; improved early diagnosis; improved access to information and support; and improved training for those delivering care.

The Nuffield Council on Bioethics is an organisation that inspects ethical issues that arise from new developments in biology and medicine. In 2009 they released a report which provided an ethical framework for how to support people with dementia and their carers (Nuffield Council on Bioethics, 2009). In line with the National Dementia Strategy, the importance of early diagnosis was advocated but emphasis was placed on diagnosis being given at the appropriate time for each individual. It was suggested that some people in the early stages might find the worry of knowing that they have dementia outweighs the benefits of early diagnosis. The report also advised that health care professionals should actively encourage people with dementia to share the diagnosis with their family. This

recognises that family members typically play a significant part in providing support and meeting care needs and consequently need access to information which helps with their new role and the transition. A noteworthy point made in the report was that despite dementia being a progressive illness, the provision of care services is often split across health care and social care. This presents a problem when the needs of people with dementia are not seen as a high priority in the social care setting and the allocation of support resources is often not made until a crisis point is reached. The report recommended that support should be offered where needed, irrespective of whether it falls under the health care sector or social care sector. The potential conflict between the autonomy and quality of life of the person with dementia and that of their informal carer was also noted. This conflict can arise when the carer neglects their own needs, resulting in feeling overburdened and in need of a break from caring. The framework acknowledges that when the carer's quality of life is looked after the quality of care they provide is enhanced, leading to a positive knock-on effect on the quality of life of the person with dementia. Research has found that improving the quality of life of a family carer results in the person with dementia staying at home for a longer time before moving into long-term residential care (Mittelman, Haley, Clay & Roth, 2006).

Dementia research can historically be categorised into three main types: service redesign, pharmacological and psychosocial. With budgetary pressure on the NHS, trying to achieve the same outcome at a lower cost through service redesign is increasingly popular. The key to successful service redesign is identifying what currently works well and is thought of as best practice, and targeting areas where resources are not being used efficiently and can be reallocated. Interventions can be targeted either directly at the carer, or indirectly through affecting the quality of life of the person being cared for. Pharmacological interventions primarily target the person with dementia, but psychosocial interventions can target both the carer and the person being cared for as either individuals or dyads.

In the United Kingdom, in 2009 a Ministerial Dementia Research Summit was held to identify key areas of future research, covering both medical knowledge of dementia and also care services. Representatives from public bodies, industry, charities and voluntary organisations were present. Four care service areas of priority were identified: first, how to involve people with dementia in all stages of research, as current ethics often prevent people with severe dementia participating unless they are able to explicitly give consent;

second, the need to collate existing research and implement it into current practice was noted; third, the need for awareness that results from randomised controlled trials do not always translate directly into real world results, and that the physical and social environment of each person with dementia will influence their quality of care; fourth, as a result of the need to treat people on a person-centred basis there is a need for a service evaluation to establish what works best in each circumstance. Proposed solutions included conducting Cochrane reviews of qualitative studies of care and ensuring that results of existing studies are considered when care services are being developed. The need to increase research capacity in dementia care was also noted (Department of Health, 2009b).

### **Methodological perspective of this thesis**

#### *Quality of life*

Over recent years, quality of life has emerged as a key outcome for service delivery in health and social care sectors. A universally agreed and consistently used definition of quality of life is hard to find as the concept is dependent on context. In this thesis, which is focused on health economics research, I use the World Health Organisation's broad definition of quality of life (WHO, 1996):

*“Quality of life is defined as individuals’ perceptions of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns.”*

This definition allows quality of life measurement to take into account the perceived effect of an illness on a wide range of aspects of quality of life, rather than solely measuring the physical symptoms of the illness. In a survey of the general UK population, respondents were asked to list up to five things that affected quality of life. Over 60% stated relationships with family and other people, 43% stated their own health and 38% stated the health of somebody they were close to (Bowling, 1995). Well-being is conceptually distinct to quality of life, with its own lack of consistently used definition; well-being can refer to mental health alone or encompass other attributes such as life satisfaction. Well-being is often considered to be only one component of the broader concept of quality of life. Health is also a determinant of quality of life, and health-related quality of life refers to quality of life within the specific context of health, albeit including components such as functioning, degree and quality of social interactions, psychological well-being, somatic

sensations e.g. pain, adjustment and life satisfaction (Bowling, 1995). A review of theoretical health-related quality of life models by Taillefer et al. (2002) found that less than 40% of the articles defined quality of life, and 31% of quality of life models contained well-being as a variable. Functioning was a variable in 18% of the models and health was a variable in 15%.

### *Economic paradigms*

Economic analysis can be positive or normative. Positive economics is concerned with describing and predicting relationships between variables. Normative economics is concerned with making value judgements about the relative desirability of outcomes. While economists usually limit themselves to providing the evidence used to inform decisions rather than making decisions themselves, value judgements about how to measure the benefits and which costs to include will still influence their analysis. The three economic paradigms of interest in this thesis are all normative: welfarism, extra-welfarism and the capability approach.

### *Welfarism*

Welfare economics analyses the social desirability of a set of outcomes in terms of the utility obtained by individuals, regardless of equity or distributive justice (Sen, 1979). The two key (and normative) assumptions for welfare economics are:

1. Social welfare is the sum of each individual's utility.
2. Each individual is the best judge of their own utility.

To determine the societal preference for states of the world and the associated utility for each world state, it is necessary to aggregate individuals' preferences for that state. To do this, trade-offs between individuals' utility must be made and the framework used for this is the Pareto principle. Given an initial allocation of goods or services, a change to the allocation which makes at least one person better off but makes nobody worse off is called a Pareto improvement. Pareto efficiency is reached when no further Pareto improvements can be made. At Pareto efficient points, any further increase in utility can only be obtained by reducing another individual's utility. It is not possible to compare states where an individual's utility has increased at the expense of another individual's. There will be many different combinations of allocations that are Pareto efficient and the framework

does not offer a way of ranking different Pareto efficient states either. Kaldor (1939) and Hicks (1939) introduced the concept of compensation tests, which relax the Pareto principles. Under Kaldor-Hicks criterion, individuals gaining utility at the expense of others' utility would hypothetically pay the utility losers a monetary compensation. If the utility gainers would still be better off after paying the compensation then society as a whole has benefitted and a potential Pareto improvement has been made.

Cost-benefit analysis is the technique of choice for the welfarism framework, as all costs and outcomes are valued in monetary terms. An assumption of cost-benefit analysis is that all individuals have the same marginal utility of income and marginal utility of health as other individuals in the society; meaning that the level of monetary compensation required to pay to utility losers can be determined easily. However, in practice, individuals will have different marginal utilities to each other and it is unrealistic to assume that a unit of money has the same utility value to every individual in a society. In this case, the Kaldor-Hicks compensation tests are unlikely to help lead to (what we might judge as) an equitable distribution of utility across individuals in society. However, as stated earlier, concepts of equity or distributive justice are not considered in welfare economics. Morris, Devlin and Parkin (2007) presented the main arguments against the welfarism approach and they are summarised below:

1. While welfare economics has its roots in microeconomic theory, there is a concern as to how relevant rational choice and utility maximising behaviour is to health and health care behaviours.
2. In welfare economics utility is derived from consumption of a commodity. It is possible that utility can be derived from other sources, such as a family member's consumption of a good. Culyer called this, in relation to health care, the concept of 'caring externalities' (Culyer, 1976).
3. Welfare economics assumes that all individuals are interested in maximising their own utility, whereas some might have preferences toward voluntarily contributing to a societal good. This is similar to the concept of 'caring externalities' above.
4. Individuals' valuation of utility is affected by their personality and circumstances, meaning that measuring social welfare is difficult.

### *Extra-welfarism*

Non-welfarism rejects the principle of only valuing individuals' utility to determine societal welfare that is associated with the welfarism approach. Extra-welfarism is a form of non-welfarism which allows individual utilities to be taken into account when supplemented with additional non-utility information about a given state. This information can include attributes such as the process of care or characteristics of the individual receiving care. Allowing a focus on individual utility as opposed to societal utility is relevant for health and social care policy, as in practice it is often an aim of policymakers to reduce inequalities in the distribution of characteristics such as health, skills and education (Brouwer, Culyer, van Exel & Rutten, 2008). Extra-welfarism has its roots in Sen's work on functionings and capabilities (Sen, 1993), where an individual's valuation of a good is based not only on the utility arising from consumption but also the attributes of the good and their freedom to make choices. Functionings and capabilities are deemed by Sen to provide a more rational evaluative space for quality of life measurement than "opulence" (command over commodities) and utility (preferences) alone. The commodity requirements required to achieve a functioning will differ between individuals, therefore more information than the level of commodities available to an individual are necessary to determine their quality of life. Sen also noted that focusing solely on utility could mask the true quality of life status of individuals if they had developed "adaptive preferences" to their current status in society (Sen, 1995).

The goal in extra-welfarism is to maximise health output, rejecting the welfarism goal of maximising societal utility. Quality of life measurement is consistent with this framework, and cost-effectiveness and cost-utility analysis are used in the extra-welfarism paradigm. In cost-effectiveness analysis the outcome is measured in natural units and in cost-utility analysis the outcome of choice is the quality-adjusted life year (QALY), a unit or index combining length of life with quality of life. Whether QALYs, and their focus on physical health functioning when calculated using the EQ-5D, are the best estimation of health output is open to debate. Further discussion of the EQ-5D and its emphasis on physical functioning is in the Instruments section, page 20.

### *The capability approach*

The capability framework is even broader than the extra-welfarism framework as it allows outcomes other than health to be considered. Sen's capabilities work developed a general

concept for quality of life, rather than being specific to health related quality of life; however, the capability approach has been applied in a range of research areas including poverty, inequality, social justice, health and identity (Clark, 2006). The capability approach to quality of life measurement focuses on individuals' capability to function, i.e. the things which individuals are able to be or do, which is in contrast to the focus on individuals' utility. Focusing on capability to function rather than actual functioning is important, as sometimes individuals choose not to undertake a particular functioning. It is also worth noting that the utility derived from a functioning is distinct from the actual functioning itself.

Many health care interventions, particularly public health interventions, have effects on other sectors such as the education or social service sectors. Therefore, there is a need to reflect these benefits with broader outcome measures. Using the extra-welfarism framework of only considering health benefits accruing as a result of an intervention will underestimate the total benefit to all sectors of society. Furthermore, programmes only measuring health-related outcomes will be at a disadvantage when funding decisions are made (Coast, Smith & Lorgelly, 2008a). For carers of people with dementia, measuring physical health-related outcomes inappropriately places a 'patient' identity on carers (Al-Janabi, Flynn & Coast, 2011a).

Sen's capabilities work was concerned with social justice; this is opposed to the focus on economic growth, which tells us nothing about the quality of life of the members of a society. Sen did not provide a list of desirable functionings or capabilities, but Nussbaum (2003) suggested the following ten basic human capabilities: life; bodily health; bodily integrity; sense, imagination and thought; emotions; practical reason; affiliation; other species; play; control over one's environment. The list was developed from an initial notion of the dignity of human life and what is required to live a life with dignity. All ten capabilities are envisioned to be mutually supportive and of equal importance, though Nussbaum notes that priorities often have to be set for practical reasons. These capabilities are closely related to the human rights approach; however, the human rights approach has been criticised for being male centric (Nussbaum, 2003). Nussbaum's capabilities explicitly incorporate promotion of gender equality. Nussbaum declared that all individuals are entitled to a minimum level of these ten basic capabilities, without specifying what the minimum threshold level should be.



As well as the ambiguity of lack of specified capabilities by Sen, it is not clear whether the capability approach aims to maximise the sum of societal capability or whether equitable considerations are taken into account and the aim is to attain a minimum level of capability for all members of a society (Coast et al., 2008a). A potential solution for addressing the issue of equity distribution would be the weighting of QALYs, a concept which involves making value judgements (Donaldson et al., 2011). Alan Williams (1997) expanded the concept of ‘fair innings’ originally introduced by Harris (1985), whereby all members of a society are entitled to a minimum span of health, which could be expressed in life years. Those not achieving the minimum are deemed to have been cheated and those achieving more than the minimum are ‘living on borrowed time’. A distinction between the ‘fair innings’ concept and the capability approach is that the fair innings concept is entirely outcome based, whereas the capability approach can include resource and process-based information.

A number of instruments consistent with the capability approach have been developed. The ICECAP-O (see Appendix 3, page 182) was designed specifically to measure capabilities in adults aged 65+ (Grewal, Lewis, Flynn, Brown, Bond & Coast, 2006). The ICECAP-O is described in more detail in the Methods section, page 21. A version suitable for adults aged 18+, the ICECAP-A, has also been developed (Al-Janabi, Flynn & Coast, 2012). The Carer Experience Scale (see Appendix 4, page 183) (Al-Janabi, Flynn & Coast, 2011b) was developed to measure the caring experience. An outcome measure which captures the quality of social care, the Adult Social Care Outcomes Toolkit (ASCOT) (see Appendix 5, page 184) (Netten et al., 2011) is also available. Out of these capability based measures, published preference based utility weights are currently only available for the ASCOT.

#### *Qualitative framework analysis*

In Chapter 4, the results of qualitative research involving carers of people with dementia are presented. The framework approach was used for this analysis. Framework analysis was developed in the 1980s to analyse qualitative data gathered in applied policy research, an area with highly focused objectives (Smith & Firth, 2011). It is the opposite of more inductive approaches, such as grounded theory, as the focus is not on developing a new theory but instead on describing and interpreting participants’ views. Framework analysis

is popular as it encourages a systematic, structured approach to the analysis of qualitative research data (Ritchie & Lewis, 2003).

### **Purpose of this thesis**

The ‘reference case’ in the NICE guide to the methods of technology appraisal sets out guidance for assessing the clinical and cost-effectiveness of new and existing technologies (NICE, 2013). The guidance stipulates that when measuring the clinical effectiveness of an intervention the impact on the carer as well as the patient must be considered: ‘...*health benefits and adverse effects that are important to patients and/or their carers.* (p14). The ‘reference case’ also states that the preferred outcome measure for eliciting changes in health-related quality of life is the quality-adjusted life year (QALY) as measured by the EQ-5D (EuroQoL group, 1990).

The need to use appropriate outcome measures in health economics research has been identified (Payne, McAllister & Davies, 2012; Kelly, McDaid, Ludbrook & Powell, 2005; Craig, Dieppe, Macintyre, Michie, Nazareth & Petticrew, 2008). To be appropriate, an instrument has to be able to measure changes in the area that an intervention is targeting; if the main aim of an intervention is improved mental health, the primary outcome measure should be sensitive to changes in mental health. Focusing on a physical functioning based health measurement is not always appropriate in complex interventions which target multiple outcomes. The capability framework might offer a broader measure of quality of life than physical functioning alone in research involving carers of people with dementia.

The purpose of this thesis is to explore the theoretical suitability, validity and application of a capability based instrument, the ICECAP-O, to research involving carers of people with dementia.

### **Thesis novelty and contribution to knowledge**

The use of the ICECAP-O has not previously been explored in a research setting involving informal carers of people with dementia living in the community. This thesis offers a novel opportunity to:

- Increase the evidence base for the capability approach with carers of people with dementia.

- Support evidence based decision-making about the use of public resources to support this group of people in society.

### **Research questions**

The research questions addressed by this thesis are as follows:

1. What existing literature is there on the effectiveness and cost-effectiveness of interventions to support informal carers of people with dementia?
2. What outcome measures are used most frequently in interventions involving carers of people with dementia, and how useful are these measures for economic evaluation?
3. What do family carers of people with dementia perceive as affecting quality of life, and how well do capability based instruments capture these aspects of quality of life?
4. To what extent is the ICECAP-O valid for use with carers of people with dementia?
5. To what extent does the ICECAP-O capture the effect of a psychosocial intervention of joint reminiscence therapy for carers of people with dementia?

The structure of the thesis and how the research questions link together is shown in Figure 1.1 and described further in the Methods section of this chapter.

### **Wider generalisability of this thesis**

While this thesis is focused on exploring the application of the capability approach to measuring quality of life in informal carers of people with dementia, I believe that it has a wider generalisability. Its findings are relevant to other groups of informal carers, such as carers of disabled people or carers of people with different chronic conditions, whose physical health (as measured by the EQ-5D) may not be affected by an intervention. There is a need to consider the use of additional, capability based outcome measures in research with these carers. I have focused on informal carers of people with dementia as dementia is a major health challenge both for the United Kingdom and internationally.

### **Author contribution**

My contribution to this thesis (Carys Jones: CJ) and that of my supervisory team (Professor Rhiannon Tudor Edwards: RTE; Dr Barry Hounsome: BH) to each chapter is outlined below.

*Chapter 1:* Initial draft by CJ. RTE and BH gave conceptual advice and critical comments.

*Chapter 2:* Literature searching, data extraction and initial draft by CJ. RTE and BH gave conceptual advice and critical comments.

*Chapter 3:* Literature searching, data extraction and initial draft by CJ. RTE and BH gave conceptual advice and critical comments.

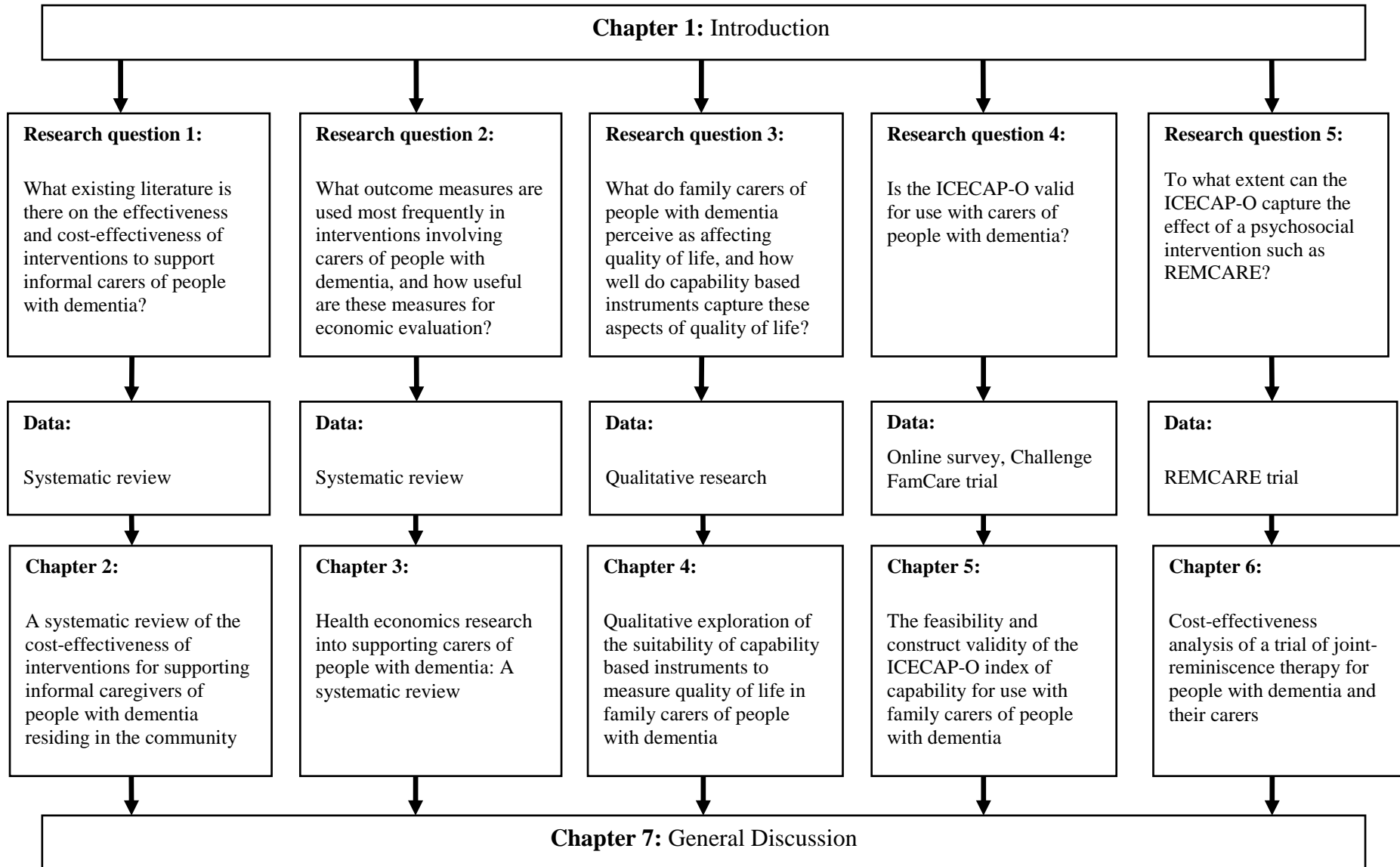
*Chapter 4:* Interviews conducted, transcribed and analysed by CJ. Initial draft by CJ. RTE and BH gave conceptual advice and critical comments.

*Chapter 5:* Online survey designed in SurveyMonkey and analysed by CJ. Ethical amendment to include the ICECAP-O arranged by CJ. Challenge FamCare data released by the Principal Investigator, Professor Esme Moniz-Cook (EMC). Data analysis and initial draft by CJ. RTE and BH gave conceptual advice and critical comments. EMC gave critical comments.

*Chapter 6:* Ethical amendment to include the ICECAP-O arranged by CJ. REMCARE data released by the Principal Investigator, Professor Bob Woods. Data analysis of ICECAP-O subsample by CJ. Initial draft by CJ. RTE and BH gave conceptual advice and critical comments.

*Chapter 7:* Initial draft by CJ. RTE and BH gave conceptual advice and critical comments.

**Figure 1.1:** Structure and layout of the thesis



### **Instruments used in this thesis**

To conform with NICE guidelines, health economists require preference based utility measures to weight QALYs (NICE, 2013); other health service researchers might use quality of life measures which do not meet this requirement but still offer information on the effectiveness of an intervention. This thesis focuses on the appropriateness of capability based instruments, in particular the ICECAP-O (see Appendix 3, page 182), for measuring quality of life in carers of people with dementia. The suitability of the EQ-5D (see Appendices 1 and 2, pages 180-181) is discussed, along with two other capability based instruments: the Carer Experience Scale (CES) (see Appendix 4, page 183) (Al-Janabi et al., 2011b) and the Adult Social Care Outcomes Toolkit (ASCOT) (see Appendix 5, page 184) (Netten et al., 2011).

The EQ-5D has been in use since the 1990's and is a commonly used instrument in health economics research. It is generic, which means it can theoretically be used to measure health-related quality of life in research involving any illness. The EQ-5D has two parts, the EQ-5D questionnaire and the EQ-VAS visual analogue scale. The EQ-5D part is a generic preference based measure with five domains. The domains are mobility, self-care, usual activities, pain/discomfort and anxiety/depression. Each domain has three possible levels: no problems, some problems and extreme problems. A total of 243 different health states can be described by the EQ-5D. The EQ-VAS is not preference based. It is a thermometer with markings from 0 (worst imaginable health state) to 1 (best imaginable health state). Respondents are asked to draw a line to the thermometer indicating how they would rate their own health at that moment. The EQ-5D is known to be prone to 'ceiling effects', where respondents frequently choose the highest level. A survey of the 11,248 members of the US general population found that 47% of respondents indicated that they had no problems when assessed using the EQ-5D, but when assessed using a different generic health-related quality of life measure- the SF-6D, only 6% rated themselves as being in full health. The EQ-5D was also found to be poor at detecting mental health problems, with 49% of respondents who indicated having no problems on the EQ-5D selecting "a little" or "some" problems with feeling tense or downhearted and low (Bharmal & Thomas, 2006). The finding that the EQ-5D is less sensitive than the SF-6D at detecting lower mental health was echoed in a study of the two instruments in mental health patients (Lamers et al., 2006), where more respondents reported having no problems

on the EQ-5D compared to the SF-6D. 65% of respondents reported having problems most or all of the time due to mental health on the SF-6D, compared to only 33% reporting extreme problems on the anxiety/depression domain of the EQ-5D. While the EQ-5D has the advantage of being concise and easy to use, its emphasis on physical functioning leads to a need to consider other outcome measures which are better able to detect quality of life changes in populations which don't expect to see large changes in their physical health as the result of an intervention, e.g. carers.

The ICECAP-O is a newer instrument (Grewal et al., 2006). It measures an individual's capability to achieve an outcome regardless of whether they carry out the functioning, for example, respondents are asked whether they *can* have all the love and friendship that they want rather than if they *have* all the love and friendship that they want. The domains of the ICECAP-O are love and friendship, thinking about the future, doing things that make you feel valued, enjoyment/pleasure and independence. There are four possible levels for each domain: I can have all, I can have a lot, I can have a little and I cannot have any. Even though the ICECAP-O is a capability based measure, it is appropriate to consider using it for quality of life research as the domains were developed to capture areas that affect a broader quality of life than physical health alone. The ICECAP-O was designed specifically for use with people aged 65+. As many carers of people with dementia are themselves older, particularly spousal carers, it is appropriate to consider the use of the ICECAP-O for use in research in this population.

The Carer Experience Scale (CES) was developed to measure care related quality of life rather than health-related quality of life (Al-Janabi et al., 2011b). One significant feature of the CES is that it covers both the positive and negative aspects of caring. The six domains of the CES are: activities outside caring, support from family and friends, assistance from organisations and the Government, fulfilment from caring, control over caring and getting on with the person you care for. Each domain has three possible levels: a lot, some and little.

The Adult Social Care Outcomes Toolkit (ASCOT) measures social care related quality of life for care recipients (Netten et al., 2011), but the domains might have relevance for quality of life of carers too: control over daily life; personal cleanliness and comfort; food

and drink; personal safety; social participation and involvement; occupation; accommodation cleanliness and comfort; and dignity.

### **Data sources used in this thesis**

This thesis uses four sources of primary data: qualitative interviews, REMCARE trial data, Challenge FamCare trial data, and survey data.

Qualitative interviews were conducted with eight family carers between November 2010 and July 2011. The aim of the interviews was to elicit the positive and negative ways in which caring can affect quality of life, and hence identify whether capability based instruments such as the ICECAP-O or CES would be suitable for use in research involving carers of people with dementia. Carers were recruited using two methods: first by the researcher giving a short presentation at local Alzheimer's Society cafes in North Wales, and second by distributing a study information sheet to carers registered with NEURODEM (Wales Dementias and Neurodegenerative Diseases Research Network), a network for people with memory problems and their carers. Semi-structured one-to-one interviews were conducted at a location that the participant chose, typically their home. Interviews lasted up to an hour and were recorded using a digital recorder and note taking. Interviews were then transcribed verbatim, taking care to remove any identifying information such as participant names. Interpretive phenomenological analysis (IPA) was used to code the data. IPA was used as the interviews were designed to probe the participants' subjective view of their own experiences. The domains of the ICECAP-O, ASCOT and CES were set as predetermined codes, and additional codes were derived through repeated reading of the interview transcripts.

REMCARE was a NIHR HTA funded trial (ISRCTN 42430123) of joint-remembrance therapy for people with dementia and their carers (Woods et al., 2009; Woods et al., 2012). Participants were randomised to receiving reminiscence therapy following the 'Remembering Yesterday, Caring Today' model (Schweitzer & Bruce, 2008), or usual care. Dyads randomised to receive reminiscence therapy attended reminiscence groups which used photographs, music and other objects to stimulate memories in the person with dementia. The inclusion of carers in the reminiscence groups was to encourage an improved relationship between the dyads. REMCARE was conducted at 8 sites across England and Wales. Reminiscence groups met weekly for 3 months, and then monthly for



a further 7 months. Outcome measures were collected at baseline, 3 months and 10 months. The primary outcome measure for the trial was the Quality of Life in Alzheimer's Disease (QOL-AD) for the person with dementia and the General Health Questionnaire (GHQ-28) for the carer. Secondary outcome measures included the Autobiographical Memory Interview (AMI), Quality of the Carer-Patient Relationship (QCPR), Cornell Scale for Depression in Dementia (CSDD), Rating Anxiety in Dementia (RAID), Hospital Anxiety and Depression Scale (HADS), Relative's Stress Scale (RSS) and the EQ-5D. General demographics and resource use data were also collected. The REMCARE trial was already set up and running when an amendment was submitted to include the ICECAP-O measure for carers. As such, ICECAP-O data were only available for REMCARE participants who entered the trial after the amendment was ethically approved by the Research Ethics Committee for Wales.

Challenge FamCare was an NIHR funded cohort study (ISRCTN 58876649) involving family carers supporting people with dementia who exhibited challenging behaviour. Outcome measures were collected at baseline, 2 months and 6 months. The primary outcome measure was the Revised Memory and Behaviour Problems Checklist (RMBPC). Secondary outcome measures for the carer were the General Health Questionnaire (GHQ-12), EQ-5D, ICECAP-O, HADS, Short Sense of Competence Questionnaire (SSCQ), Guilt Scale and Relatives Stress Scale (RSS). General demographics and resource use data were also collected.

The fourth data source was an online survey created in SurveyMonkey to gather data for a construct validity analysis of the suitability of the ICECAP-O for measuring quality of life in carers of people with dementia. Ethical approval was received from Bangor University and the survey ran between June 2011 and April 2012. The survey consisted of the ICECAP-O, EQ-5D and a number of demographic questions. To be eligible to take part, participants had to be current or former carers and aged 65+. Participants were recruited through several routes including a Uniting Carers mail out, posting on the Alzheimer's Society Online Forum and a notice in the University of the Third Age newsletter. Participants had the option of completing the survey online or requesting a postal copy. Carers who completed the survey by 31<sup>st</sup> December 2011 were entered into a prize draw to win one £250 Marks and Spencer voucher as an incentive to take part.

## **Thesis structure**

Chapter 1 has provided a general introduction to the topic and specifically the importance of carer quality of life as a research area within health economics. The novel contribution of this thesis is presented in this chapter, along with the specific research questions and how they will be answered (see Figure 1.1, page 19).

Chapter 2 provides a more detailed discussion of the existing evidence from interventions to support carers of people with dementia. To do this, a systematic literature search was conducted using a range of electronic databases. Only studies reporting a cost and health-related outcome measure for unpaid carers of people with dementia living in the community were included. No restrictions were placed on intervention target (person with dementia or their carer), intervention type (pharmacological, service delivery or psychosocial), location, gender or other carer characteristics. Studies passing the initial screening of title and abstract were retrieved for review. The following characteristics were extracted from studies meeting the systematic review criteria: intervention type, country, study design and follow-up period, number of participants, perspective of analysis, cost results and health-related outcome measure results for carers. Study quality was assessed using the Drummond ten item checklist for economic evaluations (Drummond, Sculpher, Torrance, O'Brien & Stoddart, 2005). A narrative approach was used to synthesise the main clinical and cost-effectiveness findings. An edited version of Chapter 2 has been published in *International Psychogeriatrics* (Jones, Edwards & Hounsome, 2012a).

Chapter 3 addresses the issue of which instruments are the most frequently used to measure quality of life in carers of people with dementia through a review of outcome measures used in published studies. As stated earlier, the NICE guidance for technology appraisal advocates the use of QALYs to measure cost-effectiveness (NICE, 2013). This allows cross-programme comparisons across interventions and illnesses to be made in a consistent manner. The recommended instrument to measure the health-related quality of life component of the QALY is the EQ-5D. However, as the domains covered by the EQ-5D are predominantly concerned with physical health there is a question as to how relevant it is for use in research involving carers of people with dementia. The role of caring for a person with dementia can be physically demanding if the carer assists with tasks such as lifting and attending to the toileting needs of the person being cared for. To carry out these tasks many carers already have a reasonable level of physical health. Therefore,

interventions to help promote the mental health of carers, rather than physical health might be more effective. Instruments such as the EQ-5D are not entirely appropriate in this setting as they are focused on physical health rather than mental health. As such, other instruments need to be considered. For Chapter 3, an electronic database search was conducted to find interventions with outcome measures for carers of people with dementia. The outcome measures used for carers were extracted from relevant studies and listed in a frequency table. Outcome measures appearing in four or more studies were discussed. The potential use of the ICECAP-O as a quality of life measure for carers of people with dementia is introduced in this chapter. An edited version of Chapter 3 has been published in *Health and Quality of Life Outcomes* (Jones, Edwards & Hounscome, 2012b).

Chapter 4 presents the results of the qualitative analysis exploring the suitability of the ICECAP-O, Adult Social Care Outcomes Toolkit (ASCOT) and Carer Experience Scale (CES) for carers of people with dementia. Having recognised that the EQ-5D is not an ideal instrument to measure health-related quality of life in psychosocial interventions for carers of people with dementia, there is a need to consider other instruments for use in research. Carers were questioned about their experience of caring, and the perceived impact on their quality of life. Themes that arose from the interviews were discussed in the context of choosing an appropriate health-related quality of life instrument for future health economics research. In doing so, Chapter 4 encourages a mixed methods approach to health economic evaluations by utilising qualitative methods to inform quantitative research design.

Chapter 5 presents results of an exploratory construct validity analysis for the use of the ICECAP-O with carers. As mentioned previously, the ICECAP-O was developed to measure capability in members of the general population aged 65+. The data for the validity analysis was from two sources: Challenge FamCare baseline data and additional survey data collected through SurveyMonkey. Feasibility was assessed using item completion rates. Floor and ceiling effects were assessed by examining the percentage of respondents choosing the highest and lowest level for each domain of the ICECAP-O. Cronbach's alpha was used to assess internal consistency. Construct validity was assessed using Spearman's rho to examine the correlation between the ICECAP-O and other outcome measures used. Chi-square tests were used to explore the relationship between the domains of the ICECAP-O and the domains of the EQ-5D.

Chapter 6 presents a cost-effectiveness analysis of joint-remembrance therapy for people with dementia and their carers. The data source for this chapter was a subset of 41 REMCARE participants who completed the ICECAP-O. The primary outcome measure for the analysis was the ICECAP-O. Secondary outcome measures for the analysis were the EQ-5D, Quality of Care-giving relationship (QCGR), Hospital Anxiety and Depression Scale (HADS), Relatives' Stress Scale (RSS) and the General Health Questionnaire (GHQ-28). Mean outcome measure values for the intervention and control group at baseline, 3 months and 10 months were presented, along with mean costs at 10 months. Cost and effect results were compared to results from the economic analysis sample of the main REMCARE trial. Loss of power due to small numbers of participants is acknowledged.

Chapter 7 concludes the thesis. The chapter examines the principle results from earlier chapters and synthesizes findings. It offers an interpretation of the findings and discusses the implications for a number of areas: health economics as a discipline that supports evidence-based policy making by Government; future commissioning and reviewing of proposed research in the health economics of dementia care by research funding bodies; and contribution of evidence of effectiveness and cost-effectiveness on carers in local dementia care service commissioning. Limitations of the thesis and areas for future research are also discussed.

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Chapter 2: A systematic review of the cost-effectiveness of interventions for supporting informal carers of people with dementia residing in the community

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An edited version of this chapter is published as:

Jones, C., Edwards, R.T. & Hounsome, B. (2012). A systematic review of the cost-effectiveness of interventions for supporting informal caregivers of people with dementia residing in the community. *International Psychogeriatrics*, 24(1), 6-18.

## **Abstract**

### *Background*

Dementia places a huge demand on health care services; however a large proportion of the cost is borne by informal carers. With the number of people affected by dementia set to increase in the future, there is a need for research to consider the effects of interventions on informal carers as well as persons with dementia. This chapter seeks to systematically review the existing evidence on the cost-effectiveness of interventions to support informal carers of people with dementia residing in the community.

### *Methods*

A range of electronic databases was searched. Studies were included if both costs and outcome measures for informal carers of people with dementia residing in the community were reported for an intervention. Both pharmacological and non-pharmacological interventions were included. Quality of study was assessed using the Drummond ten item checklist for economic evaluations and results were presented through narrative synthesis.

### *Results*

Twelve studies were included in the review, of these only four reported a significant difference in the outcome measure for carers.

### *Conclusions*

At present few published studies report costs in enough detail to provide evidence of the effectiveness and cost-effectiveness of interventions for supporting carers. Future trials need to collect carer data alongside patient data to increase the evidence base for intervention effectiveness. Further research is required to establish the effectiveness and cost-effectiveness of both pharmacological and non-pharmacological approaches.

## **Introduction**

Worldwide, the number of people with dementia is estimated to be over 35 million, a figure expected to rise due to an ageing population (Wimo and Prince, 2010). This increase will place a significant burden on health care services. In the United Kingdom, two-thirds of people with dementia live in private residences and are supported by informal care provided by friends and family. This informal care contributes £12.4bn (55%) of the estimated £23bn annual cost of dementia to the economy (Luengo-Fernandez et al., 2010). Estimates of the global cost of informal care are as high as US\$251bn (Wimo and Prince, 2010). As the informal care sector is such a substantial resource, it is essential that when reviewing dementia care, whether as interventions delivered by the NHS or social services, or as fiscal measures, one also considers the benefits and adverse effects on the carer. From an NHS perspective, this agrees with the National Institute for Health and Care Excellence (NICE) 'reference case' which states '*an additional QALY (Quality Adjusted Life Year) should receive the same weight regardless of any other characteristics of the people receiving the health benefit*'. The reference case explicitly states '*the perspective on outcomes should be all direct health effects, whether for patients or, when relevant, other people (principally carers)*.' (NICE, 2013). NICE has also produced specific care guidelines for carers of people with dementia (NICE, 2007).

There are many papers detailing pharmacological and non-pharmacological interventions to support people with dementia, but fewer economic evaluations of interventions to support their informal carers. Often outcomes for informal carers are secondary measures of studies primarily targeted at the persons with dementia. This paper reviews evidence on health-related outcomes for carers from interventions targeted at people with dementia and their carers, as well as interventions solely targeted at carers.

## **Method**

Studies were considered if an informal carer was looking after a person with dementia in a community setting. Carers could be any age, gender, nationality or ethnicity and both pharmacological and non-pharmacological interventions were included. Studies had to include cost data and a health-related outcome measure for the carer.

Electronic database searches were conducted on Cinahl, Biosis, Medline, PsycINFO, Science Direct, Cochrane Library, NHS Economic Evaluation Database, BMJ, The Department of Health and Rowntree Foundation. Titles, keywords, and text were searched for the terms (“Dementia” or “Alzheimer’s Disease”) and (“Caregiver\*” or “care\*”) and “Costs and Cost Analysis”. The search strategy for each database is presented in Appendix 6, page 187. The last search was conducted on January 18, 2011.

Study eligibility was based on initial screening of title and abstract by the lead author (CJ). Titles and abstracts were not screened by a second reviewer. Articles passing the initial screening were retrieved for further inspection. The type of study, country of study, study design, number of participants recruited and completing the study, study length, carer outcomes measured, cost results and outcome results were extracted.

Study quality was assessed using the Drummond ten item checklist for robust economic evaluations (Drummond et al., 2005). Differences in quality were highlighted to avoid studies with weaker methodology being given the same weight as studies of higher quality.

It was anticipated that studies would be diverse in both setting/type of intervention and participants; therefore meta-analysis would be unsuitable and a narrative approach to data synthesis was planned. The principal summary measure for the effectiveness of an intervention was the difference in means of the carer outcome measures between intervention and control groups at the end of the intervention.

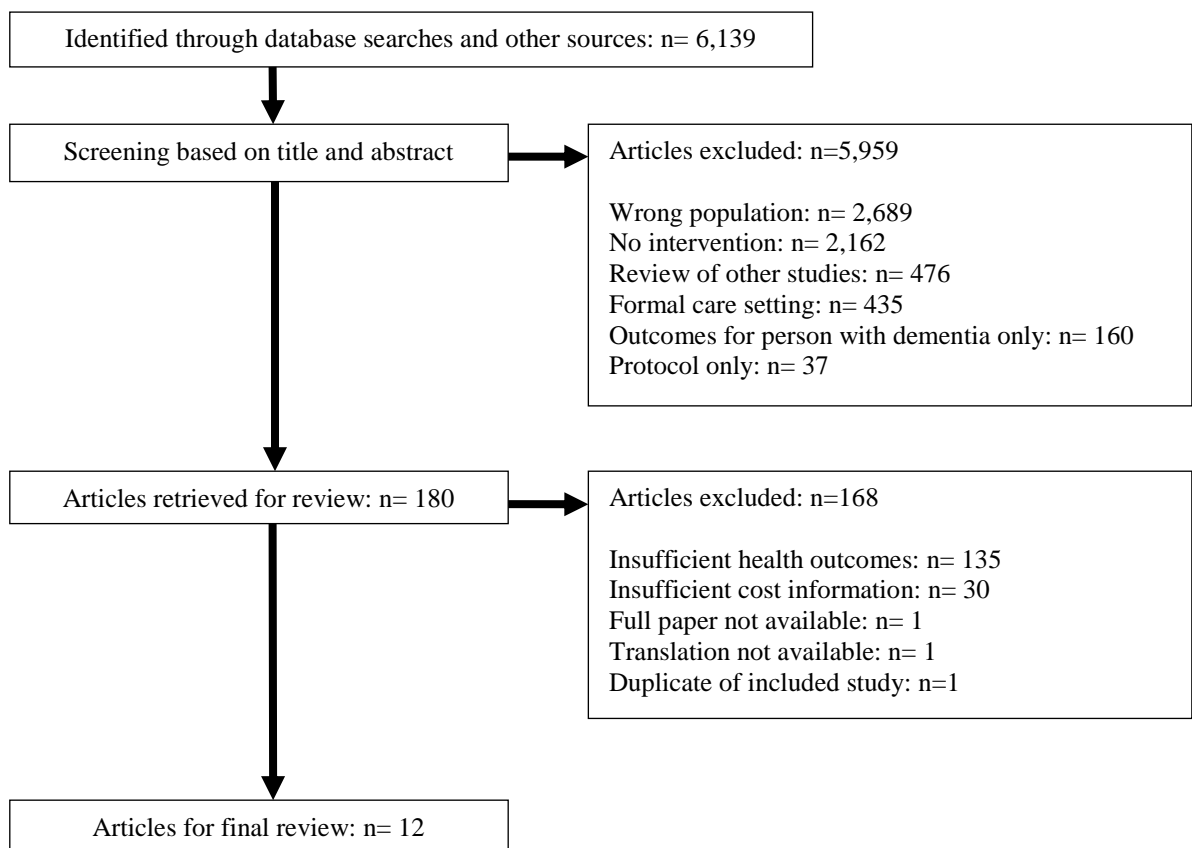
## **Results**

6,139 studies were identified after duplicates were removed from search results (Figure 2.1). 5,959 were excluded during the initial screening of title and abstract. The main reasons for exclusion were reporting survey rather than intervention data and the population being too broad, for example, interventions with carers as participants within a general population, but data not being extractable. 180 articles were retrieved for further inspection. 135 were excluded because health-related outcomes were not included, 30 were excluded due to insufficient costing information, 1 article was not available through library loans or contact with the author, and a translation in English was not available for 1 article. Two papers contained results of the same study, the BECCA befriending scheme



(Charlesworth et al., 2008; Wilson et al., 2009). To avoid repeated presentation of the same results, the cost-utility paper (Wilson et al., 2009) was excluded from the review and will not be discussed further; however the results will be covered in the discussion on the main paper, (Charlesworth et al., 2008). The twelve articles retrieved for full review are presented in Table 2.1.

**Figure 2.1:** Flow of articles retrieved through electronic searches



**Table 2.1:** Characteristics of studies included in the review

STUDY	INTERVENTION TYPE	COUNTRY, DESIGN AND FOLLOW UP	NUMBER OF PARTICIPANTS	CARER MEASURES	PERSPECTIVE AND COST RESULTS	OUTCOME RESULTS (I= Intervention group, C= Control group)
Courtney et al., 2004	Pharmacological	UK.  Double-blind RCT.  Indefinite- patients followed until withdrawal from study, full time-care or death.	566 randomised. 293 completed initial 60 week phase (52%).	Baseline, 12, 24, 36, 48 and 60 weeks.  General Health Questionnaire-30 (GHQ-30).	Health care provider.  Intervention group mean cost per patient/yr: £2842.  Control group mean cost per patient/yr: £2344.  Not a significant difference.	GHQ-30 between group difference of 0.3 points at 60 weeks, I<C. Not a significant difference.
Feldman et al., 2003, 2004	Pharmacological	Canada, Australia, France.  Double-blind RCT.  24 weeks.	290 randomised. 246 completed (85%).	Baseline, 4, 12 and 24 weeks.  Modified Caregiver Stress Scale (CSS).	Societal.  Intervention group mean cost/ dyad over 24 weeks: Can\$9904.  Control group mean cost / dyad over 24 weeks Can\$10,236.  Not a significant difference.	CSS between group difference of 1.82 points at 24 weeks, I<C. Not a significant difference.

Getsios et al., 2010	Pharmacological	UK.  Discrete-event simulation.  10 years.	Simulated individuals created from dataset of 826 participants from 3 donepezil trials.	QALY gain calculated from SF-36.	Societal.  Intervention group costs per dyad at 10 years: £166,178.  Control group costs per dyad at 10 years: £170,947.	QALY gain of 2.920 per carer in the intervention group and 2.909 in the control group.
Neumann et al., 1999a	Pharmacological	USA.  Markov model.  24 months.	Donepezil trial recruitment and completion rates not given.	QALY gain calculated from HUI2.	Societal.  Intervention group costs at 18 months: US\$72,487.  Control group costs at 18 months: US\$72,227.	Carer QALY gain in the intervention group <0.002, <0.000 in the control group.
Charlesworth et al., 2008	Psychosocial	UK.  Single-blind RCT.  24 months.	236 randomised. 190 completed (81%).	Baseline, 6, 15 and 24 months.  EQ-5D, Hospital Anxiety and Depression Scale (HADS), Positive and Negative Affect Schedule (PANAS), Carers Assessment of Difficulties Index (CADI), Brief Coping Orientation for Problem Experience (COPE).	Societal.  Intervention group mean cost per dyad over 15 months: £122,665.  Control group mean cost per dyad over 15 months £120,852.  Not a significant difference.	HADS between group difference of 0.468 points at 15 months, I<C. PANAS between group difference of 0.079 points, at 15 months, I>C. EQ-5D VAS between group difference of 2.33 at 15 months, I>C. QALY gain of 0.946 in intervention group and 0.929 in control group. Not significant differences.

Graff et al., 2006, 2008	Psychosocial	Netherlands.  Single-blind RCT.  12 weeks.	135 randomised. 105 completed (78%).	Baseline, 6 and 12 weeks.  Sense of Competence Questionnaire (SCQ), CES-D.	Societal.  Intervention group mean cost per patient at 12 weeks: €12,563.  Control group mean cost per patient at 12 weeks €14,311.	SCQ between group difference of 9.6 points at 12 weeks, I>C. A significant difference. No follow up results for CES-D.
Martikainen et al., 2004	Psychosocial	Finland.  Monte Carlo Markov model.  5 years.	206 recruited for CBFi programme, completion rate not given.	QALY gain calculated from HUI2.	Societal.  Intervention group mean cost per dyad at 5 years: €43,933.  Control group mean cost per dyad at 5 years: €46,925.  Not a significant difference.	QALY gain of 3.13 per carer in the intervention group and 3.14 in the control group. Not a significant difference.
Roberts et al., 1999	Psychosocial	Canada.  Single-blind RCT.  12 months.	83 randomised. 58 completed (70%).	Baseline, 6 and 12 months.  Psychosocial Adjustment to Illness Scale (PAIS), Caregiver Burden Interview (CBI), Duke Social Support (DSS), Indices of Coping (IOC).	Societal.  Intervention group mean cost per carer at 1 year: Can\$4,545.  Control group mean cost per carer at 1 year: Can\$2,005.	Change in PAIS psychological distress at 1 year: I +0.28, C -0.83. Not a significant difference. No significant difference in CBI or DSS. Change in logical analysis subsection of IOC at 1 year: I +0.21, C -1.03. A significant difference.

Challis et al., 2002	Service delivery	UK. Quasi-experimental double cohort. 12 months.	86 matched pairs. 80 alive at 12 months (93%).	Baseline, 6 and 12 months. Global indicator of strain, burden of care, Malaise scale for stress.	Societal. Intervention group total cost at 12 months: £23,402. Control group total cost at 12 months: £19,053. Not a significant difference.	Carer perceived burden - 1.46 points at 12 months in the intervention group, -0.39 points in the control group. A significant difference. No significant differences in strain or stress.
Drummond et al., 1991	Service delivery	Canada. Single-blind RCT. 6 months.	60 enrolled. 42 completed (70%).	Baseline, 3 and 6 months. CES-D, State-Trait Anxiety Inventory (STAI), Caregiver Quality of Life Instrument (CQLI).	Health care provider Intervention group mean cost at 6 months: Can\$3,562. Control group mean cost at 6 months: Can\$2,897. Not a significant difference.	Change in CES-D at 6 months: I -0.05, C +2.8. Change in STAI at 6 months: I -0.07, C +2.25. Change in CQLI at 6 months: I +0.09, C -0.03. Not significant differences.
Engelhardt et al., 2008	Service delivery	USA. Quasi-experimental double cohort. 20 months.	36 enrolled. 32 active participants (89%).	Baseline and 20 months. Client Experience Questionnaire, Nursing Outcome Classification (NOC).	Health care provider. Average annual intervention group total inpatient costs plus intervention costs: US\$12,988. Average annual control group total inpatient costs: US\$30,650. A significant difference.	Data only available for participants in the intervention group. Mean NOC scores increased 2.04 points between baseline and the end of the study. A significant difference.

Gaugler et al., 2003	Service delivery	USA. Double cohort. 12 months.	548 recruited. 233 completed (43%).	Baseline, 3 and 12 months. Role Overload Scale (ROS), CES-D.	Societal. Intervention group daily costs at 12 months: \$47.10. Control group daily costs at 12 months: \$41.15. A significant difference.	ROS between group difference of 1.32 points at 1 year, I<C. CES-D between group difference of 2.97 points at 1 year, I<C. Significance not clear.
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### *Pharmacological interventions*

Courtney and colleagues (2004) described a double-blind randomised controlled trial (RCT) of donepezil compared to placebo for people with dementia. Donepezil is an acetylcholinesterase inhibitor which impedes the breakdown of acetylcholine neurotransmitters. It is believed to have a beneficial effect on memory and the ability to carry out daily activities, but its effect on behavioural problems associated with dementia is currently ambiguous. Five-hundred and sixty-six people with dementia were randomised to either 5mg/day of donepezil or a placebo. At twelve weeks participants were re-randomised to receive donepezil 5mg/day, 10mg/day or placebo. Treatment continued until participants withdrew from the study, were institutionalised or died. Quality of life of the carer was measured with the General Health Questionnaire-30 (GHQ-30) at baseline, 12, 24, 36, 48 and 60 weeks. The GHQ-30 is a self completed questionnaire used to assess anxiety, social functioning and depression. A higher score indicates increased severity of symptoms. At 60 weeks, GHQ-30 scores of carers in the intervention group were 0.3 points lower than the control group; however this was not a significant difference (95% CI -0.3 to 0.9,  $p=0.3$ ). Service use was measured with the dementia-specific Caregiver Activities Scale and UK national average unit costs for the price year 2000. Annual cost per patient was £2,842 in the donepezil group and £2,344 in the placebo group, a non significant difference ( $p=0.16$ ). The authors stated that a societal perspective was taken, however the costs of medication, institutionalisation and informal care were excluded, hence a health care provider perspective is more accurate.

Results of a 24 week trial of 5-10mg of donepezil daily compared to a placebo were presented by Feldman and colleagues (2004). The study was multi-centre with 290 dyads randomised across sites in Canada, Australia and France. Informal caring time was costed at the Canadian minimum wage. Health service use was recorded by the Canadian Utilization of Services Tracking questionnaire with analysis from a societal perspective, only including service use that was considered to be Alzheimer's disease related. Service costs were calculated in 1998 Canadian dollars using Canadian unit costs for all countries. The mean cost per dyad at 24 weeks was Can\$9,904 for the intervention group and Can\$10,236 for the control group, a non-significant difference. Effectiveness results were presented in a separate paper (Feldman et al., 2003). The main outcome measure for carers was a modified version of the Caregiver Stress Scale (CSS) recorded at baseline, 4, 12, and 24 weeks. Higher scores on the CSS reflect increased stress levels. At 24 weeks there was

no significant difference in overall CSS scores between groups but a significant difference was observed on cognitive status ( $p < 0.001$ ). Carers in the control group perceived a greater cognitive decline in the persons with dementia than their counterparts in the intervention group.

The cost-effectiveness of donepezil for people with dementia was modelled by Getsios and colleagues (2010) using discrete-event simulation. Individuals were simulated using data from 826 participants across three efficacy trials of donepezil. Data on long term behavioural and functional effects associated with dementia were taken from seven donepezil trials. Costs were taken from UK national data sources and included daily donepezil costs, twice yearly physician visits, and direct and indirect care costs; hence a societal perspective was used. Carer health utility data were modelled over ten years using SF-36 scores from the donepezil trials. To make the simulated individuals typical of people in the United Kingdom with Alzheimer's disease, sex and age weighting from a national dementia report were applied. For people with dementia starting treatment in the mild-moderate stage the costs at ten years were £166,178, while costs for those in the control group were £170,947. QALY gains for carers in the intervention group were 2.920, while for the control group QALY gains were 2.909, a net difference of 0.012 after ten years.

Neumann and colleagues (1999a) assessed the cost-effectiveness of donepezil compared to usual care using a Markov model. Transition probabilities between stages of dementia and participant settings were calculated from a longitudinal study of 1,145 people with dementia in the United States. Effectiveness of donepezil was taken from a 24 week RCT investigating 5mg and 10mg daily doses on people with mild or moderate Alzheimer's disease. A societal perspective was used for analysis. Costs of care were taken from a 1993 study of 180 people with Alzheimer's disease and their carers living in California. Quality of life (QoL) weights were obtained from the HUI2 (Health Utility Index) data of a companion study of 528 carers of people with Alzheimer's disease. QALY gains for carers were not presented, however sensitivity analysis showed an incremental cost-effectiveness ratio (ICER) of US\$8,700 and US\$72,000 when the weights for carers and people with dementia were combined. It was then possible to calculate that the QALY differences between intervention and control group was less than 0.002 for carers of people with mild



Alzheimer's disease and less than 0.001 for carers of people with moderate Alzheimer's disease.

### *Psychosocial interventions*

In the Health Technology Assessment conducted by Charlesworth and colleagues (2008), 236 carers were randomised to receive either contact with a Befriender Facilitator (BF) or usual care. The hypothesis was that if a person feels more able to cope with their caring role, their service use might become lower. The BF would arrange a match between carer and befriender volunteer with the expectation that there would be weekly home visits by the befriender for at least six months. Take up was 48% and the retention rate after 24 months was 81% with similar dropout rates between groups. The primary reason for withdrawal was carer health. Data were collected at baseline, 6, 15 and 24 months. Carer measures included the EQ-5D, Hospital Anxiety and Depression Scale (HADS), Positive and Negative Affect schedule (PANAS), Carers Assessment of Difficulties Index (CADI) and Brief Coping Orientation for Problem Experience (COPE). Resource use data was collected through an interview covering areas of the Client Service Receipt Inventory (CSRI), the Caregiver Time Questionnaire, the Caregiver Activity Schedule and the Resource Utilization in Dementia Questionnaire. Unit costs were derived from national estimates. Costs for carer time were calculated using the gross average wage. Base-case analysis was on costs and effects at 15 months. Mean QALY gains per carer were calculated using EQ-5D data and were 0.946 for the intervention group and 0.929 for the control group at 15 months, a non-significant difference ( $p=0.315$ ). Differences in HADS and PANAS scores were also non-significant ( $p=0.342$  and  $p=0.94$  respectively). Mean costs per carer at 15 months were £122,665 for the intervention group and £120,852 for the control group. The highest proportion of cost was informal carer time at approximately 85% of total costs. The ICER was calculated as £105,954. Sensitivity analysis implied a 42.2% probability of the ICER being below the NICE threshold of £30,000. A societal perspective was used for base-case analysis, when the perspective was shifted the mean ICER was £117,039 for the combined health service and public sector, £77,236 for households and £521 for the voluntary sector.

The cost-effectiveness of an in-home/telephone tailored occupational therapy programme compared to usual care was discussed by Graff and colleagues (2008). The hypothesis was that if carers felt better equipped in their caring role it might lead to a delay in

institutionalisation of people with dementia, therefore reducing societal costs. Criteria for the carer were not as strict as for other studies; the carer only needed to provide care at least once a week and not have a severe illness to be eligible. The criteria for people with dementia was being in the mild to moderate stage of dementia, free from severe behavioural and psychological symptoms; not diagnosed with severe depression or co-morbid illness and at least three months into a course of a cholinesterase inhibitor or memantine. One-hundred and thirty-five dyads were randomised. The intervention spanned five weeks and consisted of four hourly sessions of problem defining and goal setting for the dyads, followed by six sessions of practical advice on problem solving, coping strategies and effective supervision. Outcome measures were collected at baseline, 6 and 12 weeks. The Sense of Competence Questionnaire (SCQ) was the primary outcome measure, a higher score indicating a higher feeling of competence. Clinical effectiveness results were presented in a separate paper (Graff et al., 2006). The Center for Epidemiologic Studies depression scale (CES-D) was applied at baseline but no follow-up data were available. At twelve weeks, adjusted SQC scores were 9.6 points higher for the intervention group compared to the control group, a significant difference (95% CI 4.7 to 14.5,  $p < 0.0001$ ). Costs were calculated using outpatient tariffs to cover staff delivery of the occupational therapy sessions and staff hourly wages for additional time such as administering interviews and analysing the data. Health service unit costs were taken from Dutch guidelines on economic health care from 2004. Informal caring hours were valued using the unit cost of a middle-aged cleaning person applied to the hours of absence from employment attributed to caring or illness. A societal perspective was used; the mean cost per patient at twelve weeks was €1,748 lower (95% CI -€4,244 to €748) in the intervention group than the control group (€12,563 vs. €14,311).

The long term cost-effectiveness of a cognitive-behavioural family intervention (CBFI), as compared to usual care in Finland, was modelled by Martikainen and colleagues (2004). The CBFI was aimed at both the persons with dementia and their carer. People with dementia received physical and recreational training, while carers received educational support and counselling. A Monte Carlo Markov model was derived from Neumann et al. (1999a), discussed previously. Transition probabilities were from Neumann et al. (1999b); the transition from moderate to mild stage was excluded as it was deemed implausible. The effect of the CBFI programme on delay to nursing home admission was taken from a study of 206 participants receiving the intervention over 3.5 years and followed up for 8

years. QoL measures were not included in the CBFI effectiveness study so were derived from a separate study of the QoL weights attached to each stage of dementia under the assumption that carers' QoL was directly related to the progression of dementia in the person that they cared for. Usual care practice was not standardised across Finland so was based on the main services provided by two regional health centres. A societal perspective was adopted; service and medication costs were taken from national estimates, intervention costs were local. Costs were discounted by 5% annually. The Bayesian approach was taken to calculate distributions for the parameters used. A second-order Monte Carlo simulation was used to calculate the distribution of mean costs and effects from the resulting model. The mean cost per dyad in the intervention group was €43,933 and €46,925 in the control group, a non-significant difference. As the cost of the intervention was lower than standard care, the CBFI programme is potentially cost saving. A cost-effectiveness acceptability curve was constructed and showed that there was a probability of 0.9 that the CBFI programme was cost-effective for all values of willingness to pay up to €100,000 per QALY gained. Carers in the intervention group gained 3.13 QALYs over five years compared to the 3.14 QALYs of carers in the control group; however this was not a significant difference.

Roberts and colleagues (1999) evaluated the effect of up to ten one-to-one problem-solving therapy sessions for carers of persons with dementia compared to usual care. The counselling sessions were nurse-led and conducted in-home or by telephone. Eighty-three carers were randomised, with 38 receiving the counselling and 45 assigned to the control group. The primary outcome was the Psychosocial Adjustment to Illness Scale (PAIS), with carers also completing the Caregiver Burden Interview (CBI), Duke Social Support (DSS) and Indices of Coping (IOC) measures at baseline, 6 months and 1 year. A significant difference between groups was found on the logical analysis sub section of the IOC (intervention group change +0.21, control group change -1.03,  $p=0.04$ ). No significant differences were recorded by the other measures. Comprehensive costs were collected and classified by carer direct and indirect expenses, cash transfers and health and social service use for the persons with dementia. The mean direct and indirect cost for intervention carers at 1 year was Can\$4,545, while the mean cost for the control carers was Can\$2,005.

### *Service delivery interventions*

Studies were classed as service delivery if the intervention involved a change in the management or delivery of existing services, either for the persons with dementia or their carer.

Challis and colleagues (2002) matched dyads on functioning, behavioural symptoms and carer stress level. Of the 86 dyads, 43 received case management. A case manager, based in a community mental health team for the elderly, regularly updated care plans for dyads in the case management arm. Dyads were selected for case management on the basis of identified unmet service needs and a significant risk of institutionalisation being present. Carer outcomes were assessed at uptake, 6, and 12 months and included an indicator of strain, the Malaise scale for stress and a measure of the burden of care. At 12 months the only significant difference between groups was in the lower perceived burden felt by carers in the intervention group (95% CI -3.37 to -0.33,  $p < 0.05$ ). Due to the design of the study there were no 'drop-outs' as such, but six individuals had died at the twelve month point. A societal perspective was used for analysis. Mean annual costs of care services per patient (including long-term, hospital and social services) were £23,402 for the intervention group and £19,053 for the control group, a non-significant difference.

Carers of persons with moderate to severe dementia were block randomised to a Caregiver Support Program (CSP) or standard care in Drummond et al. (1991). The intervention consisted of a 4 hour block of weekly in-home respite care, along with additional respite on request and regular home visits by nurses to educate about caring and neglected health problems. Carers were also told about a monthly support group in the area. Standard care for the control group was focused on the persons with dementia rather than the carer. Sixty carers were randomised and 42 (70%) completed the study. The health outcomes collected were CES-D, State-Trait Anxiety Inventory (STAI) and the Caregiver Quality of Life Instrument (CQLI) (Mohide, Torrance, Streiner, Pringle & Gilbert, 1988). At 6 months the carers in the intervention group had a small improvement in CQLI while the control group had a decrease in CQLI, the difference between groups was not significant. Cost data were reported in Canadian dollars for the year 1988, and included health service use such as nursing, respite and physician visits. The mean cost per carer in the intervention group was Can\$3,562 over the 6 month period while the mean cost in the

control group was Can\$2,897, a non significant difference. The authors calculated an incremental cost per QALY of Can\$20,036.

A pilot programme investigating the impact of a care coordination and support strategic partnership (CCSP) between a home health care agency and a telephone support service provided by nurses was presented by Engelhardt and colleagues (2008). Thirty-six people with Alzheimer's disease and their carers were recruited, with 32 actively participating. One-hundred and thirteen people with Alzheimer's disease who did not receive the CCSP programme were identified through medical records, but no further details were given of how the controls were matched to the participants of the CCSP programme. Dyads in the CCSP programme were followed for 20 months. Clinical effectiveness was determined using the Nursing Outcome Classification (NOC) system, where areas such as knowledge are rated on a scale of one to five with five being the best state possible. At baseline, the mean NOC score for carers in the intervention group was 2.14. This increased to 4.18 at 20 months, a significant difference ( $p < 0.001$ ). NOC scores were not available for the matched cohort. Programme delivery costs were calculated by multiplying the length of telephone support time by the average clinician wage plus overhead costs. Home visits were costed at US\$95.61 per visit; no explanation was given as to where this figure came from. Informal carer time was not recorded; hence a health care provider perspective was adopted. Mean annual cost of inpatient hospital stays plus intervention per participant was US\$364 in the intervention group and US\$271 in the control group.

Gaugler and colleagues (2003) discussed the effect of adult day services compared to no adult day services. Carers receiving subsidized adult day care services in New Jersey were compared to a control group who did not use adult day care services in Ohio, an area chosen because of its similar socio-demographic mix. Carers completed the Role Overload Scale and CES-D at baseline, three months and one year. Five-hundred and forty-eight carers were selected for the study, but because carers selected for analysis in the control group could not have used adult day care services, the number eligible at one year had dropped to 233. The number of hours that carers reported receiving formal services and spent on informal care tasks was multiplied by the US Department of Labor figure for average household payments for home health care services. A societal perspective was used for analysis. Loss of employment hours were costed using hourly wage estimates from the Bureau of Labor Statistics. At 12 months the average daily costs for the

intervention and control group were US\$47.10 and US\$41.15 respectively, a significant difference ( $p < 0.05$ ). Role overload was 19.96 in the intervention group and 21.28 in the control group; a lower score indicated a lower level of overload. CES-D scores were 12.77 in the intervention group and 15.74 in the control group, a lower score indicating a lower level of depression. Significance of the clinical results was not presented clearly in the paper.

### **Study Quality**

The Drummond et al. (2005) ten item checklist was applied to included studies and results are summarised in Table 2.2. The majority of papers clearly stated a research question or objective. The alternatives under evaluation were discussed, albeit briefly in papers which modelled effectiveness data from other trials, or where companion papers gave fuller descriptions of the intervention (Feldman et al., 2003, 2004; Gaugler et al., 2003; Getsios et al., 2010; Neumann et al., 1999a). Rationale behind study design was clearly stated and effectiveness was established for all studies apart from Engelhardt et al. (2008), where effectiveness data were only collected for the intervention group due to the control being a matched historical cohort. This meant that it was not possible to determine how effective the intervention was compared to usual care.

The range of costs collected in each study varied, with studies including health service use, social service use, informal care time or a mixture of all three. Unit cost sources were stated on the whole, but several studies did not report the price year (Courtney et al., 2004; Challis et al., 2002; Engelhardt et al., 2008; Graff et al., 2006; Roberts et al., 1999). Of the studies where follow-up extended beyond twelve months, discounting of costs and benefits was conducted where appropriate, apart from in Courtney et al. (2004), where annual costs were calculated by multiplying each 12 week phase up to a year rather than using costs collected over a longer time period. Half of the studies performed an incremental analysis of costs and consequences (Charlesworth et al., 2008; Drummond et al., 1991; Gaugler et al., 2003; Getsios et al., 2010; Martikainen et al., 2004; Neumann et al., 1999a). No form of sensitivity analysis was mentioned in five studies (Challis et al., 2002; Drummond et al., 1991; Engelhardt et al., 2008; Gaugler et al., 2003; Roberts et al., 1999).

**Table 2.2:** Quality assessment of included studies

	1	2	3	4	5	6	7	8	9	10
Courtney et al., 2004	Y: Research questions stated p2107.	Y: Description of intervention and comparator p2107.	Y: Effectiveness results p2109-2113.	Y: Health and social service cost, and informal carer time collected. Informal care time not included in the analysis p2107-2108.	Y: Cost breakdown p2112.	Y: National sources p2108. Costs reported in UK£, price year not stated.	N: Annual costs calculated by multiplying each 12 week period up p2112.	N	Y: Sensitivity analysis performed but not presented. 95% CIs presented.	Y: Results compared to other studies.
Challis et al., 2002	Y: Research objective stated p315.	Y: Description of intervention p316. Comparator group not described in detail.	Y: Theory presented p315-316. Effectiveness results p318-320.	Y: Health and social service costs and carer time costed p322.	N: Unit of costing unclear.	N: Sources not stated. Costs reported in UK£, price year not stated.	N/A: Follow up period of 1 year.	N	N: No sensitivity analysis reported.	Y: Results compared to other studies. Implications for future research discussed.

Charlesworth et al., 2008	Y: Research objectives stated p3.	Y: Description of intervention and comparator p5, 13-17.	Y: Theory presented p2-3. Effectiveness results p24-29.	Y: Health and social service costs, voluntary sector costs and informal care costs included. P10.	Y: Various resource use schedules used. Cost breakdown in appendices 3-4.	Y: Sources listed p 11, 18. Costs reported in UK£, price year 2005.	Y: Costs and benefits accrued between 15-24 months were discounted at 3.5% p 11.	Y: ICER calculated from various perspectives p32-36.	Y: ICERs and 95% CIs presented for different payer perspectives, along with a cost-effectiveness acceptability curve (CEAC).	Y: Results compared to other studies. Recommendations for future work discussed.
Drummond et al., 1991	Y: Research objectives stated p210.	Y: Description of intervention and comparator p211-212.	Y: Theory presented p210. Effectiveness results p213-216.	Y: Health and social service costs included p212. Informal care time not costed.	Y: Cost breakdown p215.	Y: Sources taken from health agency records or carer. Costs in Canadian\$, price year 1988 p215.	N/A: Follow up period of 6 months.	Y: ICER p216.	N: No sensitivity analysis reported.	N: Results not compared to other studies. Recommendations for future work brief.
Engelhardt et al., 2008	Y: Research objective stated p166.	N: Description of intervention p167. Comparator group were historical controls.	N: Effectiveness only presented for the intervention group p168-170.	N: Health service costs only p170-171. No informal costs collected.	N: Costs calculated by multiplying average contacts by the number of participants, rather than collecting actual contact numbers p170.	N: Health unit cost sources not given.	Y: Costs prorated for annual use but discount rate not stated.	N	N: No sensitivity analysis reported.	Y: Application and limitations of the pilot program discussed.



Feldman et al., 2003, 2004	Y: Research objective stated p644.	N: Brief description of intervention given p645. Full details given in separate paper.	Y: Effectiveness results in 2003 paper p739-742.	N: Only health services, social services and residential resource use related to dementia were recorded p645.	Y: Cost breakdown p646.	Y: National sources. Costs reported in Canadian\$, price year 1998 p645.	N/A: Follow up period of 24 weeks.	N	Y: Sensitivity analysis conducted. Appendix E1.	Y: Results compared to other studies. Limitations discussed.
Gaugler et al., 2003	N: No clear objective stated.	N: Referred to separate paper for description of intervention p121.	Y: Theory presented p118-121. Effectiveness results p126-128.	Y: Health service costs, day care costs, informal carer time and employment costs collected p125.	Y: Carer report of number of hours of contact with various services p125.	Y: Carer self-report. Costs reported in US\$, price year 1993 p6.	N/A: Follow up period of 1 year.	Y: ICER p127.	N: No sensitivity analysis reported.	Y: Limitations and implications discussed.
Getsios et al., 2010	N: No clear objective stated.	N: Efficacy data taken from several studies, no clear statement of intervention and control conditions p415.	Y: Effectiveness results p420.	Y: Health service costs and informal carer time collected p417-418.	Y: Cost breakdown p417-418.	Y: National sources and previous studies. Costs reported in UK£, price year 2007 p418.	Y: Costs and benefits discounted by 3.5% annually p411.	Y: ICER p420.	Y: Sensitivity analysis conducted on key model parameters p422. CEAC presented for societal and health care provider perspective p423.	Y: Results compared to other studies. Limitations discussed.

Graff et al., 2006, 2008	Y: Research objectives stated p2.	Y: Description of intervention p2-3. Comparator of usual care not described in detail.	Y: Theory presented p1-2. Effectiveness results in 2006 paper p3-5.	N: Health and social service costs were for the persons with dementia. Carer informal time costed.	Y: Cost breakdown p4-7.	Y: National sources p4-7. Costs reported in €, price year not stated.	N/A: Follow up period of 3 months.	N	Y: 95% CI and CEAC presented.	Y: Results compared to other studies. Recommendations for future work discussed.
Martikainen et al., 2004	Y: Research question stated p137.	Y: Description of intervention and comparator p137.	Y: Effectiveness results p141.	Y: Health and social service costs included p140. Informal care time not costed.	Y: Parameters used in model listed p 139-140.	Y: Sources listed p 139-140. Costs reported in €, price year 2001.	Y: Costs and benefits discounted by 5% annually p 139.	Y: ICER p141.	Y: 95% CI and CEAC presented p141, p138.	Y: Uncertainty surrounding model parameters and applicability to other settings discussed. No comparison to other study results. Brief recommendations for future work.
Neumann et al., 1999a	Y: Research objective stated p1138.	N: Efficacy parameters derived from separate study, brief description of intervention given p1139.	Y: Effectiveness results p1141.	Y: Direct medical costs and informal carer time costed p1140-1141.	Y: Parameters used in model listed p 1140.	Y: Unit costs taken from separate study p1140. Costs reported in US\$, price year 1997.	Y: Costs and benefits discounted by 3% annually p 1138.	Y: ICER p1141.	Y: Sensitivity analysis conducted on key model parameters p1142.	Y: Strengths and limitations discussed.

Roberts et al., 1999	Y: Research questions stated p163.	Y: Description of intervention p164. Comparator of usual care not described in detail.	Y: Theory presented p163. Effectiveness results p166-167.	Y: Health service costs, indirect costs, cash transfers and other social costs included. P164.	Y: Health and Social Utilization Questionnaire used. Breakdown of costs p167-169.	Y: National medical insurance rates. P164. Costs in Canadian\$, price year not stated.	N/A: Follow up period of 1 year.	N	N: No sensitivity analysis reported.	N: Research questions not explicitly answered. No comparison made to the results of similar studies.
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Key (taken from Drummond et al., 2005)

1. Was a well-defined question posed in answerable form?
2. Was a comprehensive description of the competing alternatives given i.e. can you tell who did what to whom, where, and how often?
3. Was the effectiveness of the programme 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 e.g. hours of nursing time, number of physician visits, lost work-days, gained life years?
6. Were the cost 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 estimates of costs and consequences?
10. Did the presentation and discussion of study results include all issues of concern to users?

## **Discussion**

### *Effectiveness findings*

The initial database search retrieved articles on a variety of intervention types and no clear trend in research focus or health economic methodology was observed. Four pharmacological, four psychosocial, and four service delivery interventions were included. Of the trials included for full review, four recorded significant differences in outcome measures for carers as the result of an intervention; lower perceived burden (Challis et al., 2002), higher sense of competence (Graff et al., 2006), improved coping skills (Roberts et al., 1999) and increased nursing knowledge (Engelhardt et al., 2008).

Despite a lack of statistically significant results for carers in the pharmacological studies, there were significant effects for the people with dementia participating in the trials. Indeed, NICE guidelines recommend donepezil for the maintenance of function and cognitive status for people with moderate stage Alzheimer's disease (NICE, 2009). Feldman et al. (2003) found that participants randomised to donepezil declined only 0.74 points on the Disability Assessment for Dementia scale compared to an 8.98 point decline for the control group at twenty-four weeks. Courtney and colleagues (2004) found that people with dementia randomised to donepezil scored 0.8 points higher on the Mini-Mental State Examination, an instrument used to measure cognition, than the control group over a two year period. It is possible that carers experienced benefits not recorded by the outcome measures selected for the trials, therefore it would be rash to state that pharmacological interventions have no benefits for carers when there is a lack of evidence overall.

The psychosocial interventions had mixed results for carers. This is supported by a 2003 meta-analysis (Brodaty et al., 2003), where a weighted average effect size of 0.32 was found for the main outcome measures of thirty studies into psychosocial interventions for carers of people with dementia. An effect size of 0.2 is considered statistically significant but weak, 0.5 is moderate and 0.8 is strong (Cohen, 1988). NICE guidelines recommend that psychosocial interventions such as cognitive behavioural therapy be available to carers who are experiencing psychological distress (NICE, 2007). The wide geographical area in Charlesworth et al. (2008) might have biased the trial costs through increased travel costs, with the likelihood being that if the befriending scheme had been set up in an urban area the costs would be lower.

Although Engelhardt and colleagues (2008) found a significant increase in carers' knowledge between baseline and the end of the service delivery intervention, the methodological quality of the study was weak. With no results available for the control group, it is difficult to quantify how much of the change was attributable to the intervention and how much was coincidental. Evidence on the effectiveness of service delivery interventions is of potential interest to health and social care providers and policy makers. With mounting pressure on health and social care budgets, the reorganisation of existing service delivery might be a more affordable and efficient short-term option than offering additional psychosocial interventions with high set-up costs.

Initial database searches returned many modelling papers, but most dealt only with the effects on the persons with dementia and did not contain data for the carer. Models are useful for extrapolating costs and effects beyond the time period that clinical evidence is available for, as evidenced by Getsios et al. (2010), where costs and QALY gains were extrapolated over ten years. A long time period for follow-up is generally preferable so that the residual effects of an intervention can be measured and valued. But as Getsios et al. (2010) only recorded a small difference over a time horizon of ten years, it leads to questions about whether the instrument used to measure carer utility, in this case the SF-36, was appropriate. While models can aid policymakers in allocation decisions when clinical data is not currently available, the quality of evidence produced by a model is dependent on the quality of the data populating the model. This means that while models are undeniably useful, clinical trials are still required to produce a portfolio of evidence to use for modelling.

The heterogeneity of carers within the sub-group of dementia carer needs to be considered; the needs of a spousal carer might differ from those of a child carer and what is effective for one sub-type of carer might not translate to another. Evidence on the effectiveness of combined psychosocial and pharmacological interventions is also sparse with further work in the area required. As well as addressing the issue of collecting relevant cost data, methodological quality also needs to be improved through larger sample sizes. Future research would benefit from a longer follow-up period, something that is often restricted due to funding reasons.

#### *Cost findings*

Three studies found significant differences in costs between intervention and control groups (Gaugler et al., 2003; Engelhardt et al., 2008; Roberts et al., 1999). Gaugler and colleagues

(2003) found the average daily cost for the control group to be \$41.15, compared to \$47.10 for the intervention group. Roberts et al. (1999) found no significant difference between health and social care costs between groups when the costs of the relative with dementia were included, although there was a significant difference for carer costs only (intervention group costs Can\$4,545, control group costs Can\$2,005). Conversely, Engelhardt et al. (2008) found significantly higher costs in the control group, mainly resulting from higher inpatient stays (control group costs were \$30,650 compared to \$12,988 for the intervention group at 20 months). However, the methodological quality of this pilot study was weak. The remaining studies did not have significant differences in cost outcomes and the balance between intervention and control group costs varied. Four studies found non-significantly lower costs in the control group at the end of the trial (Feldman et al., 2004; Getsios et al., 2010; Graff et al., 2008; Martikainen et al., 2004). It is an interesting point to consider whether funding bodies would support an intervention if it was deemed cost-saving but there was no significant benefit to the participants involved. Similarly, would funding be forthcoming if an intervention only benefitted the carer, but had no effect on the persons with dementia?

#### *Implications for funding*

The need to support carers has been historically recognised. However, in the United Kingdom the release of the Carers Strategy in England in 1999 was the start of real policy planning for carers. The most recent update of this document was 2010 (Department of Health, 2010a) and the most recent Carers Strategy for Wales was released in 2013 (Welsh Government, 2010). Key messages from the Strategy include the need for carers to stay mentally and physically well, and to not suffer financial hardship due to their role. During the financial year 2010/11 the government pledged to make £1million available to condition-specific voluntary organisations that help support carers. On top of this, £400million is to be given over four years to the NHS to fund respite breaks for carers. The figures might look large on the surface, but this financial support is to be divided between carers of all types, and to date dementia funds have never reached the level allocated to other illnesses. The level of UK funding for dementia research is estimated to be approximately £50 million annually, considerably less than funding for cancer research (£590 million) or heart disease (£169 million). The proportion of funding for research does not reflect the cost of illness to social and health care services which is £10 billion annually for dementia, £4.5 billion for cancer and £2.3 billion for heart disease (Luengo-Fernandez et al., 2010). If research funding for dementia were to increase in line with its national burden, there would be a boost to research in this field. Informal care accounts for £12billion (55%) of the annual United Kingdom cost

of dementia (Luengo-Fernandez et al., 2010). Although this could be partially funded through increased taxes, such fiscal measures are rarely popular. Private funding of care is another option, but with an estimated annual cost per dementia patient of over £27,000 (Luengo-Fernandez et al., 2010) this would not be universally affordable and the government would still need to cover costs for patients who could not afford the bill.

While health economists provide evidence on the costs and benefits of competing treatments, it is ultimately policymakers who decide which interventions are funded. NICE has set a threshold for cost-effectiveness at £20,000-£30,000 per QALY gained for an intervention to be deemed an effective use of NHS resources (NICE, 2013). As discussed above, NICE prefers instruments such as EQ-5D to be used for QALY calculation purposes; hence cost-effectiveness decisions will be biased toward interventions that improve physical health. While few would argue that physical health is the only aspect of quality of life worthy of targeting, there is an argument that interventions solely aimed at improving general quality of life are not the main duty of the NHS and should be funded by other means. Treatments that cost more than the £30,000 per QALY threshold can still be funded if public reaction to the original decision is unfavourable. An example of a funding decision being overturned is that of the breast cancer drug Herceptin (Barrett et al., 2006). One of the key concepts in health economics is opportunity cost; the true cost of funding a treatment is the value of benefits foregone by not using the resources in their best alternative use. For each NICE guidance overturned, resources need to be diverted from elsewhere to fund the treatment.

Once an intervention has been deemed cost-effective and policymakers decide to implement it beyond the trial period, re-distribution of resources toward the intervention is required. This is more complicated in the case of carers of people with dementia as interventions might fall across the divide between health and social care. Consequently, there is the issue of which sector should bear the set-up and running costs. The need for greater integration between different sectors in the provision of support for carers is clear.

#### *Implications from an outcome measures perspective*

In research involving carers, it is typically mental health improvements that are targeted (although not exclusively); therefore using an instrument capable of detecting broader changes in quality of life than changes in physical functioning is important. The reason for targeting mental health is that caring has been found to have a greater effect on mental health than on physical health due to its unpredictable nature and psychological strain (Schulz &

Sherwood, 2008). Considering the nature of the interventions included in this review (ranging from psychosocial to service delivery), the use of outcome measures sensitive to changes in carer mental health would be expected. Nine of the twelve studies in this review used instruments focussing on mental health areas such as depression, strain and coping. These instruments ranged from generic measures such as the General Health Questionnaire, to population specific measures such as the modified Caregiver Stress Scale and Caregiver Burden Interview. The three remaining studies used generic outcome measures which cover both mental and physical aspects of quality of life (Health Utilities Index, SF-36). Only one study (Charlesworth et al., 2008) included an outcome measure covering both mental and physical health (EQ-5D) alongside outcome measures solely oriented towards mental health.

NICE guidance on technology appraisals state that a standardised and generic instrument such as the EQ-5D is preferred (NICE, 2013). While the EQ-5D is good at detecting changes in physical health, it is not particularly sensitive to mental health beyond one question on anxiety/depression, as discussed in Chapter 1, page 19. One solution to the issue of selecting outcome measures sensitive enough to detect quality of life changes in populations not expected to see an improvement in physical functioning as a result of an intervention is to include a generic preference based quality of life measure alongside group-specific measures, as was seen in Charlesworth et al. (2008). However, there is a need to consider the burden of asking carers to complete extra questionnaires. None of the studies included an outcome measure developed from the capabilities approach, but this is unsurprising as instruments such as the ICECAP-O (Grewal et al., 2006) are fairly recent additions to the quality of life measurement toolkit. The capability approach allows quality of life measurement to go beyond health output, which is appropriate when exploring effects on carers. Instruments rooted in the capabilities approach offer the potential to bridge the gap between instruments focusing on the physical functioning aspect of quality of life and those focusing on the mental health aspect.

#### *Limitations of the review*

Although there is a lack of evidence of the cost-effectiveness of interventions to support informal carers of people with dementia residing in the community, this is a reflection of the current quantity of studies in this area including a health economics component, rather than a limitation of the review. A number of trials for both the persons with dementia and their carer are underway; therefore a larger evidence base will be available in the future e.g. Joling et al., 2008.



The main limitation of this review is that titles and abstracts were not screened by a second reviewer. The review was conducted as part of a PhD with limited resources available. Systematic reviews conducted as part of fully funded trials should ideally be second screened to ensure no relevant articles are excluded.

The heterogeneous nature of the interventions, participants and settings precluded conducting a meta-analysis and results were instead presented as a narrative. Meta-analysis is a useful tool to determine whether treatment effects are similar in similar situations; as the evidence base for both the effectiveness and cost-effectiveness of interventions involving carers of people with dementia increases it would be beneficial for future reviews to attempt a meta-analysis of results where possible. Evidence was from a range of countries and there is a question as to how transferrable the findings are, given the heterogeneous nature of health and social care provision across different countries.

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## Chapter 3: A systematic review of outcome measures used in research involving carers of people with dementia

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## **Abstract**

### *Background*

Advisory bodies, such as the National Institute for Health and Care Excellence (NICE) in the United Kingdom, advocate using preference based instruments to measure the quality of life (QoL) component of the quality-adjusted life year (QALY). Cost per QALY is used to determine cost-effectiveness, and hence funding, of interventions. QALYs allow policy makers to compare the effects of different interventions across different patient groups. Generic measures might not be sensitive enough to fully capture the QoL effects for certain populations, such as carers, so there is a need to consider additional outcome measures, which are preference based where possible to enable cost-effectiveness analysis to be undertaken.

### *Purpose*

This chapter reviews outcome measures commonly used in health services research and health economics research involving carers of people with dementia.

### *Methods*

An electronic database search was conducted in PubMed, Medline, the Cumulative Index to Nursing and Allied Health Literature (CINAHL), PsycINFO, the National Health Service Economic Evaluation Database (NHS EED), Database of Abstracts of Reviews of Effects (DARE) and Health Technology Assessment database. Studies were eligible for inclusion if they included an outcome measure for carers of people with dementia.

### *Results*

Two-thousand, two-hundred and sixty-two articles were identified. Four-hundred and fifty-five articles describing 361 studies remained after exclusion criteria were applied. Two-hundred and twenty-eight outcome measures were extracted from the studies. Measures were categorised into 44 burden measures, 43 mastery measures, 61 mood measures, 32 QoL measures, 27 social support and relationships measures and 21 staff competency and morale measures.

### *Conclusions*

The choice of instrument has implications on funding decisions; therefore, researchers need to choose appropriate instruments for the population being measured and the type of intervention undertaken. If an instrument is not sensitive enough to detect changes in certain populations, the effect of an intervention might be underestimated, and hence interventions which might appear to be beneficial to participants are not deemed cost-effective and are not funded. If this is the case, it is essential that additional outcome measures which detect changes in broader QoL are included, while still retaining preference based utility measures such as EQ-5D to allow QALY calculation for comparability with other interventions.

## **Introduction**

In the United Kingdom, the government faces an increasing challenge to meet the growing demands on the health care system. Despite increased public expectations of treatment availability, an ageing population and higher levels of chronic disease, the government is aiming to achieve efficiency savings of £20 billion in the National Health Service's (NHS) budget by 2014 (Department of Health, 2010b). Savings are to be made through focusing on quality, innovation, productivity and prevention. Treatments offered on the NHS must be clinically effective and cost-effective, as assessed by the National Institute for Health and Care Excellence (NICE). The NICE guide to technology appraisal (NICE, 2013) states that cost-effectiveness should be reported in quality-adjusted life years (QALYs), a measure combining length of time with quality of life (QoL). Therefore, the choice of instrument used to measure QoL is important, as the resulting QALY calculations determine whether a treatment is cost-effective and hence potentially funded. The issue of NICE cost-effectiveness funding thresholds might only be applicable to the United Kingdom; however, the methodological issue of measuring and valuing carer benefits has international application.

Dementia places a large burden on the economy, with costs incurred by the health care sector, social care sector and informal carers (Luengo-Fernandez, Leal & Gray, 2010). The largest proportion of the cost (55%) is incurred by informal carers looking after a friend or relative, and is indicative of the burden faced by carers. Carer burden can predict institutionalisation of the person with dementia (Elmståhl, Ingvad & Annerstedt, 1998; O'Donnell et al., 1992); therefore evidence of effective methods to support carers in their role needs to be established to delay institutionalisation and the associated costs. Burden can affect QoL through decreased mental health caused by stress and worry, and also the opportunity cost of reduced time for leisure activities and self-care (Bell, Araki & Neumann, 2001).

The need to use appropriate outcome measures in health economics research has been recognised (Kelly, McDaid, Ludbrook, Powell, 2005; Craig et al., 2008; Payne, McAllister & Davies, 2012). Interventions involving people with dementia and their carers might be complex with multiple objectives; therefore it is necessary to consider multiple outcome measures. Focusing on one attribute, such as QoL, might lead to other benefits being overlooked. Moniz-Cook et al. (2008) argued that a more cohesive approach to outcome measurement in dementia care research will lead to a more robust evidence base. Health economists require preference based utility measures for calculating QALYs. However, restricting benefit measurement to health-related outcomes in carer research places a patient

identity on the carer, which might not be appropriate (Al-Janabi, Flynn & Coast, 2011a). This chapter aims to address the question ‘what outcome measures are used most frequently in interventions involving carers of people with dementia, and how useful are these measures for economic evaluation?’

## **Methods**

A systematic literature search of electronic databases was conducted on 1st March, 2012. PRISMA reporting principles were used as guidance (Liberati et al., 2009). PubMed (1946-2012), Medline (1950-2012), the Cumulative Index to Nursing and Allied Health Literature (CINAHL) (1981-2012), PsycINFO (1806-2012), and the NHS Centre for Reviews and Dissemination (containing the National Health Service Economic Evaluation Database (NHS EED), Database of Abstracts of Reviews of Effects (DARE) and Health Technology Assessment database) (1960-2012) were searched. Titles, keywords and abstracts were searched for the terms caregiver, randomised controlled trials and dementia or Alzheimer’s disease using MeSH terms where possible. The search strategy is given in Appendix 7, page 189. No restriction on publication year was set. Study eligibility was based on initial screening of title and abstract by the lead author (CJ). Titles and abstracts were not screened by a second reviewer. Articles passing initial screening were retrieved for further review.

Studies were considered if they reported an intervention with outcome measures for carers of people with dementia. Carers could be paid workers or informal carers, such as friends and family members. We included outcomes for paid carers to get a broader indication of which aspects of health and social care provision are typically measured. No gender, age or nationality restrictions were applied. The person being cared for could be living in residential care, a medical facility or the community.

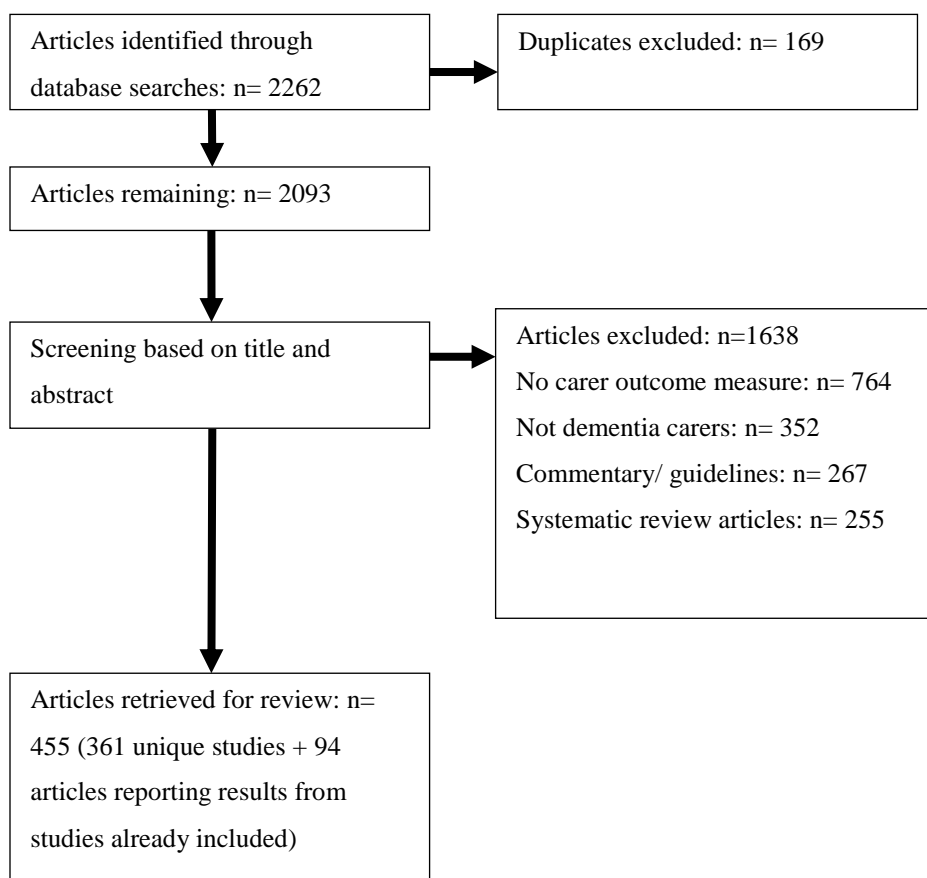
Carer outcome measures were extracted and categorised. The categories used in Moniz-Cook et al. (2008) were a starting point: burden, mood, quality of life and staff competency and morale. Two additional categories were developed after reviewing the data: mastery and social support and relationships.

## **Results**

Two-thousand, two-hundred and sixty-two records were retrieved, 2093 articles remained after duplicates were removed (Figure 3.1). After screening titles and abstracts, 1638 articles

were excluded. Exclusion reasons included no carer outcome measure (764 articles), the population not consisting of dementia carers (352 articles), commentary articles or clinical practice guidelines (267 articles) and systematic review articles (255 articles). 455 articles reporting on 361 studies remained. 228 outcome measures were extracted. A full list of extracted outcome measures, the number of studies they appeared in and their earliest and most recent year is in Appendix 8, page 190. Table 3.1 presents key properties of outcome measures appearing in four or more studies (1% of included studies). Table 3.2 shows the change in composition of carer outcome measures used over the years.

**Figure 3.1:** Flow of articles retrieved through electronic searches



**Table 3.1:** Properties of the most frequently used outcome measures

Category	Outcome measure	Number of studies	Publication year	Region of development	Number of items	Number of levels per item	Dementia specific?
Burden	Zarit Burden Interview	76 (21.1%)	1983	USA	22	5	Yes
	Revised Memory and Behavior Problems Checklist (RMBPC)	44 (12.2%)	1992	USA	24	6	Yes
	Relatives Stress Scale	13 (3.6%)	1982	UK	15	5	Yes
	Novak Caregiver Burden Inventory	11 (3.0%)	1989	Canada	24	4	Yes
	Screen for Caregiver burden	11 (3.0%)	1991	USA	25	5	Yes
	Perceived Stress Scale	11 (3.0%)	1983	USA	14	5	No
	(Revised) Caregiving Burden Scale	6 (1.7%)	1994	Netherlands	13	5	Yes
	Caregiver Stress Scale	4 (1.1%)	1990	USA	15	3-5	Yes
Mastery	Sense of Competence Questionnaire	12 (3.3%)	1996	Netherlands	27	2-5	Yes
	Brief Coping Orientation for Problems Experienced (COPE)	6 (1.7%)	1997	USA	28	4	No
	Ways of coping scale	6 (1.7%)	1985	USA	64	2	No
	Revised Scale for Caregiving Self Efficacy	6 (1.7%)	1999	USA	19	Rated 0-100	No
Mood	Center for Epidemiologic Studies Depression Scale (CES-D)	57 (15.8%)	1977	USA	20	4	No
	General Health Questionnaire (GHQ) <sup>1</sup>	31 (8.6%)	1978	UK	28	4	No
	Neuropsychiatric Inventory- Carer Distress (NPI-D)	30 (8.3%)	1998	USA	12	6	Yes
	Geriatric Depression Scale	19 (5.3%)	1982	USA	30	2	No
	Beck Depression Inventory (BDI)	17 (4.7%)	1961	USA	21	4	No
	Neuropsychiatric Inventory Questionnaire (NPI-Q)	12 (3.3%)	2000	USA	12	6	Yes
	Brief Symptom Inventory	8 (2.2%)	1983	USA	53	5	No
	Hamilton Depression Scale	8 (2.2%)	1960	UK	17	3-5	No
	Pittsburgh Sleep Quality Index	8 (2.2%)	1989	USA	24	1-4	No
	Hospital Anxiety and Depression Scale	7 (1.9%)	1983	UK	14	4	No
	State-Trait Anxiety Inventory	7 (1.9%)	1970	USA	20	4	No
	Positive and Negative Affect Scale (PANAS)	5 (1.4%)	1988	USA	20	5	No
	Hopkins Symptoms Checklist	4 (1.1%)	1974	USA	58	4	No
	Positive Aspects of Caregiving	4 (1.1%)	2004	USA	9	5	Yes

Quality of life	Short Form-36 (SF-36)	32 (8.8%)	1988	USA	36	2-6	No
	EuroQoL (EQ-5D)	18 (5.0%)	1990	Europe	5	3	No
	World Health Organization Quality of Life-Bref (WHOQOL-BREF)	8 (2.2%)	1996	Global	26	5	No
	Health Utilities Index Mark <sup>2</sup>	4 (1.1%)	1990	Canada	8	5-6	No
Social support and relationships	Social Support Questionnaire	7 (1.8%)	1983	USA	27	6	No
	Stokes Social Support network List	4 (1.1%)	1983	USA	N/A Matrix	N/A	No
Staff competency and morale	Maslach Burnout Inventory	10 (2.6%)	1981	USA	22	3-7	No
	Approaches to Dementia questionnaire	4 (1.1%)	2000	UK	19	5	Yes

<sup>1</sup> Information given for the 28 item version (GHQ-28)

<sup>2</sup> Information given for the Health Utilities Index Mark III (HUI3)

**Table 3.2:** Composition of outcome measures across the years

Years	Number of included papers	Number of measures used, percentage composition for the time period					
		Burden	Mastery	Mood	QoL	Social support and relationships	Staff competency and morale
1985-89	5	2 (12%)	1 (6%)	11 (65%)	3 (18%)	0	0
1990-94	10	8 (42%)	1 (5%)	7 (37%)	1 (5%)	0	2 (11%)
1995-99	33	27 (36%)	12 (16%)	30 (40%)	1 (1%)	4 (5%)	1 (1%)
2000-04	86	68 (32%)	31 (15%)	69 (33%)	23 (11%)	16 (8%)	4 (2%)
2005-09	148	99 (26%)	60 (16%)	131 (34%)	44 (11%)	33 (9%)	19 (5%)
2010-12	79	50 (21%)	40 (17%)	67 (29%)	38 (16%)	24 (10%)	14 (6%)



### *Burden measures*

The 44 measures in this category consisted of burden, stress and strain. Burden was the second most popular category of measure used in dementia carer research. The Zarit Burden Interview (ZBI) (Zarit, Reever & Bach-Peterson, 1980) was most popular, appearing in 76 studies (21%). The ZBI is dementia specific, originally a 29-item instrument but also available as a shorter 12-item version (Bédard et al., 2001). Domains of the ZBI cover physical health, psychological well-being, finances, social life, and relationship with the person with dementia. The earliest paper retrieved which included the ZBI was published in 1994; the ZBI is still used currently. The Revised Memory and Behavior Problem Checklist (RMBPC) (Teri et al., 1992) was the second most popular measure, appearing in 44 studies (12%). It is also dementia specific and contains 24 items adapted from the Memory and Behavior Problem Checklist (MBPC) (Zarit, Orr & Zarit, 1985). The MBPC assesses the frequency and severity of problems exhibited by a person with dementia and their carer's reaction to these problems. As with the ZBI, the RMBPC has also been in use since 1994 and is still used today.

### *Mastery*

Forty-three measures encompassing the family carer's coping, self-efficacy and competence were extracted. As can be seen in Table 3.2, mastery measures were infrequently used in earlier studies. Currently, mastery measures account for 17% of the outcome measures included in dementia carer research. The Sense of Competence Questionnaire (SCQ) (Vernooij-Dassen, Persoon & Felling, 1996) was most popular, appearing in 12 studies (3%) since the year 2000. The SCQ was developed to measure the ability of carers to cope with looking after people with dementia living at home. Three domains are covered: satisfaction with the person receiving care, satisfaction with one's own performance as a carer and the impact of caring on the personal life of the carer.

### *Mood*

Mood measures were included the most frequently, and currently account for almost one third of dementia carer measures included. Sixty-one mood measures covering anxiety, depression, sleep and general mental health were extracted. The Center for Epidemiologic Studies Depression Scale (CES-D) (Radloff, 1977) was the most frequently used measure, appearing from 1989 onwards. CES-D was followed in frequency by the General Health Questionnaire (GHQ) (Goldberg, 1978) and the Neuropsychiatric Inventory-Distress (NPI-D) (Kaufert et al.,

1998). The NPI-D primarily assesses the frequency and severity of behavioural disturbances occurring in people with dementia, but also asks carers to rate their reaction to the behaviours. The NPI-D is one of the more recently developed mood measures, first appearing in the year 2000. The next most popular measures were the Geriatric Depression Scale (Yesavage et al., 1982) which was developed for use in an elderly population, the Beck Depression Inventory (BDI) (Beck, Ward, Mendelson, Mock & Erbaugh, 1961) and the Neuropsychiatric-Questionnaire (NPI-Q) (Kaufer et al., 2000), a version of the NPI-D suitable for use in a clinical setting which has appeared in publications from 2006 onwards.

#### *Quality of life measures*

Thirty-two QoL measures were identified. While QoL measure inclusion has increased over the years, only 16% of included outcome measures are currently for QoL. Four outcome measures were used most frequently: the Short Form-36 (SF-36) (Ware & Sherbourne, 1992), the EuroQoL (EQ-5D) (EuroQoL Group, 1990); the World Health Organization Quality of Life-brief (WHOQOL-BREF) (World Health Organisation [WHO], 1996) and the Health Utilities Index (HUI) (Furlong, Feeny, Torrance & Barr, 2001). The SF-36 and EQ-5D appeared in publication from 2001 onwards, while the WHOQOL-BREF is a more recent, appearing 2007 onwards.

The SF-36 evolved from the RAND Health Insurance Experiment, a fifteen year study of American health policy; and the Medical Outcome Study of patients with chronic illnesses (Ware & Sherbourne, 1992). The SF-6D was subsequently developed; enabling preference based utility scores and QALYs to be calculated from the SF-36 or SF-12 (Brazier, Usherwood, Harper & Thomas, 1998; Brazier, Roberts & Deverill, 2002). While it is possible to use the SF-6D directly in a study, developers recommend using the SF-36 or SF-12 and then translating results into the SF-6D. The six domains of the SF-6D are physical functioning, role limitation, social functioning, pain, mental health and vitality.

The EQ-5D was developed in Europe and consists of a questionnaire (EQ-5D) and a visual analogue scale (EQ-VAS). The EQ-5D comprises five domains: mobility; self care; usual activities; pain and discomfort; and anxiety and depression. A scoring algorithm converts responses into an index score which can be used to calculate a QALY. On the EQ-VAS, respondents are presented with a thermometer with markings representing the worst and best imaginable health state. Respondents are asked to draw a line to mark the level they would

describe their QoL as being. While the scoring of the EQ-5D is preference based, the EQ-VAS is not.

The WHOQOL-BREF is derived from the WHOQOL-100, an instrument developed by a global research team and intended to be applicable cross-culturally (WHO, 1996). The domains of the WHOQOL-BREF can be broadly categorised into physical health, psychological well-being, social relationships and the environment. Preference based utility scores are not available for either the WHOQOL-BREF or WHOQOL-100.

The Health Utilities Index has two main versions: the HUI2 used with children, and the HUI3 used with adults. The HUI3 has eight domains: vision, hearing, speech, ambulation, dexterity, emotion, cognition and pain (Furlong, Feeny, Torrance & Barr, 2001).

#### *Social support and relationships*

The earliest published use of a social support or relationship measure was in 1999. Twenty-seven measures were identified in this category. Only the Social Support Questionnaire (Sarason, Levine, Basham & Sarason, 1983) and the Stokes Social Support Network List (Stokes, 1983) were used consistently, neither was developed for dementia carers. The Social Support Questionnaire assesses the respondent's perceived number of social support contacts and their satisfaction with the social support available. The Stokes Social Support Network List asks respondents to list people they have contact with on a regular basis and whether or not they are relatives. The respondent's social network size and composition is then determined. The Stokes Social Support Network List is a recent measure, appearing in publications dated 2006-2010.

#### *Staff competency and morale*

Staff competency and morale measures were included from 1994 onwards. Twenty-one measures were identified. Only two questionnaires were used in four or more studies; the Maslach Burnout Inventory (Maslach & Jackson, 1981) and the Approaches to Dementia Questionnaire (Lintern, Woods & Phair, 2000). Burnout is described as the emotional exhaustion and cynicism experienced by staff involved with people-facing roles (Maslach & Jackson, 1981), and the consequences of burnout are low quality of care, low morale and high staff turnover. The Approaches to Dementia Questionnaire assesses the carer's attitude toward the care recipient.

## **Discussion**

The key to selecting appropriate outcome measures is defining what an intervention targets, and therefore what a measure has to be able to capture. As can be seen in Table 3.2, the composition of measures included in dementia carer research has changed over time. In earlier years, mood measures were the most prevalent. While this is still true of current research, the gap between use of mood and burden measures has narrowed. Measures capturing social support and relationships are more commonly used now.

Whichever instrument is used, NICE prefers results to be converted into a QALY to allow comparisons across different illnesses and interventions (NICE, 2013). To satisfy QALY methodology, quality weights must be based on preferences and anchored on an interval scale which contains full health and death points (Drummond et al., 2005). Preference-based generic instruments, such as the EQ-5D are preferred (NICE, 2013). Validity of the instrument selected is important for results to be meaningful. The most popular measures in the QoL category have been validated with members of the general population.

The aggregation of carer and patient QALYs is rarely undertaken; however, one trial of befriending for carers of people with dementia presented the incremental cost-effectiveness ratio (ICER), as calculated with the EQ-5D for the QALY component, for both the carer alone and the carer and person with dementia combined (Charlesworth, Shepstone, Wilson, Thalanany, Mugford & Poland, 2008). The intervention was not cost-effective when the ICER was calculated for the carer alone, but became cost-effective when the effects on the person with dementia were incorporated. Aggregation of QALYs needs to be undertaken cautiously, with the information used to calculate resulting ICERs explicitly stated to allow for comparisons with interventions where QALYs have not been aggregated.

Out of the most popular instruments in the QoL category, only weights for the EQ-5D were derived using the time trade-off method. The SF-6D and HUI3 were valued using a visual analogue scale and standard gamble; the WHOQOL-BREF does not have preference based scoring. Three possible explanations for differences in health state valuations between measures have been put forward: coverage of descriptive systems, sensitivity of dimensions and valuation methods (Brazier, Ratcliffe, Salomon & Tsuchiya, 2007). Instruments which describe more health states will pick up smaller changes in health status and are more appropriate for research where smaller health gains are expected to be made (Grieve,

Grishchenko & Cairns, 2009), such as research involving carers. The HUI3 can describe 972,000 health states; the SF-6D either 7,500 or 18,000 depending on the version, while the EQ-5D only describes 243 health states. A ‘ceiling effect’, where higher health states are chosen more frequently, is known to be a feature of the EQ-5D. In contrast, the SF-6D appears to have a ‘floor effect’, with responses clustered at the lower end of the scale. The floor effect is amplified in population groups with more physical health problems, so might not be an issue when conducting research with carers of people with dementia. This is because although many carers do have health issues, one might assume that they already have reasonable physical health to be able to cope with the physical aspects of caring.

The World Health Organisation defines health as ‘*a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity*’; a definition unchanged since 1948 (WHO, 2012a). Furthermore, the seven determinants of health are suggested as: income and social status, education, physical environment, social support networks, genetics, health services and gender (WHO, 2012b). This reinforces the idea that we need to go beyond physical health measurement, and consider other attributes affecting QoL. This is particularly relevant for dementia carers, as research is primarily aimed at relieving burden rather than improving physical health.

While the EQ-5D covers physical domains well there is only one question on mental health. Due to the dominance of physical domains, it is not particularly sensitive to changes in carers of people with dementia, who might not see changes in their physical health over time though their QoL is still affected. This issue was raised by Al-Janabi et al. (2011a), who posed that measuring health-related outcomes for carers places a ‘patient’ identity on carers. In a cross-sectional study involving carers of people with dementia completing the HUI2, Neumann et al. (1999b) found that the stage of Alzheimer’s Disease was a negative predictor of patient utility (as reported by carers completing the HUI2 as a proxy); however, the utility that carers reported for themselves was insensitive to the stage of the care recipients dementia. For research involving carers of people with dementia it might be necessary to include additional outcome measures alongside a generic primary outcome measure for cost-effectiveness analysis.

It has been found that disease specific instruments are better at detecting QoL changes than generic instruments (Pickard, Yang & Lee, 2011). The main advantage of disease specific instruments is that they are sensitive to changes associated with the disease in question;

therefore studies do not need a large sample size. A disadvantage is that co-morbidities might be overlooked; by focusing on QoL changes associated with one particular illness, separate health issues are ignored. As people with dementia and their carers tend to be older, co-morbidities and side effects are particularly relevant. Disease specific instruments are typically focused on the person with the illness; therefore using a population group measure might be more appropriate for carers. Population specific measures cover a broader range of domains than disease specific instruments, with the additional benefit of being more sensitive than a generic instrument. This review found that the most popular instruments in the burden category were developed specifically to measure burden in dementia carers, combining disease specific with population specific domains.

This review found 29 studies which included details of costs; however, most of these were only partial economic evaluations which provided cost-outcome descriptions. Where cost-effectiveness analyses had been performed the unit of effect was typically time, for example cost per additional year that the person with dementia lived at home, cost per reduction in hours spent on care tasks per day. Cost-utility analysis was included in three studies (Charlesworth et al., 2008; Neumann et al., 1999a; Drummond, Mohide, Tew, Streiner, Pringle & Gilbert, 1991); the outcome measures used were the EQ-5D, HUI2 and the Caregiver Quality of Life Instrument (CQLI) (Mohide et al., 1988). The EQ-5D and HUI2 are frequently used measures discussed briefly in the results section. The CQLI was only used in one study (Drummond et al., 1991). It was developed to capture QoL of carers of elderly people and utility values were elicited using the time trade-off technique. The domains of the CQLI include: time to socialise with friends and family; quality of the relationship with the person being cared for; physical wellness and energy; sleep; happiness and freedom from anxiety/ worry. The CQLI was found to be feasible, responsive and demonstrated construct validity; however, it was not widely adopted by the health economics community after its development.

All three measures are suitable for QALY calculations. The study that included the cost-utility analysis using the HUI2 (Neumann et al., 1999a) aggregated carer and patient QALYs, which as mentioned above is not consistent with traditional QALY methodology. Nine of the studies listing costs were protocols, seven planned to conduct cost-utility analysis using the EQ-5D and two planned to conduct cost-utility analysis using the SF-12 or SF-36.

Overall, burden and mood measures were the most frequently used. The earliest article retrieved from the searches was published in 1987 and included four mood measures and one QoL measure. Outcome measures in the mood category covered a broad range of symptoms from overall mental health, anxiety, depression and sleep quality. A variety of social support measures were used; the two most frequently used measures were not specific to dementia carers. Social support measures have grown in popularity but are still not as frequently used as burden, mastery, mood or QoL measures. The least frequently used category of measure was the staff competency and morale category. A large number of unspecified measures were found, mainly due to poor reporting of study methods precluding the authors of this review being able to identify the measure used. The increased use of guidelines such as CONSORT (Schulz, Altman, Moher & The CONSORT Group, 2010), has improved the quality of reporting of trials in recent years.

### **Future Directions**

The ICECAP index of capability has been developed to measure attributes of QoL rather than influences on QoL, for example health (Coast, Flynn, Natarajan, Lewis, Louviere & Peters, 2008b). The theory is that QoL does not decrease due to poorer health, but instead decreases through limitations in what one can do as a result of poor health, i.e. individuals value the activities that they can undertake rather than health itself. In this sense, instruments such as the EQ-5D are only a proxy measure for QoL rather than a direct measure (Grewal, Lewis, Flynn, Brown, Bond & Coast, 2006). Two versions of the ICECAP are available: the ICECAP-O, suitable for ages 65+; and the ICECAP-A, suitable for ages 18+. The domains of the ICECAP-O are: love and friendship; thinking about the future; doing things that make you feel valued; enjoyment and pleasure and independence. These domains were developed to measure capability in older members of the general population (Coast, Peters, Natarajan, Sproston & Flynn, 2008b) and have a certain degree of overlap with the categories of burden, mastery, mood, quality of life, and social support and relationships. The domains of the ICECAP-A are similar: security; love and friendship; independence; achievement and enjoyment and pleasure. Currently, an algorithm to convert ICECAP scores into a QALY is not yet available. One way around this is to perform a mapping exercise of ICECAP scores onto EQ-5D scores. To be valid this would require considerable time and financial resources to construct the necessary data set.

The capability framework has also led to the development of the Adult Social Care Outcomes Toolkit (ASCOT) (Netten et al., 2011), an instrument to measure social care-related QoL.

While the ASCOT does not specifically measure carer QoL, it is a step toward acknowledging the importance of the care environment that a person is living in. Domains of the instrument include: control over daily life; personal cleanliness and comfort; food and drink; personal safety; social participation and involvement; occupation; accommodation cleanliness and comfort; and dignity. While the domains are similar the ICECAP, the advantage that the ASCOT tool has is that it is a preference-based measure with scoring reflecting preferences of the general population (Netten et al., 2012).

## **Conclusion**

Few studies currently incorporate economic evaluations alongside clinical trials as routine practice. The choice of instrument used to measure QoL has implications for whether or not a treatment is considered cost-effective and potentially funded. Health economists need to choose instruments appropriate for the population and expected outcomes. Researchers need to consider ease of administration and clarity of instrument to ensure as many participants as possible complete questionnaires. For carers of people with dementia, available time is already restricted so there is a need to avoid overburdening participants with lengthy questionnaires. If an instrument is not sensitive enough to detect changes in QoL for carers of people with dementia, the effect of an intervention which might appear to be beneficial to participants are underestimated. Capability based instruments which capture the impact of an intervention on both health and broader aspects of quality of life offer the opportunity for decision-makers to evaluate interventions that improve QoL without necessarily improving health.



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Chapter 4: Qualitative exploration of the suitability of capability based instruments to measure quality of life in family carers of people with dementia

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## **Abstract**

### *Background*

In our ageing population, many individuals find themselves becoming a carer for an elderly relative. Caring is associated with increased burden; it is timely for policymakers to consider whether instruments focusing on physical health gains, such as the EQ-5D, are appropriate for carer research. The capability approach offers a potential alternative. This qualitative study explores aspects of quality of life affected by caring for a person with dementia, with the aim of identifying whether capability based instruments are more suitable for measuring quality of life in carers than the EQ-5D.

### *Methods*

Semi-structured interviews lasting up to an hour were conducted between November 2010 and July 2011 with eight family carers of people with dementia living in North Wales. Interviews typically took place at the participants' home, and were recorded and transcribed verbatim. Framework analysis was used to code and analyse data. Domains from three capability based instruments (the ICECAP-O, Carer Experience Scale and the Adult Social Care Outcomes Toolkit) were used as initial codes. Similar codes were grouped into categories, and broader themes were developed from these categories.

### *Results*

Four themes were identified: social network and relationships; interactions with agencies; recognition of role and time for oneself.

### *Conclusions*

By identifying what affects carer quality of life, a more appropriate choice can be made when selecting which instruments to use in future dementia carer research. The themes identified in this chapter had a high degree of overlap with the capability instruments, suggesting that the capabilities approach would be suitable for future research involving carers of people with dementia.

## Introduction

Dementia is a growing problem, affecting over 800,000 people in the United Kingdom at an annual cost of £23 billion (Wimo & Prince, 2010). Informal care by friends and family members accounts for 55% (£12bn) of this cost (Luengo-Fernandez, Leal & Gray, 2010); this would have to be covered by health and social services if carers were unable to cope. Care can include activities of daily living such as assistance with personal care, or instrumental activities of daily living such as helping with shopping, managing finances or supervision to ensure the safety of the person with dementia.

The number affected by dementia is expected to rise with the ageing population, placing a heavier burden on both families and health and social services in the future. From an economic perspective, it is important to support informal carers in their role to enable people with dementia to remain living at home as long as possible. In response to the increasing pressure being placed on scarce health and social care resources, the UK Government has made a commitment to double spending on dementia research to £66 million per year by 2015 following calls by the Alzheimer's Society to increase funding substantially (Lakey, Chandaria, Quince, Kane & Saunders, 2012); therefore it is timely to consider the appropriateness of current health economics instruments used to measure quality of life.

As discussed in Chapter 1, page 10, quality of life is a concept with many definitions and interpretations. In this chapter, the World Health Organisation concept is used (WHO, 1996) as it is a broad interpretation, not restricting itself to measuring physical health or wealth: *“Quality of life is defined as individuals’ perceptions of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns.”* In a survey of the general UK population, respondents were asked to list up to five things that affected quality of life. Over 60% stated relationships with family and other people, 43% selected their own health and 38% selected the health of somebody they were close to (Bowling, 1995). Caring has been found to affect mental health more than physical health (Schulz & Sherwood, 2008), with up to 30% of carers of people with dementia experiencing depression (Schoenmakers, Buntinx & Delepeleire, 2010). However, some carers of people with dementia develop resilience to the caring situation. Longitudinal research has shown that some carers report stability over time in the areas of role overload and depression (Gaugler et al., 2000).

### *Measuring quality of life*

With increased public expectations of what treatments should be made available on the National Health Service (NHS), demand for health care is infinite but resources are scarce; therefore choices have to be made about whether or not to fund specific health care interventions. Informed funding decisions can only be made after a rigorous economic analysis of the costs and benefits of competing alternatives has taken place. In the United Kingdom, the National Institute for Health and Care Excellence (NICE) assesses evidence on the clinical-effectiveness and cost-effectiveness of treatments. The NICE guide to technology appraisal (NICE, 2013) states that effectiveness should be reported in quality-adjusted life years (QALYs), with the generic EQ-5D (EuroQoL, 1990) as the preferred instrument for measurement of the health-related quality of life component of the QALY. Guidance on whether the NHS should fund a treatment is made by NICE based on whether or not the cost per QALY falls below an arbitrarily chosen funding threshold of £20,000- £30,000. Treatments falling above this threshold are not recommended for NHS funding; however, local trusts have the option of overruling the guidance and offering more expensive treatments.

The recommended utility instrument for measuring the quality of life part of the QALY, the EQ-5D, consists of two parts; a five item questionnaire and a visual analogue scale (see Appendices 1 and 2, pages 180-181). Strengths of the EQ-5D including ease of completion and potential for use with a range of illnesses. However, despite NICE favouring the use of the EQ-5D, there are arguments for including alternative outcome measures. The EQ-5D is dominated by physical health questions, which places a 'patient' identity on the carer (Al-Janabi, Flynn & Coast, 2011a). Carer interventions can cross the health and social care sector; therefore instruments focused on physical functioning underestimate the full effects on quality of life. In this chapter we argue for the routine inclusion of broader quality of life measures alongside the EQ-5D in research involving carers of people with dementia. This is necessary to capture the full effects of an intervention, while satisfying the NICE requirement of using the EQ-5D to enable comparability across interventions.

### *An alternative approach*

Capability theory is a growing area in health economics as it allows the opportunity of exploring quality of life beyond physical health gain. Recent developments in the capability framework include the development of three instruments: the ICECAP-O (Grewal et al., 2006) (see Appendix 3, page 182), the Carer Experience Scale (CES) (Al-Janabi et al., 2008,

Al-Janabi et al., 2011b) (see Appendix 4, page 183) and the Adult Social Care Outcomes Toolkit (ASCOT) (Netten et al., 2011) (see Appendix 5, page 184). The ICECAP-O was developed through prior research into quality of life of members of the general population aged 65+ (Grewal et al., 2006), an appropriate instrument as many carers of people with dementia fall into this age group; the Carer Experience Scale was developed through qualitative research with carers (Al-Janabi et al., 2008), and the ASCOT was developed to measure social care related quality of life for the care recipient but the domains might be applicable for evaluation of quality of life for carers too. (Netten et al., 2011). While similar in format to the EQ-5D, these instruments contain domains which measure the capability of an individual to achieve a range of outcomes.

Developing a new instrument to measure quality of life requires a rigorous process beginning with interviewing relevant groups to discover what aspects of quality of life are important to them (Brazier et al., 2007). This qualitative research allows a deeper exploration of a subject and can be conducted alongside quantitative research to enhance understanding and put results into a meaningful context (Coast, 1999). While it is not often possible to generalise findings across a whole population, qualitative research is a useful tool for informing the design of instruments used in quantitative methods.

Measuring quality of life in carers of people with dementia could potentially be undertaken using the ICECAP-O, CES or ASCOT. To explore which of these three instruments would be more suitable for use with this population a qualitative approach was adopted. The qualitative research described in this paper involved a small sample of family carers of people with dementia living in North Wales, and was undertaken to explore the question ‘What do family carers of people with dementia perceive as affecting quality of life, and how well do capability based instruments capture these aspects of quality of life?’

## **Methods**

### *Design*

A framework analysis approach was used to analyse the data. Framework analysis was selected as it is systematic and allows transparency in the data analysis (Smith & Firth, 2011). The framework approach is popular in healthcare research. It is the opposite of more inductive approaches, such as grounded theory, as the focus is not on developing a new theory but instead on describing and interpreting participants’ views. The COREQ checklist (Tong, Sainsbury & Craig, 2007) is used to report the qualitative research presented in this chapter.

### *Participants and recruitment*

Carers were recruited through distributing information sheets face-to-face at Alzheimer's Cafes in North Wales and through the mailing list of the NEURODEM (Wales Dementias and Neurodegenerative Diseases Research Network) Research Participant Register, a register of carers and people with memory problems who have given permission to be contacted about research projects. The information sheet explained the aim of the study, and that information was being collected as part of a PhD study examining quality of life measurement in carers of people with dementia. Carers were asked to contact the lead author (CJ) if they were interested in participating. CJ did not know any of the participants prior to recruitment. It did not matter whether participants were current or former carers because a 'lived experience' viewpoint was sought. Convenience sampling was used and participants were selected opportunistically to ensure as homogeneous a sample as possible. No target sample size was set; participants were recruited until data saturation occurred. Twelve potential participants were approached through the NEURODEM mailing list, and four took part (a 33% response rate).

### *Interview procedure*

Due to the potentially sensitive nature of the topic, one-to-one interviews were held rather than focus groups. Semi-structured interviews were held at a location convenient to the carer, typically their home, between November 2010 and July 2011. Interviews were planned to be held with only the interviewer and participant present; however, in two of the interviews the person with dementia was also present. Interviews were conducted by the lead author (CJ), a female PhD student. Before the interview commenced, CJ asked participants to read the information sheet again and provide written consent to take part. Participants were reminded that they could stop the interview or ask questions at any time. Repeat interviews were not conducted.

An interview schedule containing open ended questions about the participant's experiences as a carer was used (see Appendix 9, page 198). Questions were designed to encourage participants to talk about both past experiences and concerns about the future. Prompts were used to encourage the participant to elaborate more on topics. The interview schedule was not tested prior to use; however after each interview the schedule was reviewed to determine whether modifications were needed. Interviews lasted between 22 and 54 minutes and were recorded using a digital recorder, with additional notes taken during the interview.

### *Data Analysis*

Interviews were transcribed verbatim by CJ with identifying information, such as names, changed to protect confidentiality. Analysis was undertaken in QSR International's NVivo 8 qualitative data analysis software (NVivo, 2008).

The lead author (CJ) familiarised herself with data through repeated listening to the recordings and reading of all of the transcripts. Provisional codes were assigned to transcripts in NVivo by CJ using a line by line coding method. The domains of the ICECAP-O, Carer Experience Scale and ASCOT were used as predetermined codes and were sought in the data. Additional codes were derived inductively. A sample of transcripts were reviewed by co-authors RTE and BH to improve rigour; however, as CJ had led the research and conducted the interviews she was more immersed in the data and was ultimately responsible for coding decisions. Similar ideas thought to affect quality of life were grouped into categories, which were then refined into broader themes by CJ. The original transcripts were cross-checked to ensure that the themes and their interpretation were grounded in the participant's descriptions. Table 4.1 shows the initial codes identified, along with the resulting broader themes. Negative cases were sought to identify contradictions. Transcripts were not returned to participants for comment or correction.

Quotes presented in the text were selected for clarity and relevance. Sections not relevant to the theme have been removed and replaced by ellipses (...). Repetition and hesitations not thought to add meaning, such as 'erm', 'you know' and 'umm' have been removed without ellipses.

### *Data protection*

In compliance with the terms of the Data Protection Act (1988), contact details for participants were stored securely in a password protected file on a computer that only CJ had access to. Anonymised transcripts were also stored securely on the computer. Hard copies of consent forms were archived in a locked cabinet in a locked room, the key being held by CJ.

### *Ethical approval*

Ethical approval for the study was received from Bangor University.

**Table 4.1:** Codes, categories and themes

Predetermined codes	Inductively derived codes	Category	Theme
Social participation and involvement (ASCOT)	Participation	Long term effects of caring; concern for the future	Social network and relationships
Enjoyment and pleasure (ICECAP-O)	Positive coping		
Love and friendship (ICECAP-O)	Being positive		
Anxiety/Depression (EQ-5D)	Blame		
Support from family and friends (CES)	Safety		
Activities outside caring (CES)	Embarrassment		
Getting on with the person you care for (CES)	Isolation		
Fulfilment from caring (CES)			
Assistance from organisations and the government (CES)	Dealing with professionals	Feelings about involvement with decisions; beliefs about health and social care agencies	Interactions with agencies
	Recording experiences		
	Need for information		
Control over caring (CES)	Raising awareness	Helping others	Recognition of role
Doing things that make you feel valued (ICECAP-O)	Respect		
Occupation (ASCOT)	Person with dementia awareness		
Self-care (EQ-5D)	Own health	Direct impact of caring	Time for yourself
Usual activities (EQ-5D)	Feeling overwhelmed		
Independence (ICECAP-O)	Frustration		
Thinking about the future (ICECAP-O)	Guilt		
Control over daily life (CES)	Dignity		
Accommodation, cleanliness and comfort (CES)	Difficulty articulating		
Personal cleanliness and comfort (CES)			

Notes: CES: Carer Experience Scale; ASCOT: Adult Social Care Outcomes Toolkit



## Results

Eight carers were recruited; participant characteristics are displayed in Table 4.2. Four themes were identified: social network and relationships; interactions with agencies; recognition of role and time for oneself.

**Table 4.2:** Characteristics of participants and recipients of their care

		N (%)
Sex of carer	Female	5 (62.5%)
	Male	3 (37.5%)
Relationship to person with dementia	Spouse	7 (87.5%)
	Child	1 (12.5%)
Location of care recipient	Living with carer	4 (50%)
	Long-term residential care	2 (25%)
	Deceased	2 (25%)
Carer employment status	Retired	5 (62.5%)
	Long-term sick	1 (12.5%)
	Part-time employment	1 (12.5%)
	Full-time employment	1 (12.5%)
Mean age of carer*		69.4

\*3 carers did not wish to disclose their age

### *Social network and relationships*

This theme encompasses the social support that carers perceive they have, and how their relationships with both the person with dementia and others had changed. Spousal carers looked first to their husband/wife for social support. A change in the ability to communicate with the person being cared for was a source of upset:

We always had this very strong relationship and we always used to think the same things...Once the Alzheimer's started all his personality changed, that all went, as if we weren't on the same wavelength at all. (C1; female, bereaved spouse)

In the case of the child carer, as dementia had progressed it facilitated a closer relationship than had been experienced before:

I had my arm around her and I always try and massage her back or just touch her hand or just try and be quite tactile with her. And I was thinking, my god she would have hated this.... She wasn't a very tactile person at all... it's come to it...that this has got to happen for us to actually give her a hug. (C4; female, parent in long-term care)

Family and friends were seen as a secondary social support network, both for practical care tasks and emotional support:

We've got two very good sons that live close by...one doesn't do much with his dad but he'll come and say to me 'Oh tidy yourself up and I'll take you out for a meal'....The other son, we see him nearly every day, and he does what his dad can't do anymore. He mends things... Listening to a lot of people, I think I'm alright. (C6; female, spouse)

Relationships with friends and family could also become strained if there was a lack of understanding about dementia and how it was affecting both the person with dementia and the carer:

He came from a big family...they used to come here and the first questions they used to ask him 'do you remember how we used to do so and so?' In the end I had to tell them not to remind him, or ask him questions. Because you'd see then that Robert would get quite frustrated. He wasn't able to remember these things. (C2; female, bereaved spouse)

They're not interested, won't listen to you. It's family, recently I've been trying to get through (to) them there's a problem but they're not interested, as far as they're concerned you look fine so you are fine. (C7; male, spouse)

Carers were anxious about socialising in a wider circle if they felt that the behaviour of the person with dementia might cause embarrassment:

I've been to the memory clinic, or memory café rather, on two or three occasions, but I don't feel that either Brenda or myself have benefitted from that. What it amounts to is that you sit at a table possibly with other people, have a cup of tea and a biscuit, and you might have a talk by the fire brigade or the police, which in the case of Brenda really is of no interest and occasionally she makes adverse remarks very loudly during the lecture, which was an embarrassment. (C8; male, spouse)

Social activities, such as dining out or shopping, were restricted if it was felt that the person with dementia was not enjoying the experience:

He'd become very agitated if he was somewhere strange, and with strange people. So I never stayed long. We used to go to the Christmas dinner, but he always wanted to come home. He was quite safe in his own surroundings. (C2; female, bereaved spouse)

### *Interactions with agencies*

This theme refers to carers' perceptions about their experiences with medical staff, social services and organisations such as Crossroads and the Alzheimer's Society. All carers spoke of difficulty in getting a dementia diagnosis. This was typically caused by the person with dementia not acknowledging that there was a problem and refusing to see a doctor; or once an appointment was made, the doctor not confirming the symptoms as being dementia. The lengthy process of getting a diagnosis caused stress and self-questioning about whether there really was something wrong with the person being cared for:

Naturally for self preservation reasons, clinicians, doctors are very reluctant to say that the patient has Alzheimer's disease. They will mention all sorts of things without actually saying it. (C8; male, spouse)

It took a year to get the diagnosis. Which I think is probably actually fairly quick compared to some people. But it was actually almost a bit of a relief to actually know that we weren't sort of imagining that it, that it was just stress or depression or something like that, and that there was an actual reason for it all. (C3; female, spouse)

Carers felt that information received from the various agencies was fragmented and not received at an appropriate stage of the illness. Some stated that they would have liked more information at the time of diagnosis; others mentioned that at the start of the transition from spouse to carer they did not want to hear about practical care tasks that might become necessary as the person with dementia deteriorated, such as dealing with incontinence and feeding needs:

I think I would have liked to have a little bit more information when, right at the beginning, when we had the diagnosis. But on the other hand I didn't, because we were together. I didn't really want to ask too much, because I didn't want to ask anything that might upset Charles. (C3; female, spouse)

Some carers found the amount of extra support received after a dementia diagnosis was overwhelming:

We had all kinds of people come in....They sent people in to put ramps. I had ramps everywhere in this house. Outside, inside, everything. They put fire alarms in. The physio came. A social worker came.... Somebody else came to see if we had enough benefits...Constant, constant visitors. Perhaps it was a bit much, but....they were trying to help. (C1; female, bereaved spouse)

Two carers suggested that support was difficult to access during the night, with one reluctant to use out of hours services unless in an emergency in case of being seen as a burden:

The CPN gave good support and she gave good advice all along. Occasionally I refused it because I thought I could go my own way, but I was in the wrong. She was very supportive; I could even ring her at night... One of the things that was missing in my case, ... having any support... at night...The CPN said 'You can ring me any time' which wasn't quite true as her mobile would be turned off because she was tending someone else or something else, and at night time I didn't like calling because it's her time off (C5; male, spouse in long-term care)

Where services had not met the carer's expectations a sense of mistrust and cynicism was harboured:

It got to a stage where it was 2 o'clock in the afternoon and she was still in bed, and I felt pretty desperate about it. I got in touch with the social services that suggested that perhaps if someone came in, she would respond to a figure of authority... They used to come in for perhaps ten or fifteen minutes, if she would still not get up they said because they're not allowed to physically intervene they would come and ask for my help anyway. I felt that was a bit of a fiasco. At the end of the couple of months or whatever I had a bill for £300, and I didn't really feel that I'd had very much in the way of assistance. (C8; male, spouse)

Whenever I went to the Alzheimer's Society to ask for advice they say 'Oh, we're not at liberty to give out specific advice' like which homes shall I go to. ... The CPN, community health care people, they said the same: 'We're not allowed to recommend homes.' But frankly I've found out the local psychiatric unit ..., the nurses, they were a bit more outgoing with advice about where to go and where not to go. (C5; male, spouse in long-term care)

### *Recognition of role*

Recognition of role was perceived as an important theme. Caring can be associated with increased levels of stress and depression; however positive aspects such as fulfilment from caring were identified:

In the past Charles would have dealt with a lot of the things that I now... maybe don't deal with but I help him with... He would have dealt with all the financial side of things and the paying of the bills and although I drove, he used to like to do a lot of the driving.... He wasn't a great DIY person or anything like that but it's all the sort of small things that you don't really think that he automatically used to do that I now find myself doing. So I'm probably busier than I have been for quite a few years (laughing). (C3; female, spouse)

Seven participants mentioned events which they believe triggered or accelerated the onset of dementia in the person they cared for. Carers felt guilt or blamed themselves for those events even if they were unavoidable, such as having their own health problems:

When I had the operation I was away from her for two weeks 'cause I had the operation in Cambridge and she had to look after the dogs at home. So she didn't see me for two weeks and when I came back she was quite distressed. She was

quite agitated. (C5; male, spouse in long-term care)

All of the carers had successfully adapted to the change in role from being a spouse/child to a carer:

I'm very much like 'Right, ok, things happen, I need to work on it. I need to be positive and find out more and do things.' I just tend to react to things like that a little bit I think. (C4; female, parent in long-term care)

He's now behaving like one of the boys but in a different (way), because they were learning when they asked you a question but Dale's always asking the same thing and he's not learning any more. It's like having a little boy that's not learning; that's how I describe it. (C6; female, spouse)

Carers were keen to help others through recording their own experiences and raising awareness of dementia and its progression over time:

And I do try and talk to people that I know are going through it, right at the beginning and try and give as much of my information as I can, the things that we went through. That's why I'd quite like to get this little diary that I've got published as a little book.... I just think you could go through and go 'God, that's normal, that bit that happened.' (C4; female, parent in long-term care)

I went to a research meeting with the Alzheimer's Society...and during that course it transpired that anti-psychotics should not be prescribed for more than 3 months without review... I got in touch with the head of the community health people in (memory clinic) and explained to her. She got quite uppity to start. She said 'Well, I did everything according to the book.' I said 'Well perhaps the book isn't right'... And she said 'Oh, I take on board what you said' ... And she's now made sure that the GP is aware. (C5; male, spouse in long-term care)

### *Time for oneself*

The theme of time for oneself recognises the perceived value of having time away from the person being cared for, and hence time away from caring. Participants spoke of being able to resume activities they had previously discontinued because the person being cared for had not shared their interest:

This sounds strange; I've got my life back. My wife and I were opposites when we met...At the time I thought this might be a good marriage, because we can then each benefit from each other's experience. But it never really worked out like that; I tended to abandon all my academic interests...I didn't mind, I didn't begrudge at the time, but now that she's off, and I have every other day to myself and doing things which I did in my youth...I'm taking up movie making again, and things like this, which I did before we got married. (C5; male, spouse in long-term care)

A sense of frustration was evident when the carer was not able to spend time away from the person being cared for:

Television now has become just action; all he can watch is these action things like Schwarzenegger...I can't stand them but I've got to sometimes go along with it and try and read...We do have two televisions at home but if I go to watch he'll find (me) and say 'What you watching? I think I'll watch that then' 'cause he wants to be with me. That's awkward. (C6; female, spouse)

As well as having a greater feeling of independence, the carers of people who had moved into long-term care also spoke of the guilt they experienced at having to make the decision:

It's almost like having rent-a-wife...It's awful, in one way it gives me the freedom but on the other hand I feel awful picking her up for a little while, you know, having a good time and then just dumping her. (C5; male, spouse in long-term care)

## **Discussion**

As Coast (1999) discussed, there is a role for qualitative work in the traditionally quantitative field of health economics; however, researchers must be careful to use methods appropriately to produce work which passes the scrutiny of both health economists and qualitative researchers. The aim of this study was to elicit what carers of people with dementia perceived as impacting on their quality of life, and hence whether capability based instruments capture these aspects of quality of life and are appropriate for use in future health services and health economics research. Four themes were identified: social network and relationships; interactions with agencies; recognition of role and time for oneself.

### *Social network and relationships*

A desire for increased social support was a recurring topic. Spouses felt a sense of loss for the person they used to know and sometimes found it difficult to communicate with the person their spouse had become. A change in personality in the person with dementia could be perceived as positive if the carer felt that the person with dementia had become less inhibited and more affectionate. The subsequent layer of social support was the wider network of friends and family, reflecting the findings of Etters, Goodall and Harrison (2008), who in a review of carer burden noted the importance of positive family relationships and support. Etters et al. (2008) also found that the closer the kinship to the person being cared for, the higher the level of burden perceived. Carers in this study had experienced a reduction in their extended social support network as a result of avoiding social situations and loss of friends

due to normal aging. In a trial of a counselling and support intervention compared to usual care for spouses of people with Alzheimer's disease it was found that carers who utilised support services were able to keep their partner at home longer than those who did not (Mittelman, Haley, Clay & Roth, 2006). The mechanism behind this was thought to be an improvement in response to behavioural problems and increased carer satisfaction with social support, which relates to the theme of social network and relationships found in this study. The social network and relationships theme overlapped with all three capabilities based instruments, with the Carer Experience Scale exhibiting the most overlap. Only the anxiety/depression domain of the EQ-5D was thought to have a clear conceptual overlap with the social network and relationships theme.

### *Interactions with agencies*

Interactions with agencies was closely tied to the domain of 'Assistance from organisations and the Government' on the Carer Experience Scale. Consistent with the findings of Livingston et al. (2010), the diagnosis process was a source of frustration, caused by the reluctance of the person with dementia to admit to there being a problem and a perceived lack of help from medical professionals. Often both factors interacted, with the person with dementia appearing to be fine during an appointment, and staff only assessing symptoms and behaviour observed on the day. Previous qualitative work involving practitioners revealed four obstacles that delayed a formal diagnosis; therapeutic nihilism; risk avoidance; concerns about self competency in managing dementia care; and availability of resources (Iliffe, Wilcock & Haworth, 2006). In our study the carers perceived the delay in diagnosis to be mainly concerned with risk avoidance and therapeutic nihilism on the part of the professionals. Under the Carers (Equal Opportunities) Act 2004 (NICE & SCIE, 2007), carers are entitled to an assessment of their needs by the Social Services. If there is a delay in diagnosis it will hold up the process of the carer being assessed and supported. Opinions were mixed about whether carers should be given a lot of information at the time of diagnosis, or whether knowing the potential outcomes associated with dementia would be distressing. Greater dementia knowledge has been linked to a higher desire to institutionalise previously, with the authors hypothesising that those with more awareness of the progression of dementia recognise sooner that they might not be able to cope with the future demands of caring (Spitznagel, Tremont, Davis & Foster, 2006). In our study, praise was given to staff and services regarded as giving practical advice and information. These findings suggest that the level of information offered to carers should be judged on a case by case basis, with further information given freely if requested.

### *Recognition of role*

The most notable change in role was for the child carer, who described a role reversal whereby she had assumed the parental role and her mother had regressed to childlike. Other carers compared the experience to looking after a child, a perception also found in the Quinn, Clare and Woods (2009) review of dementia caring. Quinn et al. also found that those with a close emotional relationship with the person being cared for prior to the commencement of caring had lower levels of burden and saw caring as rewarding (Quinn et al., 2009). As well as describing stress and burden, carers spoke of positive experiences arising from their new role. These ranged from closer relationships between the carer and person being cared for, to a sense of feeling appreciated. One bereaved carer had become a volunteer befriender to continue giving in a carer role. Some control had been relinquished by the carers of people in long-term care, with decisions being made jointly with the care home. These carers had subsequently become actively involved in fundraising and raising awareness of dementia as a way of helping others. Recognition of role is reflected in the control over caring domain of the Carer Experience Scale, the occupation domain of the ASCOT and the doing things that make you feel valued domain of the ICECAP-O. The domains of the EQ-5D were not thought to describe the recognition of role theme well.

### *Time for oneself*

As well as experiencing fulfilment from caring, participants acknowledged a need for time away from caring. This time was used to catch up on chores as well as pursuing leisure activities. The ability to be independent depends on the level of support received from the social network and agencies so this theme is closely linked to the first two. Younger carers often have more competing time demands as they juggle caring, working and looking after their own family (Quinn et al., 2009), and this was found to be the case for the child carer included in our study. However, a review of caregiver burden and depression suggested that adult child caregivers are more likely to have alternative roles and social activities outside of caregiving, which might moderate the stresses associated with caregiving (Pinquart & Sörensen, 2003). The ASCOT has three domains that cover the time for oneself theme: control over daily life; accommodation, cleanliness and comfort; personal cleanliness and comfort. The ICECAP-O domains of independence and thinking about the future were also linked to the theme. Two themes of the EQ-5D were thought to overlap with the theme: self-care and usual activities.



## **Conclusion**

Exploring quality of life and how experiences shape the capability of individuals to successfully cope with caring is of great importance in dementia care. The EQ-5D, which focuses on physical health, had the least amount of conceptual overlap with the identified themes. Two EQ-5D domains were not thought to fit in with the quality of life themes identified by this work: pain/discomfort and mobility. As NICE prefer cost-effectiveness to be reported as a cost per QALY, calculated with a preference based utility measure, there is a need to continue using the EQ-5D in research involving carers of people with dementia. The themes emerging from this exploratory qualitative analysis suggests that capability based instruments are a potential addition to the health economists' toolkit for measuring quality of life in carers of people with dementia.

All domains of the ICECAP-O and Carer Experience Scale were thought to overlap with the identified themes. This is unsurprising as both instruments were designed to measure capability based quality of life in similar populations; the ICECAP-O for an older population and the Carer Experience Scale for carers. The ASCOT had three domains that did not fit with the themes: dignity, food and drink and personal safety. However, the ASCOT was developed primarily for use with the person receiving social care rather than their carer. For this reason it might be that the ICECAP-O and Carer Experience scale are more suitable for carer research. Both instruments have their strengths and limitations in this area. The ICECAP-O was designed to capture capability based quality of life of people aged 65 years and over, so while it will be suitable for a lot of spousal carers it will not be appropriate for younger people, such as child carers. The advantage of it is that some people in the caring role do not self-identify as carers, so might feel more comfortable completing a broader quality of life measure than one specifically asking about caring. The Carer Experience Scale was developed using interviews with carers for a range of illnesses, and results from this study suggest that it would be equally suitable for dementia carers. In research aiming to capture a greater insight into the pragmatic experience of caring for people with dementia the Carer Experience Scale would be preferable to the ICECAP-O.

## **Limitations**

Qualitative work should be interpreted in its context, which restricts the generalisability of results. Carers in this study were recruited through the Alzheimer's Society and a research register. As such, participants were engaged with a number of local services already and might not be representative of families who do not yet have a formal dementia diagnosis.

Carers were offered a choice of interview location; in two interviews the person with dementia was present, which might have made the carer uncomfortable discussing the negative impact of caring. All participants were white and living in suburban or semi-rural locations; it is unclear whether different themes would emerge from the experiences of carers of different ethnicities or living in an urban area with better access to services and this is an area that needs to be explored in future work.

### **Policy Implications**

Interventions involving carers of people with dementia might have multiple objectives, such as improving burden, coping skills and general quality of life. The need to select appropriate outcome measures for economic evaluations of public health outcomes has been recognised (Kelly et al., 2005; Craig et al., 2008). By focusing on health-related quality of life measurement, the NICE guide to technology appraisal (NICE, 2013) overlooks non-physical benefits of interventions. Bodies allocating research funding should check that outcome measures listed for proposed research match the objectives of the intervention rather than relying on the use of historically popular measures. There is also scope for commissioners of care support services in the community to incorporate the use of capability based measures into audit and service evaluations to ensure a high quality of care is received by those in need.

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Chapter 5: Construct validity of the ICECAP-O for carers of people with  
dementia

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## **Abstract**

### *Background*

Informal care is a large proportion of the cost of caring for people with dementia. Therefore, there is an economic argument for measuring family carers' quality of life with a view to providing appropriate support services.

### *Purpose*

This paper explores the feasibility and construct validity of the ICECAP-O, a capability based quality of life measure, for use with this population.

### *Methods*

Baseline data from the Challenge FamCare (CF) study of family carers supporting people with dementia and challenging behaviour were used. Additional data was collected through a support group sample (SG) survey of people with dementia caring experience. Item completion rates provided feasibility information. Floor and ceiling effects were examined. Cronbach's alpha and Spearman's rho were used to evaluate internal consistency and construct validity respectively.

### *Results*

157 CF and 151 SM carers participated. Feasibility was high; 99% of CF and 96% of SM carers completed all five ICECAP-O items. Ceiling effects were present on 'attachment', 'role' and 'control'. Floor effects were present on 'security'. Good internal consistency was demonstrated, with  $\alpha=0.77$  for CF and  $\alpha=0.84$  for SG carers. Construct validity analysis demonstrated a weak to moderate relationship between the ICECAP-O and other instruments in mostly the expected direction. Mean ICECAP-O and EQ-5D scores were lower than population norms, indicating that carers in our sample experienced poorer health and capability based quality of life than the general population.

### *Conclusions*

This is the first study to examine the construct validity of the ICECAP-O for carers of people with dementia; our results suggest that it is valid and feasible for use with this population.

## **Introduction**

The need to use appropriate outcome measurement instruments in health economics research is being increasingly recognised (Kelly et al., 2005; Craig et al., 2008). To be appropriate, an instrument has to be able to measure what an intervention is targeting; there is little point in using an instrument which measures physical health functioning, such as the EQ-5D, in an intervention targeting improved mental health. Interventions involving carers of people with dementia can target improvements in both health and social care; therefore instruments which offer a broader measure of quality of life than physical health functioning are needed.

Capability theory offers the opportunity to go beyond measurement of physical health gain and obtain a broader measurement of quality of life. Instruments with a theoretical underpinning in the capability framework measure the ability of an individual to undertake activities that they value regardless of whether they subsequently carry out the activities (Grewal et al., 2006). This is a move away from paradigms which aim to maximise utility or health output. Capability theory does not reject the notion of utility, only the notion of relying entirely on utility and excluding all non-utility information (Sen, 1999).

Three relatively recent instruments developed in the capability field are the ICECAP (Coast et al., 2008b; Al-Janabi, Flynn & Coast. 2012), the Adult Social Care Outcomes Toolkit (ASCOT) (Netten et al., 2012) and the Carer Experience Scale (Al-Janabi et al., 2008). The qualitative work described in Chapter 4 found that the domains of these three instruments covered the aspects of quality of life that were important to carers of people with dementia well. We were interested in examining the validity of using a generic capability-based instrument in research involving carers of people with dementia and selected the ICECAP-O as it had been included as an outcome measure in the NIHR funded Challenge Famcare study of family carers supporting people with challenging behaviour in dementia living at home (ISRCTN 58876649); therefore an existing data set were readily available.

As the ICECAP-O is a relatively recent development in the capability field there is a need to explore its suitability for use with different populations through validation work. In its simplest interpretation, validity implies that an instrument measures what it is supposed to measure; an essential requirement for results to be interpreted meaningfully. The validity of an instrument can be established through testing its feasibility, reliability, internal consistency and construct validity. To date, the ICECAP-O has been validated for the general population (Coast et al., 2008c) and in a Dutch nursing home population (Makai et al., 2012). This study

explores the feasibility, internal consistency and construct validity of the ICECAP-O for carers of people with dementia, and the relationship between the ICECAP-O and other instruments.

## **Methods**

### *Participants*

Two data sources were used. The primary source was baseline data from Challenge FamCare, cohort study of family carers supporting people with challenging behaviour in dementia living at home (ISRCTN 58876649). People with dementia and their carers were recruited into Challenge FamCare by Mental Health Team Gatekeepers across 6 NHS Trusts in Humber, Sheffield, Grimsby, Oxford, North East London and Manchester. All participants were new referrals to the Mental Health Teams. The inclusion criteria for the study were: people with dementia living at home; with a family carer; fulfilling diagnostic criteria for dementia based on Diagnostic and Statistical Manual of Mental Disorders- 4th edition (DSM-IV) (American Psychiatric Association, 1994); and fulfilling behavioural criteria for challenging behaviour using the Revised Memory and Behaviour Problems Checklist (RMBPC). Exclusion criteria included being in residential care or respite residential care at the time of recruitment, in the palliative stage of disease at the time of recruitment, and unable to speak or understand English. No explicit exclusion criteria for carers were set. Questionnaires were administered face to face in the participants' home by a member of the Challenge FamCare research team. The carer section of the interviews lasted 60 minutes on average. Ethical approval was received from the York Research Ethics Committee of the National Research Ethics Service. Carer baseline data were collected August 2010 to November 2011.

Additional data were collected through an online survey created in SurveyMonkey (SurveyMonkey, 2011), for people aged 65+ with experience of caring for a person with dementia (see Appendix 11, page 201). Recruitment took place using a variety of methods including a Uniting Carers mail out, attending local Alzheimer's cafes in North Wales, posting on the Alzheimer's Society Online Forum, an advert in the University of the Third Age Newsletter and a Bangor University press release (see Appendix 12, page 208). Carers were given contact details to request a copy of the survey by post if they preferred. As an incentive to take part, carers who completed the survey by 31<sup>st</sup> December 2011 were entered into a prize draw to win one £250 Marks and Spencer voucher. The average length of time to complete the survey online was 12 minutes. Ethical approval was received from Bangor University (see Appendix 10, page 200) and the survey ran between June 2011 and April 2012.

### *Instruments*

Challenge FamCare carer variables included the following socio-demographic information: age, gender, relationship between carer and care recipient, living situation and the amount of time that the person with dementia could be left alone. The following outcome measures were included: ICECAP-O, EQ-5D, General Health Questionnaire (GHQ-12), Quality of Care-giving Relationship (QCGR), Hospital Anxiety and Depression Scale (HADS), Short Sense of Competence Questionnaire (SSCQ) and Relatives Stress Scale (RSS).

The support group questionnaire contained similar socio-demographic questions, along with a self-report health status question ('how would you describe your health in general?'), the ICECAP-O and EQ-5D. The full range of outcome measures collected as part of Challenge FamCare were not included in the support group sample to minimise burden.

The ICECAP-O is a five item measure of capability designed for members of the general population aged 65+ (Grewal et al., 2006). The domains are: 'love and friendship', 'thinking about the future', 'doing things that make you feel valued', 'enjoyment and pleasure' and 'independence'. Respondents rate their capability on a four point scale. Using an algorithm, results are converted into an index score of between 0 and 1, with 0 representing no capability and 1 full capability. The algorithm to calculate the index score was developed using best-worst scaling with a sample of the UK population aged 65+ (Coast et al., 2008b). Best-worst scaling asks individuals to select the best and worst option in a given set of samples (Finn and Louviere, 1992), revealing the preference for different options without asking people to trade one thing for another.

The EQ-5D is a generic health-related quality of life measure consisting of a five item questionnaire and a visual analogue scale (EuroQoL, 1990). The domains of the EQ-5D are: 'mobility', 'self-care', 'usual activities', 'pain and discomfort' and 'anxiety and depression'. Each domain has three levels (no problems, some problems, extreme problems). Responses are converted into an index score of between -0.594 and 1 using an established algorithm (MVH Group, 1999), with 0 representing dead and 1 representing the best possible health state. Preference based tariffs for the different states were elicited using the time trade-off method with a selection of the UK population, enabling comparisons to be made between the EQ-5D index scores obtained in studies and population norms (Kind et al., 1999). The visual analogue scale is a thermometer marked from best possible health to worse possible health,

where respondents are asked to draw a line to the point on the thermometer which best represents their level of health at that moment.

The GHQ-12 assesses the carer's mental current health and how it differs from their usual state. Likert scoring was used with missing items classified as the lowest score in accordance with the standard guidelines (Goldberg, 1978). Domain scores are summed to produce a total GHQ-12 score ranging from 0-36. Lower GHQ-12 scores indicate a better level of health.

The QCGR is a 14 item scale consisting of statements about warmth and relationship negativity (Spruyette, van Audenhove, Lammertyn & Storms, 2002). Respondents are asked to rate their agreement with the statements on a five point scale. Responses are summed to produce a total QCGR score ranging from 14-70. A higher score indicates a better relationship between carer and care recipient than a lower score.

The HADS contains fourteen statements relating to anxiety and depression (Zigmond & Snaith, 1983). Carers are asked to rate the frequency of each statement on a four point scale. Responses are summed to produce a total HADS score ranging from 0-42. A high score indicates higher levels of anxiety and depression.

The SSCQ is a dementia specific instrument designed to measure the ability of carers to cope with looking after people with dementia living at home (Vernooij-Dassen, Persoon & Felling, 1996). The three domains are: satisfaction with the person receiving care, satisfaction with one's own performance as a carer and the impact that caring has on the life of the carer. Respondents rate their agreement on a five point scale, with responses summed to produce a total SSCQ score of 0-35. A higher score indicates a higher sense of satisfaction and competency.

The RSS is a fifteen item scale which asks carers to rate their reactions to caring situations (Greene, Smith, Gardiner & Timbury, 1982). The three sub-scales are emotional distress, social distress and negative feelings toward the care recipient. Respondents rate their agreement to the 15 items on a 5 level scale. Responses are summed to produce a total RSS score of between 0-60. A higher score indicates a higher degree of stress.



### *Data Analysis*

There are estimated to be 670,000 carers of people with dementia in the UK (Alzheimer's Society, 2012b). To achieve a 95% confidence level that the true population would pick an answer that lies within a 5 point confidence interval on an outcome measure would require a sample size of 384. If I were undertaking a definitive construct validity analysis this sample size would be targeted; however, this Chapter presents an exploratory validity analysis of the ICECAP-O with carers of people with dementia.

We followed the approach of Couzner, Ratcliffe and Crotty (2012) in their exploration of the relationship between the ICECAP-O and other health status measures in a population of older adults in a clinical rehabilitation setting. Data were analysed using SPSS version 16.0. Descriptive statistics are reported for the socio-demographic variables. Feasibility was assessed by reporting item completion rates for the ICECAP-O.

Floor and ceiling effects were tested by examining the percentage of respondents who chose the highest and lowest score by each domain. If more than 15% of respondents choose the lowest level of capability, floor effects are present. If more than 15% of respondents choose the highest level of capability, ceiling effects are present (McHorney & Tarloy, 1995). If either floor effects or ceiling effects are present the ICECAP-O cannot be considered able to detect changes in a significant number of the target population.

Internal consistency of the ICECAP-O was assessed using Cronbach's alpha. A Cronbach's alpha value above 0.7 is considered acceptable (Nunnally, 1978).

The relationship between the domains of the ICECAP-O and the domains of the EQ-5D were assessed using a Chi-squared test. The Chi-squared test calculates the expected distribution of two variables based on the null hypothesis that they are not related. A p-value of less than 0.05 indicates a significant difference between observed and expected values.

Construct validity of the ICECAP-O was examined by testing the correlation between the ICECAP-O and other instruments and socio-demographic information using Spearman's rho. The following hypotheses were tested:

1. There is a strong positive relationship between capability, as measured by the ICECAP-O, and carer health status, as measured by the EQ-5D and self-reported

health status question. In their valuation of the ICECAP-O, Coast et al. (2008b) found mean EQ-5D scores of 0.76 (s.d.  $\pm 0.27$ ) and mean ICECAP-O scores of 0.81 (s.d.  $\pm 0.15$ ) for their sample of the general population aged 65+, indicating some correlation between the measures. As the GHQ-12 is scored so that high scores indicate high levels of mental distress it was anticipated that there would be a negative relationship between the ICECAP-O and the GHQ-12, indicating that carers with higher mental health have higher levels of capability

2. Age is negatively associated with capability. Population norms for the mean EQ-5D score decrease with age (Kind, Hardman & Macran, 1999) and it was expected that the ICECAP-O would follow this pattern, indicating that carers of people with dementia experience lower capability as they age.
3. There is a positive relationship between the ICECAP-O and the SSCQ and QCGR, indicating that carers with higher levels of competence and a good relationship with the person being cared for would have a higher level of capability.
4. There is a negative relationship between the ICECAP-O and the RSS and the HADS, indicating that carers with higher levels of capability had lower stress and lower anxiety and depression.

## **Results**

Socio-demographic information for both samples is presented in Table 5.1. Summary statistics for the validity analysis are presented in Table 5.2.

**Table 5.1: Socio-demographic information**

	Challenge FamCare n=157	Support group sample n=151
Participant is currently a carer	157 (100.0%)	84 (55.6%)
Care recipient age	80.34 (s.d. 7.66)	-
Carer age	66.13 (s.d. 13.06)	74.13 (s.d. 6.76) <sup>a</sup>
Gender of care recipient: Male	64 (40.8%)	85 (56.3%)
Gender of care recipient: Female	93 (59.2%)	66 (43.7%)
Gender of carer: Male	46 (29.3%)	42 (27.8%)
Gender of carer: Female	111 (70.7%)	109 (72.2%)
Relationship to care recipient: Spouse	83 (52.9%)	120 (79.5%)
Son/daughter	59 (37.6%)	18 (11.9%)
Son/daughter in law	9 (5.7%)	2 (1.3%)
Sibling	1 (0.6%)	-
Other relative	3 (1.9%)	6 (4.0%)
Friend	1 (0.6%)	3 (2.0%)
Neighbour	1 (0.6%)	-
Missing	0	2 (1.3%)
Carer and care recipient living together	111 (70.7%)	69 (48.9%) <sup>b</sup>
Carer accommodation: Owner occupied	120 (76.4%)	139 (92.1%)
Privately rented	8 (5.1%)	3 (2.0%)
Rented from housing association	16 (10.2%)	7 (4.6%)
Residential/Nursing home	0	1 (0.7%)
Living in friend/relative's house	2 (1.3%)	-
Missing	11 (7.0%)	1 (0.7%)
Amount of time that care recipient can/could be left alone:		
Not at all	39 (24.8%)	24 (15.9%)
An hour or two	39 (24.8%)	29 (19.2%)
Up to half a day	17 (10.8%)	10 (6.6%)
Up to a whole day	9 (5.7%)	5 (3.3%)
Overnight	7 (4.5%)	1 (0.7%)
N/A, care recipient is now deceased	-	33 (21.9%)
Missing	46 (29.3%)	49 (32.5%)
Self-reported health status: Excellent	-	20 (13.2%)
Good	-	86 (57.0%)
Fair	-	36 (23.8%)
Poor	-	6 (4.0%)
Very Poor	-	1 (0.7%)
Missing	-	2 (1.3%)

Key: - Indicates that the variable was not collected as part of the study; <sup>a</sup>n=148, <sup>b</sup>n=149

**Table 5.2:** Summary statistics for validity analysis

	Challenge FamCare (n=157)	Support group sample (n=151)
Completion rate for all 5 ICECAP-O items	99%	96%
Items with ceiling effects present	Attachment, Role, Control	Attachment, Role, Control
Items with floor effects present	Security	Security
Internal consistency	Cronbach's $\alpha = 0.768$	Cronbach's $\alpha = 0.840$
Mean ICECAP-O score	0.72 (s.d. $\pm 0.19$ )	0.76 (s.d. $\pm 0.15$ )
Mean EQ-5D score	0.47 (s.d. $\pm 0.32$ )	0.77 (s.d. $\pm 0.22$ )
Mean EQ-VAS score	52.86 (s.d. $\pm 20.78$ )	-
Correlation between ICECAP-O and EQ-5D	Spearman's rho= 0.194	Spearman's rho= 0.502
Correlation between ICECAP-O and EQ-VAS	Spearman's rho= 0.209	-
Correlation between ICECAP-O and GHQ-12	Spearman's rho= -0.430	-
Correlation between ICECAP-O and age	Spearman's rho= 0.013	Spearman's rho= -0.050
Correlation between ICECAP-O and SSCQ	Spearman's rho= 0.467	-
Correlation between ICECAP-O and QCGR	Spearman's rho= -0.162	-
Correlation between ICECAP-O and RSS	Spearman's rho= -0.254	-
Correlation between ICECAP-O and HADS	Spearman's rho= -0.429	-

**Key:**

ICECAP-O domains have been abbreviated as follows:

Attachment= 'love and friendship';

Security= 'thinking about the future without concern';

Role= 'doing things that make you feel valued';

Control= 'independence'

- indicates that the variable was not collected as part of the support group sample questionnaire. This was to minimise participant burden.

GHQ-12= General Health Questionnaire-12; SSCQ= Short Sense of Competence Questionnaire; QCGR= Quality of Care-Giving Relationship; RSS= Relatives Stress Scale; HADS= Hospital Anxiety and Depression Scale

## **Challenge FamCare**

### *Socio-demographic information*

All 157 Challenge FamCare participants were current carers. 71% were female. Spousal carers constituted 53% of participants. Most of the carers and care recipients lived together, and the most frequent type of accommodation was owner occupied. Mean carer age was 66.13 (s.d.  $\pm 13.06$ ). The mean age of the care recipient was 80.34 (s.d.  $\pm 7.66$ ).

### *Feasibility*

Table 5.3 shows the distribution of responses for each of the ICECAP-O domains. The response rate for the ICECAP-O questions was high, with 99% of Challenge FamCare participants completing all 5 items.

### *Ceiling and floor effects*

Ceiling effects were present for the items 'love and friendship', 'doing things that make you feel valued' and 'independence'. 'Enjoyment and pleasure' was verging on exhibiting a ceiling effect, with 14.6% of participants selecting the highest level of capability for that domain. Floor effects were present for the item 'thinking about the future without concern'.

### *Internal consistency*

The Cronbach's alpha coefficient was 0.768, indicating good internal consistency (Nunnally, 1978). The lowest correlation with the total ICECAP-O score was found to occur on the 'doing things that make you feel valued' item ( $\alpha=0.546$ ), although the 'independence' item also returned a similar value ( $\alpha=0.556$ ).

### *Construct validity*

The mean ICECAP-O score was 0.72 (min-max 0.0-1.00, s.d.  $\pm 0.192$ ). This is lower than the mean score of 0.81 (s.d.  $\pm 0.152$ ) reported by Coast and colleagues in their valuation work of the ICECAP-O for members of the general population aged 65+ (Coast et al., 2008b). Carers reported an EQ-5D mean of 0.47 (min-max -0.43-1.00, s.d.  $\pm 0.316$ ), which is considerably lower than the population norm of 0.78 (s.d.  $\pm 0.26$ ) for members of the general population aged 65-74 (Kind, Hardman & Macran, 1999). The mean EQ-VAS was also lower than the population norms (mean EQ-VAS of 52.86, min-max 0-100, s.d. 20.779 compared to the population norm of 77.32, s.d.  $\pm 18.05$ ). The degree of correlation between the ICECAP-O score and EQ-5D score was weak, with a Spearman's rho value of 0.194, indicating a weak positive relationship. The relationship between the ICECAP-O and the EQ-VAS was similarly

weak, returning a Spearman's rho value of 0.209. Figure 5.1 illustrates the correlation between ICECAP-O scores and EQ-5D scores for the Challenge FamCare and SurveyMonkey and data sets respectively. Data points were spread widely around the line of best fit, indicating a poor correlation.

**Table 5.3:** Distribution of responses to ICECAP-O items

Challenge FamCare sample (n=157)					
	I can have/ do all that I want	I can have/ do a lot	I can have/ do a little	I cannot have/ do any	Item missing
Attachment	56 (35.7%)	<b>62 (39.5%)</b>	28 (17.8%)	11 (7.0%)	0
Security	15 (9.6%)	34 (21.7%)	48 (30.6%)	<b>60 (38.2%)</b>	0
Role	45 (28.7%)	<b>61 (38.9%)</b>	43 (27.4%)	6 (3.8%)	2 (1.3%)
Enjoyment	23 (14.6%)	59 (37.6%)	<b>65 (41.4%)</b>	10 (6.4%)	0
Control	35 (22.3%)	<b>62 (39.5%)</b>	42 (26.8%)	18 (11.5%)	0
Support group sample (n=151)					
	I can have/ do all that I want	I can have/ do a lot	I can have/ do a little	I cannot have/ do any	Item missing
Attachment	33 (21.9%)	55 (36.4%)	<b>57 (37.7%)</b>	4 (2.6%)	2 (1.3%)
Security	14 (9.3%)	38 (25.2%)	<b>67 (44.4%)</b>	30 (19.9%)	2 (1.3%)
Role	35 (23.2%)	<b>64 (42.4%)</b>	45 (29.8%)	2 (1.3%)	2 (1.3%)
Enjoyment	22 (14.6%)	53 (35.1%)	<b>71 (47.0%)</b>	2 (1.3%)	3 (2.0%)
Control	<b>70 (46.4%)</b>	50 (33.1%)	23 (15.2%)	4 (2.6%)	4 (2.6%)

Key:

Domains have been abbreviated as follows:

Attachment= 'love and friendship';

Security= 'thinking about the future without concern';

Role= 'doing things that make you feel valued';

Enjoyment= 'enjoyment and pleasure';

Control= 'independence'

Table 5.4 shows the relationship between dimensions on the ICECAP-O and EQ-5D using a Chi-squared test for the Challenge FamCare sample and support group sample respectively. In the Challenge FamCare sample the following dimensions were statistically significantly related: mobility and enjoyment; self-care and role; usual activities and love and friendship; usual activities and role; anxiety/depression and thinking about the future.

**Table 5.4:** Relationship between dimensions on the ICECAP-O and EQ-5D (p-values) using a Chi-squared test

Challenge FamCare sample (n=157)					
	Mobility	Self-care	Usual activities	Pain/ Discomfort	Anxiety/ Depression
Attachment	0.293	0.721	0.034	0.149	0.361
Security	0.573	0.916	0.726	0.122	0.007
Role	0.310 <sup>1</sup>	0.047 <sup>1</sup>	0.023 <sup>1</sup>	0.801 <sup>1</sup>	0.129 <sup>1</sup>
Enjoyment	0.009	0.228	0.564	0.197	0.171
Control	0.111	0.647	0.114	0.328	0.397
Support group sample (n=151)					
	Mobility	Self-care	Usual activities	Pain/ Discomfort	Anxiety/ Depression
Attachment	0.064 <sup>2</sup>	0.714 <sup>3</sup>	0.016 <sup>2</sup>	0.053 <sup>3</sup>	0.000 <sup>2</sup>
Security	0.003 <sup>2</sup>	0.110 <sup>3</sup>	0.003 <sup>2</sup>	0.661 <sup>3</sup>	0.000 <sup>2</sup>
Role	0.021 <sup>4</sup>	0.568 <sup>5</sup>	0.000 <sup>4</sup>	0.013 <sup>5</sup>	0.000 <sup>4</sup>
Enjoyment	0.057 <sup>6</sup>	0.851 <sup>2</sup>	0.000 <sup>6</sup>	0.002 <sup>2</sup>	0.000 <sup>6</sup>
Control	0.000 <sup>6</sup>	0.000 <sup>6</sup>	0.000 <sup>5</sup>	0.069 <sup>6</sup>	0.000 <sup>5</sup>

Key:

Domains have been abbreviated as follows:

Attachment= ‘love and friendship’;

Security= ‘thinking about the future without concern’;

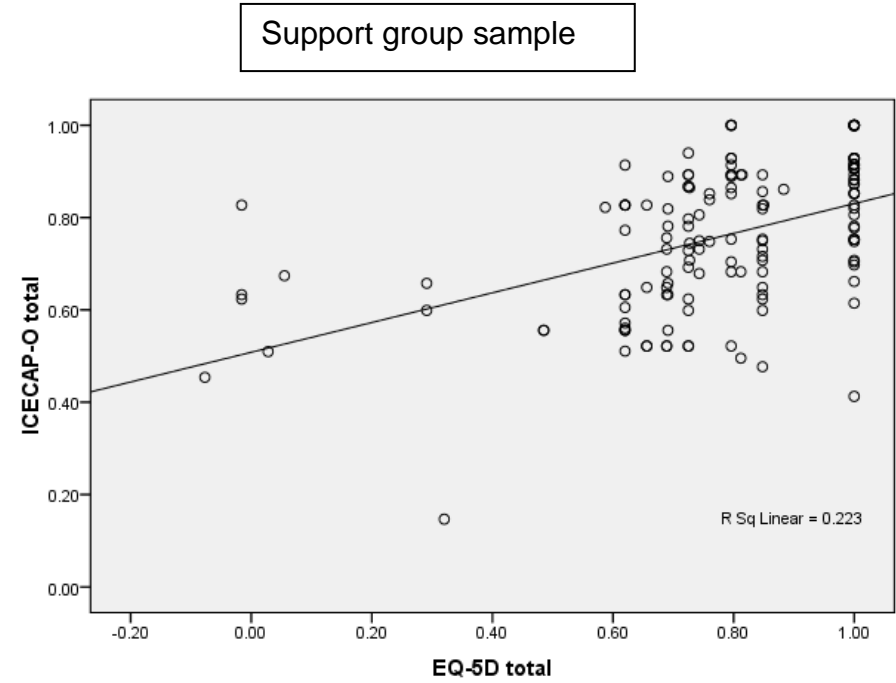
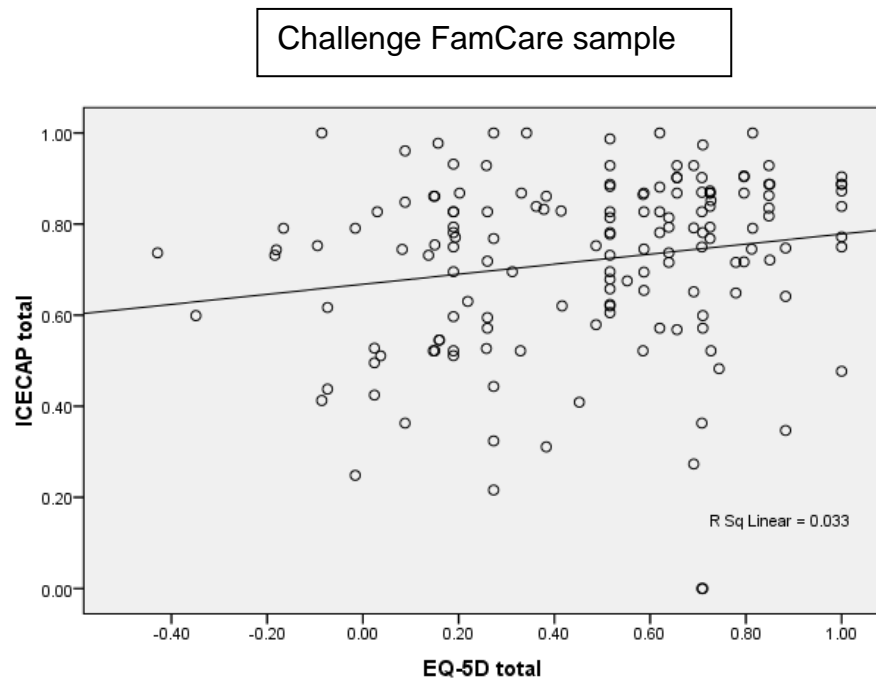
Role= ‘doing things that make you feel valued’;

Enjoyment= ‘enjoyment and pleasure’;

Control= ‘independence’

<sup>1</sup>n=155, <sup>2</sup>n=147, <sup>3</sup>n=148, <sup>4</sup>n=144, <sup>5</sup>n=145, <sup>6</sup>n=146

**Figure 5.1:** Scatter plots showing the relationship between the ICECAP-O and EQ-5D





A moderate negative relationship was found between the ICECAP-O and mental health, as measured by the GHQ-12. The Spearman's rho value for the correlation was -0.430. Carer age was not found to be correlated to the ICECAP-O (Spearman's rho= 0.013). A Mann-Whitney U test found no significant differences in ICECAP-O scores between male and female carers (p= 0.409). Kruskal-Wallis tests found no significant differences in ICECAP-O scores for carers living in different types of accommodation (p= 0.327). Although there was no significant difference in ICECAP-O scores were found between carers who could leave the person with dementia overnight and those who could not leave the person being cared for at all (p= 0.142), the results indicated that carers who could leave the person with dementia on their own for longer periods had higher ICECAP-O scores than carers who could not leave the person with dementia alone.

A moderate positive relationship was found between the ICECAP-O and the Short Sense of Competence Questionnaire (Spearman's rho= 0.467). A weak positive relationship was found between the ICECAP-O and the Quality of Care-Giving Relationship (Spearman's rho= -0.162). A weak negative relationship was found between the ICECAP-O and the Relative's Stress Scale (Spearman's rho= -0.254). A moderate negative relationship was found between the ICECAP-O and the Hospital Anxiety and Depression Scale (Spearman's rho= -0.429).

### **Support group data**

#### *Socio-demographic information*

Data were collected for 151 carers through the support group questionnaire (Table 5.1). Two hundred carers accessed the questionnaire online. Of these, 38 participants were excluded for failing survey eligibility screening questions (32 indicated that they were not aged 65 or over, 3 indicated that they had not read or understood the study information sheet, 3 indicated that they had never been carers); 11 participants were excluded for not continuing the survey beyond the screening questions. 106 (70%) completed the support group questionnaire online, 45 (30%) completed by post or face to face with a researcher.

Over half of support group participants were current carers. As with the Challenge FamCare data set, the majority of participants were female (72%) and the most common relationship between the carer and person with dementia was spousal (80%). 49% of the

carers and care recipients lived together, and the most frequent type of accommodation was owner occupied. Mean carer age was 74.13 (s.d.  $\pm 6.76$ ).

### *Feasibility*

The distribution of responses for each of the ICECAP-O domains by support group carers can be seen in Table 5.3. As with the data from Challenge FamCare carers, the response rate was high, with 96% of support group carers completing all 5 items.

### *Ceiling and floor effects*

Ceiling and floor effects were present in the same domains as in the Challenge FamCare data; ceiling effects were present for the items 'love and friendship', 'doing things that make you feel valued' and 'independence', floor effects were present for the item 'thinking about the future without concern'.

### *Internal consistency*

The Cronbach's alpha coefficient was 0.840, indicating good internal consistency. The lowest correlation with the total ICECAP-O score was found to occur on the 'independence' item ( $\alpha=0.521$ ).

### *Construct validity*

The mean ICECAP-O score was 0.76 (min-max 0.15-1.00, s.d.  $\pm 0.153$ ), which was lower than the mean score of 0.81 (s.d.  $\pm 0.152$ ) reported by Coast et al. in their valuation work of the ICECAP-O for members of the general population aged 65+ (Coast et al., 2008b). Participants reported a mean EQ-5D of 0.77 (min-max -0.08-1.00, s.d.  $\pm 0.224$ ), which is close to the population norm of 0.78 (s.d.  $\pm 0.26$ ) for members of the general population aged 65-74 (Kind, Hardman & Macran, 1999). A moderate positive relationship was found between the ICECAP-O and EQ-5D (Spearman's rho value of 0.502), which is in contrast to the findings in the Challenge FamCare data.

Support group participants were asked to rate their health status on a five point scale from 'Excellent' to Very Poor'. A Kruskal-Wallis test showed that there was a significant difference ( $p=0.001$ ) in ICECAP-O scores between participants in the different health status categories, with respondents in the 'Excellent' category reporting the highest

ICECAP-O scores and respondents in the 'Very Poor' category reporting the lowest ICECAP-O scores.

As with the Challenge FamCare data, carer age was not found to be correlated to the ICECAP-O (Spearman's  $\rho = -0.050$ ). A Mann-Whitney U test found no significant differences in ICECAP-O scores between male and female carers ( $p=0.951$ ). Kruskal-Wallis tests found no significant differences in ICECAP-O scores for carers living in different types of accommodation ( $p=0.106$ ). No significant difference in ICECAP-O scores were found between carers who could leave the person with dementia overnight and those who could not leave the person being cared for at all ( $p=0.056$ ).

Table 5.4 shows that there were more statistically significant relationships between the domains of the ICECAP-O and the EQ-5D in the support group sample than the Challenge FamCare sample. All domains of the ICECAP-O were significantly related to the anxiety/depression and usual activities domains of the EQ-5D. The independence domain of the ICECAP-O was also significantly related to the mobility and self-care domains of the EQ-5D. Mobility was significantly related to thinking about the future and doing things that make you feel valued. Pain/discomfort was significantly associated with the doing things that make you feel valued and enjoyment and pleasure domains.

## **Discussion**

The findings from this study provide evidence to support the feasibility and construct validity of the ICECAP-O for carers of people with dementia.

Consistent with census findings where 58% of all carers were female and 42% male (The Office of National Statistics, 2002), the majority of participants were female in both samples of carers surveyed, but compared with the census where the most common ages for caring are 50-59, our participants were older, reflecting the fact that dementia predominantly affects older people who are typically cared for by spouses of a similar age. Challenge FamCare carers were also different in that they were looking after somebody who exhibited challenging behaviour. Data on whether or not challenging behaviour was experienced by the support group participants was not collected.

A high item completion rate was achieved, indicating a high level of feasibility. Participants completing the support group questionnaire were given a choice between completing the survey online or by post. While offering participants the opportunity to complete questionnaires online provides convenience for researchers there is the potential for selection bias to occur. Problems known to occur with online surveys include: the intended population might not be computer literate, leading to a low response rate; participants might complete the survey more than once; and some people are more likely than others to be willing to complete surveys (Wright, 2005). To a certain extent, surveys conducted by post will also suffer from self-selection bias. The mean age of support group participants was 74.13 (s.d.  $\pm 6.76$ ) and 70% chose to complete the survey online. As yet, the ICECAP-O has not been validated for online completion so the response rate obtained from older carers in this study suggests that this population is reasonably computer literate and it is feasible to complete the ICECAP-O online. This has implications for future research, as it indicates that online data collection for older carers is possible, offering a potentially cheaper way of collecting data than researchers conducting face to face interviews.

Both ceiling and floor effects were found in the items of the ICECAP-O. Unsurprisingly, floor effects were found for the item 'thinking about the future without concern', with 38% of Challenge FamCare participants and 20% of support group participants reporting the lowest level of capability for the item. The coping strategy of 'taking one day at a time' has been identified in dementia caring literature (Nolan, Ingram & Watson, 2002; Ekwall, Sivberg & Hallberg, 2007), and not being able to think about the future without concern would be consistent with a 'taking one day at a time' strategy. Ceiling effects were present for the items 'love and friendship', 'doing things that make you feel valued' and 'independence'. The high degree of capability reported for the items 'love and friendship' and 'doing things that make you feel valued' support the findings of Quinn et al. that dementia carers often perceived their new role to be a rewarding experience (Quinn, Clare & Woods, 2009). The ceiling effect for the item 'independence' was unexpected as almost half of current carers in this survey reported that they could only leave the care recipient alone up to an hour or two or not at all. The presence of floor and ceiling effects indicates that the ICECAP-O would not be able to detect changes for a large proportion of carers of people with dementia. It is worth noting that the current gold standard instrument for quality of life measurement in the United Kingdom, the EQ-5D, suffers from ceiling

effects when used with certain populations (Brazier, Roberts, Tsuchiya & Busschbach, 2004; Hounsome, Orrell & Edwards, 2011). To overcome this issue, the creators of the EQ-5D have developed a 5 level response system as an alternative to the popular 3 level system. The ICECAP-O currently has 4 response levels, so any benefit gained from introducing extra levels to eliminate floor and ceiling effects would need to outweigh the additional workload involved in updating the instrument.

Internal consistency of the ICECAP-O was within acceptable limits. Scales with ten or fewer items typically have a low Cronbach's alpha but this was not the case for the five item ICECAP-O.

The relationship between capability, as measure by the ICECAP-O, and health, as measured by the EQ-5D and GHQ-12 was weak to moderate. The direction of the relationship was as expected; higher levels of capability were associated with higher levels of physical health and mental health. Previous research assessing the use of the ICECAP-O in a falls prevention clinical setting found a moderate relationship between the ICECAP-O and EQ-5D and determined that they were complements rather than substitutes (Davis, Liu-Ambrose, Richardson & Bryan, 2012). The original validation of the ICECAP-O for members of the general population aged 65+ found a strong association between the ICECAP-O and general health for all items apart from 'love and friendship' (Coast et al., 2008c).

The Chi-squared test of association showed mixed results across the two samples. The only significant findings that were consistent across the two samples was the relationship between the EQ-5D domain of usual activities and the ICECAP-O domains of love and friendship and doing things that make you feel valued; and between the EQ-5D domain of anxiety/depression and the ICECAP-O domain of thinking about the future. The Couzner et al. (2012) study presented a Chi-squared test of associations between the domains of the ICECAP-O and the EQ-5D. The only finding consistent with my study is the association between the usual activities domain of the EQ-5D and the doing things that make you feel valued domain of the ICECAP-O.

Both samples of carers in this study had a lower ICECAP-O score than the mean ICECAP-O score of 0.81 (s.d.  $\pm 0.15$ ) for members of the general population aged 65 and over

(Coast et al., 2008b). Carers were enrolled in the Challenge FamCare trial if they were looking after a person with dementia who exhibited challenging behaviours, so the lower than average ICECAP-O score could be an indication of the burden of caring. While the support group carers had a mean EQ-5D score close to the general population norm of 0.78 (s.d.  $\pm 0.26$ ), Challenge FamCare carers had a very low mean EQ-5D of 0.47 (s.d.  $\pm 0.32$ ). This suggests that the EQ-5D was better at picking up on the burden of caring experienced by Challenge FamCare carers than the ICECAP-O. Contributing to the low EQ-5D mean, Challenge FamCare carers reported 'moderate' problems the most frequently across all domains of the EQ-5D, and a third of carers reported being unable to do their usual activities at all. Carers in Challenge FamCare reported being able to leave the person being cared for on their own the least amount of time, which would affect their ability to undertake their usual activities and could explain the low mean EQ-5D scores. Finding out which aspects of caring are compromising or enhancing carer quality of life is important as it will enable health and social care provision to be tailored to meet the specific needs of carers.

Correlation between the ICECAP-O and EQ-VAS was weak (Spearman's  $\rho = 0.209$ ), indicating that carers' level of capability bore little relationship to their self-rated health status. Previous work has found that the EQ-VAS does not always correspond well to the health status scores obtained by the EQ-5D (Feng, Parkin & Devlin, 2012), therefore it is not surprising that the EQ-VAS did not correlate well to the ICECAP-O score in this study.

The expected direction of relationship between the ICECAP-O and the SSCQ, QCGR, RSS and HADS were observed but the strength of these relationships was weak to moderate. The anticipated relationship between the ICECAP-O and age was not observed. This contrasts with the findings of Coast and colleagues (2008c), who found a highly significant relationship between age and the ICECAP items 'doing things that make you feel valued' and 'independence'. No significant differences in ICECAP-O scores were observed for the socio-demographic variables of gender and carer accommodation type in this study. These findings indicate moderate construct validity, with several expected relationships being observed in the data.

## **Limitations**

We did not reach the sample size of 384 required for a statistical comparison. However, this chapter presents early descriptive data from two samples which supports a trend for construct validity of the ICECAP-O for use with carers of people with dementia. A larger study sample would be needed to confirm construct validity.

One of the key differences between the participants in the two data sets was the fact that all Challenge FamCare carers were current carers, whereas the SurveyMonkey questionnaire included responses from people who were previously carers but the care recipient was now deceased or had moved into long-term residential care. We also were limited by the different variables collected in the two data sources. Challenge FamCare included more variables, while for reasons of practicality the support group questionnaire was restricted to socio-demographic information, the ICECAP-O and EQ-5D. The support group questionnaire did not include a question about whether the care recipients exhibited challenging behaviours; therefore it was not possible to determine how comparable the carers in the two data sets were on this attribute.

We were not able to test reliability of the ICECAP-O for carers of people with dementia through test-retest consistency as data were cross-sectional rather than longitudinal. Future evaluations of construct validity would benefit from collecting data at more than one time point to observe the stability of ICECAP-O scores over time.

## **Future work**

To be able to confirm construct validity, a sample size of 384 would be required and any future work should aim to meet this requirement.

The ICECAP is available in two versions; the ICECAP-A for members of the general population aged 18+ and the ICECAP-O for members of the general population aged 65+. The ICECAP-O was used in this chapter. The 2001 UK census found the most common age category for carers to be 50-59 (The Office of National Statistics, 2002) so it is not unreasonable to use an instrument designed for people aged 65+; however, future work investigating the validity of the ICECAP instrument with carers of people with dementia would benefit from using the ICECAP-A as it would include all adults aged 18+.

It would also be interesting to explore the validity of the other two capability based instruments mentioned in the introduction, the ASCOT and the Carer Experience Scale, to allow for a critical comparison between capability based instruments with respect to their suitability for research involving carers of people with dementia.

### **Conclusion**

Use of the ICECAP-O in research involving carers of people with dementia offers the opportunity to explore a broader quality of life than physical health alone. The carers in this study had poorer physical health and capability levels than members of the general population of a similar age. Carers reported that they could not leave the person being cared for alone for a long period time, which hindered their usual activities and could explain the impact on health status scores. Identifying the aspects of quality of life that are affected by caring for a person with dementia will go some way toward shaping support services for this population. In summary, it is concluded that the ICECAP-O has demonstrated good feasibility and construct validity for use with carers of people with dementia.



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Chapter 6: Cost-effectiveness analysis of a trial of joint-remembrance therapy  
for people with dementia and their carers: the effect of choice of quality of  
life measure

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## **Abstract**

### *Background*

This chapter describes the results of a methodological bolt on study to the HTA funded REMCARE trial of joint reminiscence therapy (RT) for people with dementia and their carers (ISRCTN42430123, HTA 06/304/229). The bolt-on study investigated the use of the ICECAP-O for carers.

### *Methods*

The ICECAP-O questionnaire was administered, as an additional measure, to a sample of carers from the REMCARE trial. REMCARE was a multi-centre randomised controlled trial of RT for people with dementia and their carers, compared to usual care. Assessments were carried out by blinded researchers at baseline, 3 months and 10 months. The primary outcome measures for effectiveness in REMCARE was the GHQ-28 for carers. Secondary measures included the EQ-5D, Quality of Care-giving relationship (QCGR), Hospital Anxiety and Depression Scale (HADS) and the Relatives' Stress Scale (RSS).

### *Results*

In the bolt on study, 41 carers (19 intervention, 22 control) completed the ICECAP-O. EQ-5D values for the bolt on study sample were slightly higher than the REMCARE sample at baseline. Carers in the intervention group had a significant increase in social distress on the RSS sub scale (mean difference 1.47, 95% CI 0.45 to 2.49) at 10 months. Carers in the control group had a significant decrease in relationship warmth on the QCGR (mean difference -1.86, 95% CI -3.61 to -0.10).

The mean difference in ICECAP-O scores between groups was -0.02 (bootstrapped 95% CI of -0.105 to 0.066) at 10 months (the intervention group being lower than the control group) and the mean difference in costs was £1,464 (bootstrapped 95% CI of £758 to £2,313), which would result in RT being dominated by usual care. The cost-effectiveness acceptability curve indicated a 2% probability that RT was cost-effective at an arbitrarily chosen willingness to pay threshold of £20,000 per point improvement and an 8% probability that RT was cost-effective at a threshold of £30,000 per point improvement on the ICECAP-O.

The mean difference in QALYs between groups was 0.06 (bootstrapped 95% CI of -0.017 to 0.182), leading to an ICER of £24,400 (53% probability of RT being cost-effective at a maximum willingness to pay threshold of £20,000 per QALY gained and a 71% probability of RT being cost-effective at a maximum threshold of £30,000 per QALY gained).

### *Conclusions*

The bolt-on study did not find any evidence of effectiveness or cost-effectiveness for RT compared to usual care. Cost-effectiveness planes demonstrated the probability of the intervention being cost-effective at different levels of willingness to pay, and hence demonstrated the uncertainty surrounding the ICER point estimates. Cost-effectiveness analysis demonstrated that the control condition was more effective (although this was not statistically significant) and less costly than the intervention when the ICECAP-O was the unit of effect. At present no threshold for societal willingness to pay for a point improvement on the ICECAP-O has been established. When the QALY was the unit of effect the ICER point estimate was £24,400 (53% probability of RT being cost-effective at a threshold of £20,000 per QALY gained and a 71% probability of RT being cost-effective at a threshold of £30,000 per QALY gained). The findings from the sub group of REMCARE carers who completed the ICECAP-O support the findings from the main REMCARE trial, that there was no evidence of effectiveness or cost-effectiveness for RT compared to usual care.

## **Introduction**

### *Reminiscence therapy*

There are two main approaches to interventions involving people with dementia; pharmacological and psychosocial. Pharmacological interventions involve drug regimens such as donepezil, galantamine, rivastigmine and memantine. Psychosocial interventions involve therapeutic techniques that aim to address the psychological and social aspects of an individual's situation. While pharmacological interventions primarily target the person with dementia, psychosocial interventions can be targeted at either the person with dementia, their carer or the dyad together. Reminiscence therapy (RT) is a popular form of psychosocial intervention for people with dementia (Woods, Spector, Jones, Orrell, & Davies, 2005). During RT participants recall their life experiences, typically in a group setting. Objects such as photographs, music recordings and props can be used to initiate discussions and trigger memories (Schweitzer & Bruce, 2008). The aim of RT is to increase psychological well-being and improve social functioning.

Evidence for the effectiveness of RT is at present inconclusive. A meta-analysis of 15 studies found no significant differences in life satisfaction or self esteem between people with dementia who received RT and the control group; however, the intervention group had a higher level of happiness and lower depression (Chin, 2007). The Woods et al. (2005) Cochrane review found only five studies involving RT. Extractable data was available for 144 participants. None of the included trials included rigorous economic evaluations but significant effects for cognition, mood and general behaviour in the person with dementia were reported. Only one of the studies included carer data (Thorgrimsen, Schweitzer, & Orrell, 2002); carers in the intervention arm reported significantly lower levels of strain after receiving RT. An additional two randomised controlled trials involving RT were identified in a recent review (Cotelli, Manenti, & Zanetti, 2012); one assessed whether the order that a person with dementia received RT and drug regimen made a difference, and the other assessed RT compared to reality orientation therapy. Outcome measures for carers were not collected in either study.

### *Aim of the chapter*

This chapter is split into three parts. Part one briefly describes the REMCARE randomised controlled trial of joint-RT compared to usual care (ISRCTN42430123, HTA 06/304/229). Full details and results are published in Woods et al., 2012. Part two describes the bolt on

study involving a sub sample of carers who also completed the ICECAP-O capability measure. Part three compares findings from both studies and relates them to results from other studies. As discussed in Chapter 3, the choice of outcome measure used in trials is important as instruments have different strengths and weaknesses; generic instruments can be used for a wide range of illnesses but lack sensitivity, whereas condition-specific instruments are sensitive to the population being measured but study results are not easily comparable to studies of different illnesses. In the United Kingdom the National Institute for Health and Care Excellence (NICE) assesses the evidence of effectiveness and cost-effectiveness for new and existing interventions, and their preferred instrument for measuring quality of life is the generic EQ-5D (EuroQoL Group, 1990), as data can easily be converted into quality-adjusted life years (QALYs) (NICE, 2013). For carers, four out of the five EQ-5D domains are concerned with physical health (mobility, self-care, usual activities, pain/discomfort), with only one domain concerned with mental health (anxiety/depression). An intervention such as RT is unlikely to have an effect on physical health of carers so there is a need to consider alternative measures of well-being. Measuring capability using the ICECAP-O is a novel approach to well-being measurement and to our knowledge no previous studies involving carers of people with dementia have used the ICECAP-O. The aim of this chapter is to explore the extent to which the ICECAP-O can capture the effects of joint RT for carers of people with dementia, and compare the findings to the results of the main REMCARE study.

### **Part 1: The REMCARE trial**

Full details of the REMCARE trial design and results are in Woods et al., 2012. The following section presents an overview of the trial to put the findings from the ICECAP-O bolt on study in context.

### **Methods**

#### *Design*

The study was a single-blind randomised controlled trial with two parallel arms- an intervention arm and usual care arm. Allocation was on a 1:1 basis, restricted to ensure a minimum of 8 dyads per intervention group and a maximum of 12 dyads. Assessments were carried out in the participant's home by a blinded researcher at baseline, 3 months and 10 months.

### *Participants*

Participants with dementia met the following inclusion criteria:

- Met the DSM-IV criteria for dementia at baseline.
- Were in the mild to moderate stage according to the Clinical Dementia Rating.
- Were able to communicate and understand communication, rated by the Clifton Assessment Procedures for the Elderly- Behaviour Rating Scale.
- Could engage with group activity.
- Lived in the community at baseline and had a carer willing and able to participate in the intervention.

People with severe dementia were excluded, as were individuals unable to engage in group activity. All participants provided signed informed consent before they were randomised. There were no exclusion criteria relating to carers.

### *Recruitment strategy*

Participants with dementia were recruited through a range of mental health services, in particular memory clinics and community mental health teams for older people. Recruitment took place between May 2008 and July 2010.

### *Ethical approval*

Ethical approval was granted by the Multi-centre Research Ethics Committee (MREC) for Wales (ref no. 07/MRE09/58). Each site received approval from their Local Research Ethics Committee (LREC) and the appropriate NHS Trust Research and Development department.

### *Intervention*

Eligible participants were randomised to receive either joint RT or usual care. Usual care consisted of the health, social services and voluntary services available to participants in their local area. Participants randomised to the intervention group attended 12 weekly sessions of joint RT with their carers, followed by monthly maintenance sessions for a further 7 months. The joint RT followed the 'Remembering Yesterday, Caring Today' model (Schweitzer & Bruce, 2008). Topics covered in the reminiscence sessions included introductions; childhood and family life; school days; starting work; going out; courting

and marriage; homes, gardens and animals; food and cooking; babies and children; holidays and journeys; festivals and special days and a final evaluation session. Groups were run in a community setting by two trained facilitators, aided by volunteers.

#### *Carer outcome measures*

A number of measures for both the person with dementia and their carer were included. This chapter reports on the following carer outcome measures: the EuroQoL (EQ-5D and EQ-VAS; EuroQoL Group, 1990), Quality of Care-giving relationship (QCGR; Spruytte, Van Audenhove, Lammertyn, & Storms, 2002), Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983), Relatives Stress Scale (RSS; Greene, Smith, Gardiner, & Timbury, 1982) and General Health Questionnaire-28 (GHQ-28; Goldberg, 1978).

The EQ-5D measures health-related quality of life and consists of two parts; a self-report questionnaire (EQ-5D) and a visual analogue scale (EQ-VAS). The five domains of the EQ-5D are mobility, self care, usual activities, pain and discomfort, and anxiety and depression. Each domain has three levels (no problems, some problems, extreme problems). Responses are converted into an index score of between -0.594 and 1 using an established algorithm (MVH Group, 1999), with 0 representing dead and 1 representing the best possible health state. Preference based tariffs for the different states were elicited using the time trade-off method with a selection of the UK population, enabling comparisons to be made between the EQ-5D index scores obtained in studies and population norms (Kind et al., 1999). On the EQ-VAS, carers are asked to draw a line on a thermometer to mark their quality of life, with 0 representing the worst imaginable health state and 100 the best imaginable health state.

The QCGR has two sub-scales, relationship warmth and relationship negativity. Carers rate their agreement with 14 statements on a 5 level scale from totally disagree to totally agree. Responses are summed to 8-40 for warmth (higher scores indicating higher warmth) and 6-30 for negativity (higher scores indicating absence of negativity).

The HADS asks carers to rate the frequency of 14 statements relating to anxiety and depression. Each statement has 4 levels of response which are then summed to produce separate scores for anxiety and depression of between 0-21, higher scores indicating worse symptoms.

The RSS is a 15 item questionnaire which measures the reaction of carers to the care recipients. There are three sub-scales; emotional distress in relation to the care recipient, social distress as a result of caring, and negative feelings toward the care recipient. Domains are rated on a 5 level scale. The RSS is summed to a total score of between 0-60, with 0 representing no problems and 60 the highest level of problems on every item.

The GHQ-28 assesses the carer's current mental well-being and how it differs from their usual state. Symptoms are scored as 1 if present and 0 if missing, resulting in a total score of between 0-28 with 28 representing the worst state.

#### *Measurement of costs*

An NHS (including costs incurred using dementia services, primary and secondary care) and social services sector perspective was adopted. No discounting was applied to costs or outcomes as the follow-up period was less than a year.

A micro costing of the reminiscence therapy intervention was undertaken costing REMCARE staff, materials and venues.

An adapted Client Service Receipt Inventory (CSRI) was used to retrospectively collect information about the use of health and social care services (Beecham & Knapp, 2001; Ridyard & Hughes, 2010). National unit costs for resource use were obtained from the Department of Health (Department of Health, 2010c) and Curtis (2011) for the year 2010.

#### *Missing data*

For items missing within a measure, the rule for completing missing data for the relevant measure were applied e.g. for the HADS a missing item is replaced by the mean of the remaining items. For measures which were entirely missing at a time point, a linear regression model was applied, taking into account the following variables: age, gender, spousal care, centre, wave and other baseline scores. For imputing missing scores at first follow-up, baseline values were used in the model to predict the missing score. For imputing missing scores at second follow-up a combination of baseline and first follow-up scores were used to predict the missing score.



Briggs et al. (2003) discusses different approaches to handling missing service use data and while imputation to replace missing data with statistical estimates is considered more favourably, the authors note that simpler methods such as complete case analysis may be acceptable in data sets with a low amount of missing data. In the REMCARE dataset, service use data were available for 336 out of the 350 dyads who completed the trial (96%) and a complete case analysis approach was used.

#### *Data analysis*

A linear-mixed model analysis of covariance (ANCOVA) using baseline values as covariates was used for the effectiveness analysis. The treatment group was a fixed factor and the centre was a random factor.

For the health economics analysis, a cost-effectiveness analysis using the GHQ-28 as the measure of effectiveness for carers was undertaken. Non-parametric bootstrapping with 1,000 replications was used. Bootstrapping estimates the distribution of a statistic through repeated sampling with replacement. Bootstrapping is used to address the uncertainty associated with point estimates of cost-effectiveness ratios (Briggs et al., 1997). A secondary cost utility estimating the cost per QALY was done using the EQ-5D area under the curve method to calculate QALYs.

A cost-effectiveness plane displaying the distribution of the 1,000 bootstrapped incremental cost-effectiveness ratio (ICER) plots was generated. A cost-effectiveness acceptability curve (CEAC) was constructed to illustrate the uncertainty surrounding the cost-effectiveness estimate and depict the probability that the intervention is cost-effective compared to usual care given different assumptions about maximum willingness to pay for outcomes (Fenwick & Byford, 2005). CEACs offer a more flexible approach to representing uncertainty around ICER point estimates than estimating confidence intervals since they show the probability that an intervention is cost-effective for a range of potential maximum willingness to pay values (Brazier et al., 2007).

## **Results**

### *Effectiveness findings relating to carers*

2908 dyads were referred to the REMCARE trial and 488 were randomised. The majority of exclusions between referral and randomisation occurred due to potential participants not

wishing to take part (36%) or researchers being unable to make contact (16%). Of the 488 randomised, 350 completed the 10 month assessment.

This thesis focuses on carer quality of life measurement so Chapter 6 does not report on the results of the REMCARE trial for people with dementia in detail; however, no significant effects were detected on the primary outcome measure for people with dementia, the QOL-AD. No significant differences were detected between carers in the intervention group and control group on the GHQ-28, EQ-5D, RSS, HADS or QCGR at any time point. Carers allocated to the intervention group reported a significant increase in anxiety on the GHQ-28 anxiety sub scale at 10 months (mean difference 1.25, SE 0.5;  $F=8.28$ ;  $p=0.04$ ). The mean carer QALY at 10 months for the intervention group was 0.632 and the mean carer QALY for the control group was 0.633, a negligible difference.

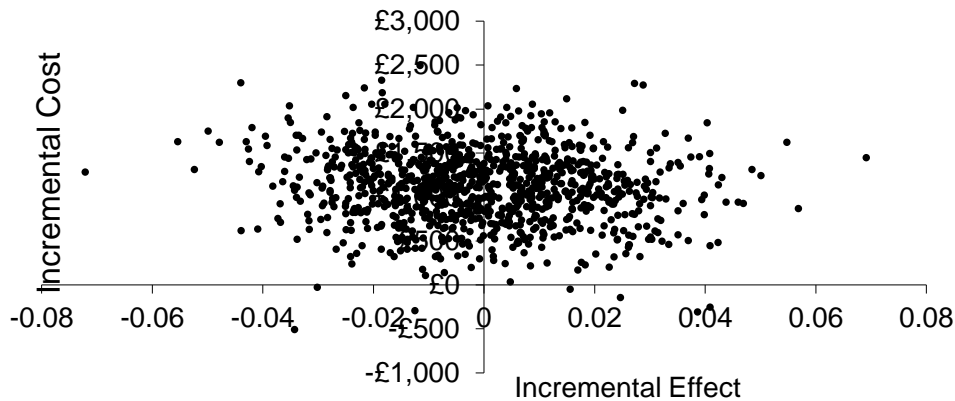
#### *Resource use and associated costs*

Researchers estimated the mean cost of running the joint RT sessions as £964 per dyad, based on 10 dyads attending each session. The largest proportion of cost was staffing, which accounted for approximately two-thirds of the cost. Transport for participants was the next highest cost category. Venue costs were modest and in some instances venues were provided for free. Mean cost per carer was £2,495 (s.d.  $\pm$ £3,866) in the intervention group and £1,359 (s.d.  $\pm$ £3,743) in the control group, a mean difference of £1,136 (bootstrapped 95% CI of £322 to £1,941).

#### *Cost-effectiveness findings relating to carers*

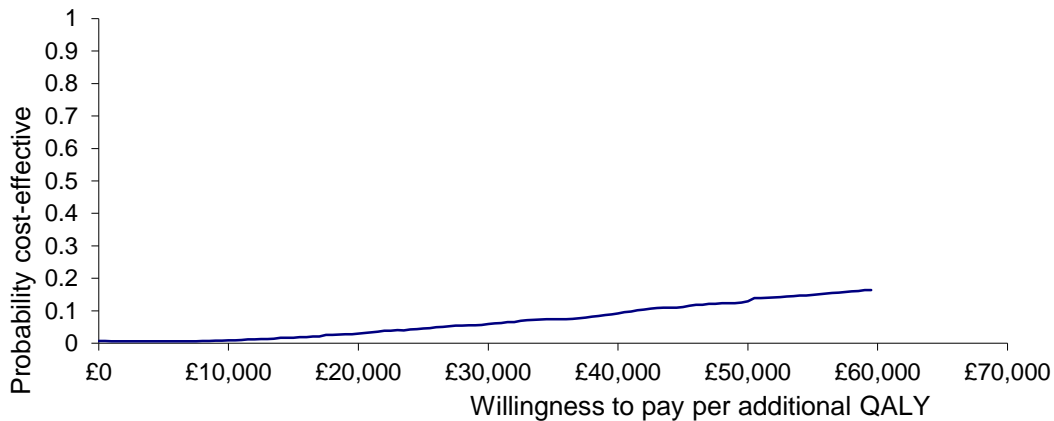
Of the 350 dyads completing the 10 month trial, 336 were used for the economic analysis. The difference in cost between groups at 10 months was estimated as £1,136 (bootstrapped 95% CI £322 to £1,941) and the difference in QALYs was estimated as -0.0004344 (bootstrapped 95% CI of -0.0352 to 0.0365). The intervention is dominated by the control condition. Figure 6.1 shows the cost-effectiveness plane for the QALY with 1,000 bootstrapped estimates. 52% of the plots fell in the North West quadrant, indicating that the intervention is more costly and less effective than the control condition.

**Figure 6.1:** Cost-effectiveness plane for the QALY with 1,000 bootstrapped ICER estimates



The cost-effectiveness acceptability curve in Figure 6.2 shows the probability that the intervention is cost-effective at a range of cost per QALY thresholds. There was a 3% probability that the intervention was cost-effective at a willingness to pay threshold of £20,000 per QALY gained and a 6% probability that the intervention was cost-effective at the £30,000 per QALY gained threshold.

**Figure 6.2:** Cost-effectiveness acceptability curve for the QALY



## Conclusion

The REMCARE trial took a traditional approach to measuring cost-effectiveness in terms of cost per change in the primary outcome measure (for carers this was the GHQ-28), with a secondary cost-utility analysis using QALYs with weights calculated using the EQ-5D. The trial and concurrent economic evaluation led to the conclusion that there was no evidence for effectiveness or cost-effectiveness of RT using the selected outcome measures. Indeed, there was some increased carer anxiety observed in the intervention group.

## **Part 2: The ICECAP-O methodological bolt on study**

Part 2 of this chapter describes a bolt on study conducted towards the end of the REMCARE trial. The bolt on study was conceived after the main trial had begun recruiting. While eight centres were involved in the main trial (with recruitment running May 2008- July 2010), this analysis includes carers from the Bangor and South London centres as these were the only centres still recruiting when the bolt on study began in late 2009. Carers in these centres were asked to complete an additional instrument, the ICECAP-O, at all time-points to see whether the use of a capability based measure embracing a broader concept of quality of life than physical functioning alone might produce a different conclusion of the effectiveness and cost-effectiveness of RT compared to usual care. ICER point estimates are presented for this sub sample.

### **Methods**

#### *Ethical approval*

The inclusion of the ICECAP-O as an outcome measure for carers was approved by the MREC in November 2009.

#### *Outcome measures*

The primary outcome measure for this bolt on study was the ICECAP-O, an instrument designed to capture the level of capability of individuals aged 65+ (Coast, Peters, Natarajan, Sproston, & Flynn, 2008b). Carers were asked to rate their capability in five domains (attachment, security, role, enjoyment and control) on a four point scale. Results are converted into an index score of between 0 and 1, with 0 representing no capability and 1 full capability. The algorithm to calculate the index score was developed using best-worst scaling with a sample of the UK population aged 65+ (Coast et al., 2008b). Best-worst scaling asks individuals to select the best and worst option in a given set of samples (Finn and Louviere, 1992), revealing the values placed on different options without asking people to trade one thing for another.

#### *Data analysis*

SPSS version 16.0 was used for data analysis. Variables were checked for out of range values before any analysis was undertaken. Data normality was checked using the Kolmogorov-Smirnov statistic (a p-value of >0.05 indicating normality), visual inspection of histograms (a bell shape indicating normality) and examining detrended normal Q-Q

plots (no clustering, and points collecting around the zero line suggesting normality) as recommended in Field, 2009. Differences in costs and outcomes at baseline were not adjusted for at follow-up.

The primary analysis was the estimation of the cost-effectiveness of joint RT compared to usual care at 10 months using the ICECAP-O as the effectiveness measure. Confidence intervals around differences in costs and outcomes were estimated using non parametric bootstrapping (1000 replications were run). Cost-effectiveness analysis was repeated for the secondary outcome measures. A cost-utility analysis using QALYs calculated by the area under the curve method was undertaken. Cost-effectiveness planes and cost-effectiveness acceptability curves are presented.

Two pre planned exploratory subgroup analyses were conducted; effect of fidelity was investigated for carers who attended an arbitrarily chosen cut-off of least 6 out of the 12 weekly sessions, and effect of age was investigated for carers aged 65+ as this is the intended respondent age for the ICECAP-O.

#### *Missing data*

No service use data were missing for the ICECAP-O sample so no imputation was necessary.

## **Results**

Forty-one carers (19 intervention, 22 control) completed the ICECAP-O at three time-points and were included in this analysis. The flow of participants is shown in Figure 6.3.

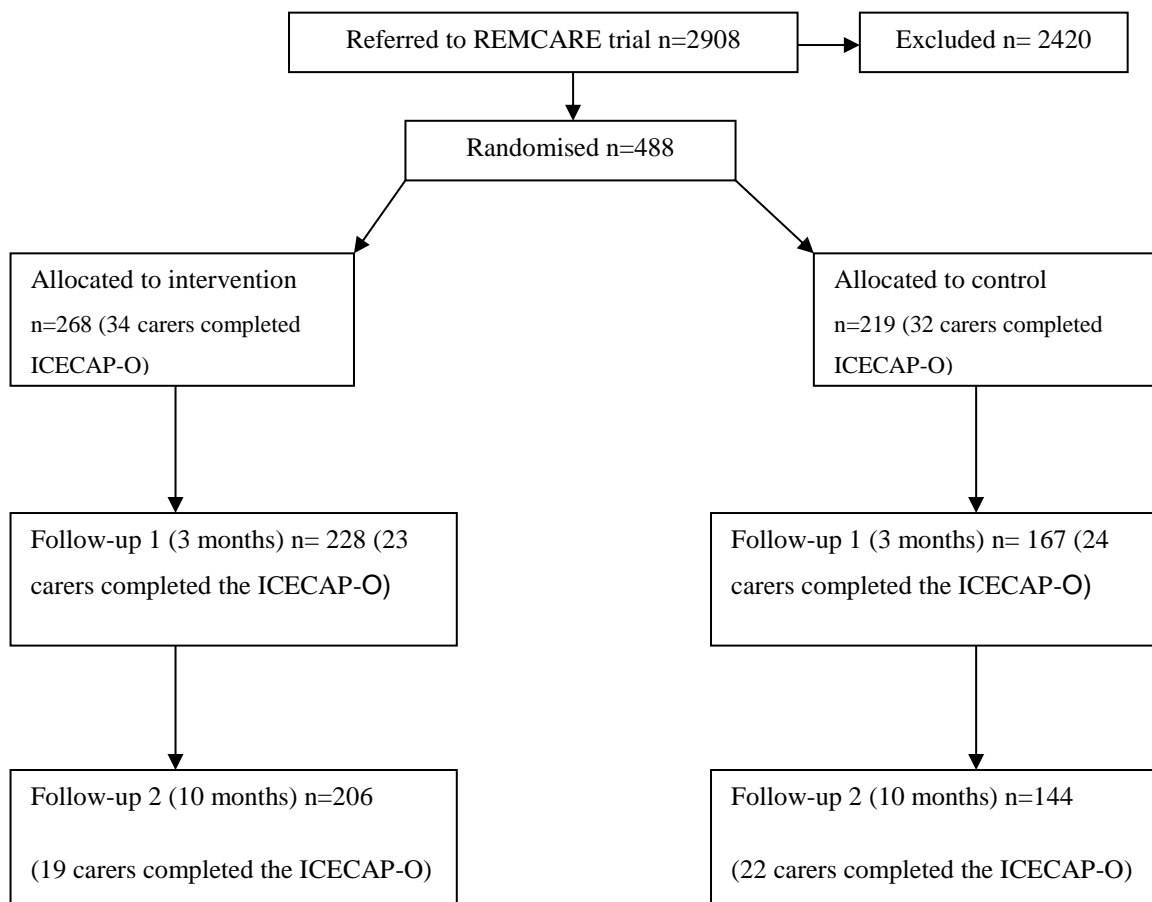
#### *Representativeness of sample to main REMCARE economic analysis sample*

Baseline demographics and service use data were used to assess whether the 41 participants used for this analysis were representative of the 336 participants in the REMCARE economic analysis sample. Table 6.1 shows baseline values for the whole REMCARE sample and the sub sample of carers used in this bolt-on study. Mann-Whitney U tests were used to detect significant differences between the ICECAP-O sample and main economic analysis sample. No significant differences were found on carer age, person with dementia age, the number of adults living in the house, the number of hours

the person with dementia could be left alone or the number of hours spent by the carer on caring tasks.

At baseline, the mean EQ-5D index value for carers in the intervention group of the main REMCARE study was 0.78 (s.d.  $\pm 0.23$ ) and was 0.77 (s.d.  $\pm 0.24$ ) in the control group. In the ICECAP-O sub sample, mean EQ-5D index values at baseline were slightly higher. The mean EQ-5D index score at baseline was 0.82 (s.d.  $\pm 0.12$ ) in the intervention group and 0.79 (s.d.  $\pm 0.23$ ) in the control group (see Table 6.3 for carer outcome measures at baseline). GHQ-28 scores were slightly lower in the ICECAP-O sample compared to the main sample, indicating a better mental health in the sub sample. We believe the 41 carers used in the ICECAP-O sub sample to be representative of the 336 carers used in the main REMCARE economic analysis sample, but cannot guarantee this as there may be unobserved differences in outcomes not measured.

**Figure 6.3:** Flow of participants through the REMCARE trial



### *Effectiveness findings relating to carers*

Baseline demographics show that the intervention group and control group were similar on key variables (Table 6.1). Inspection of the Kolmogorov-Smirnov test (Table 6.2) indicated that most carer outcome measures were normally distributed, with the exception of the ICECAP-O for the control group, the EQ-VAS for the intervention group and the HADS depression scale for both groups. Histograms and detrended normal Q-Q plots are presented in Appendix 13, page 211. Visual inspection of the histograms for the non-normally distributed measures showed that the ICECAP-O and EQ-VAS were negatively skewed, while the HADS depression scale was negatively skewed. Parametric t-tests were used for all normally distributed outcome measures. Non-parametric Mann Whitney U tests were used with the non-normally distributed ICECAP-O, EQ-VAS and HADS depression sub scale.

**Table 6.1:** Baseline demographic information

	Intervention (n=19)	Control (n=22)	All sub sample (n=41)	All REMCARE sample (n=488)
Female participant	8 (42.1%)	7 (31.8%)	15 (36.6%)	240 (49.5%) <sup>1</sup>
Female carer	14 (73.7%)	19 (86.3%)	26 (63.4%)	325 (67.0%) <sup>1</sup>
Participant ethnicity: White	19 (100%)	22 (100%)	41 (100%)	447 (95.3%) <sup>2</sup>
Carer ethnicity: White	19 (100%)	22 (100%)	41 (100%)	448 (95.9%) <sup>3</sup>
Participant and carer live together	17 (89.5%)	19 (86.4%)	36 (87.8%)	362 (74.5%) <sup>4</sup>
Dyad relationship:				
Spousal	16 (84.2%)	16 (72.7%)	32 (78.0%)	345 (70.7%)
Non spousal	3 (15.8%)	6 (27.3%)	9 (22.0%)	143 (29.3%)
Type of dementia:				
Alzheimer's disease	8 (42.1%)	15 (68.2%)	23 (56.1%)	106 (57.9%) <sup>5</sup>
Vascular	7 (36.8%)	1 (4.5%)	8 (19.5%)	23 (12.6%) <sup>5</sup>
Mixed	1 (5.3%)	1 (4.5%)	2 (4.9%)	17 (9.3%) <sup>5</sup>
Unknown/ Missing	3 (15.8%)	5 (22.7%)	8 (19.5%)	37 (20.2%) <sup>5</sup>
	Mean (s.d.), min-max	Mean (s.d.), min-max	Mean (s.d.), min-max	Mean (s.d.), min-max
Age of participant	79.84 (7.22), 65-93	77.91 (8.89), 54-91	78.80 (8.12), 54-93	77.53 (7.3), 54-95 <sup>6</sup>
Age of carer	72.00 (11.25), 45-83	68.91 (8.61), 47-86	70.34 (9.91), 45-86	69.65 (11.6), 23-91 <sup>4</sup>
Cost of carer's service use in the 3 months before baseline	£466.18 (£1,116.89), £0- £4,629.97	£109.33 (£226.70), £0- £1,069.00	£274.70 (£787.90), £0- £4,629.97	Missing <sup>7</sup>

Notes:

<sup>1</sup>Percentages are based on n= 485, <sup>2</sup>percentages are based on n=469, <sup>3</sup>percentages are based on n=467, <sup>4</sup>percentages are based on n=486, <sup>5</sup>percentages are based on n=183, <sup>6</sup>percentages are based on n=487, <sup>7</sup>Data on baseline costs are not available for the whole REMCARE sample as they were not reported in the HTA report (differences between groups were tested through frequency of service use).



**Table 6.2:** Tests for normality

		Kolmogorov-Smirnov <sup>a</sup>			Shapiro-Wilk		
		Statistic	df	Sig.	Statistic	df	Sig.
ICECAP	Intervention	.196	19	.053	.919	19	.106
	Control	.199	22	.023	.881	22	.013
Carer EQ5D	Intervention	.137	19	.200*	.949	19	.379
	Control	.184	22	.051	.771	22	.000
EQ-VAS	Intervention	.208	19	.029	.851	19	.007
	Control	.161	22	.142	.926	22	.102
HADS-Depression	Intervention	.202	19	.040	.935	19	.215
	Control	.191	22	.036	.905	22	.037
HADS-Anxiety	Intervention	.127	19	.200*	.940	19	.264
	Control	.119	22	.200*	.951	22	.327
QCGR total	Intervention	.149	19	.200*	.931	19	.184
	Control	.107	21	.200*	.965	21	.619
RSS_total	Intervention	.127	19	.200*	.968	19	.735
	Control	.128	21	.200*	.968	21	.679
GHQ_total	Intervention	.168	17	.200*	.911	17	.102
	Control	.159	22	.155	.883	22	.014
Cost	Intervention	.356	19	.000	.464	19	.000
	Control	.315	22	.000	.476	22	.000

\*. This is a lower bound of the true significance.

a. Lilliefors Significance Correction

At baseline, mean age of carer in the ICECAP-O sample was 70.34 (s.d.  $\pm 9.91$ ). As can be seen in Table 6.3, both the control group and intervention group had EQ-5D values higher than the population norm of 0.78 (s.d.  $\pm 0.26$ ) for the age group 65-74. The intervention group had a slightly higher EQ-5D than the control group (mean 0.82, s.d.  $\pm 0.12$  vs. mean 0.79, s.d.  $\pm 0.23$ ). At the three month follow up the EQ-5D score in the intervention group had a non significant increase to a mean of 0.84 (s.d.  $\pm 0.09$ ), whereas the control group EQ-5D mean had decreased to 0.73 (s.d.  $\pm 0.23$ ), which is lower than the general population norm. At the 10 month follow up the intervention group had a mean EQ-5D score of 0.83 (s.d.  $\pm 0.13$ ) compared to the control group mean of 0.76 (s.d.  $\pm 0.26$ ).

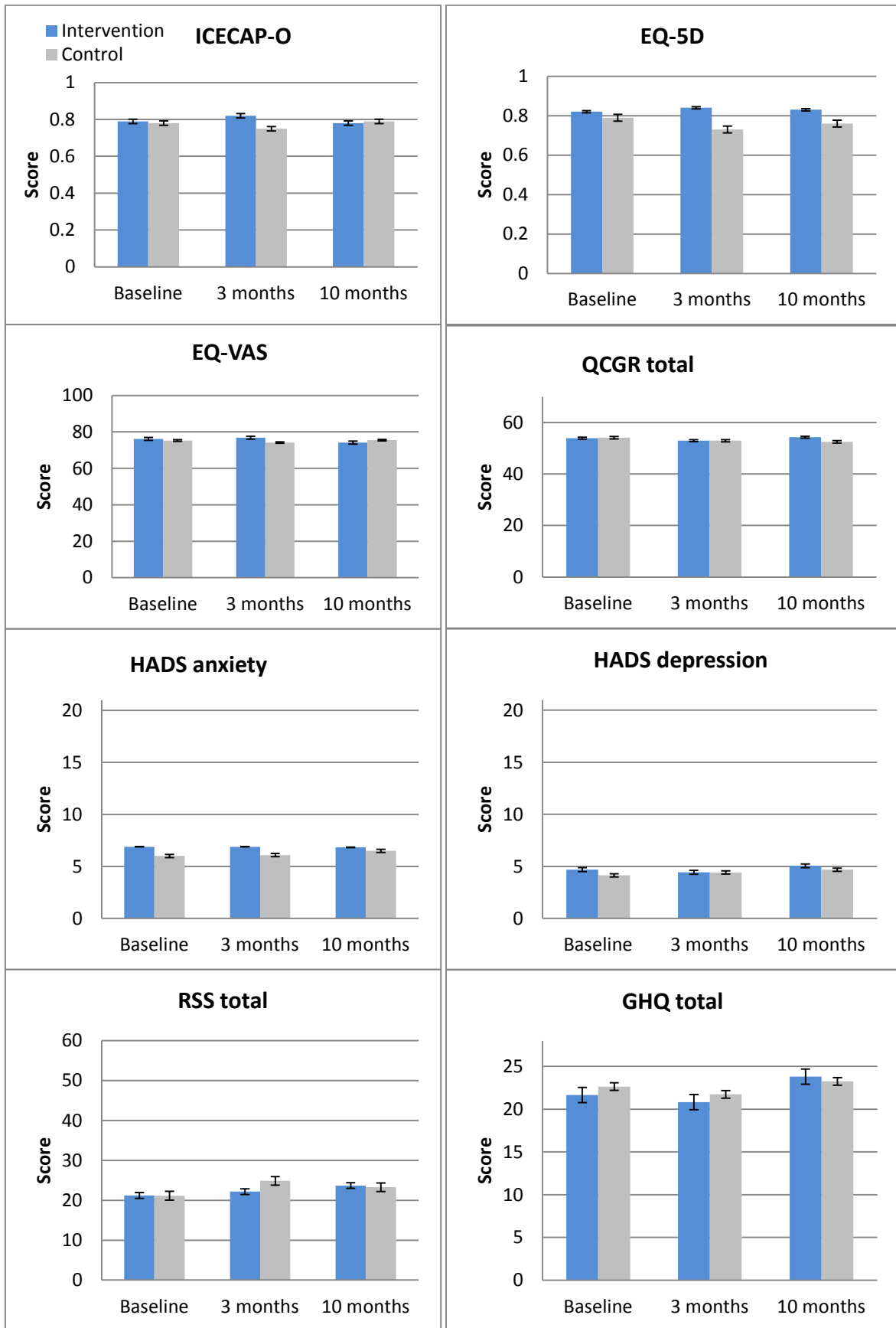
Figure 6.4 shows the mean scores for the intervention and control groups at each of the time points.

**Table 6.3:** Mean treatment group values for each of the carer outcome measures at each of the time points

	Baseline: Mean (s.d.)			3 months: Mean (s.d.)			10 months: Mean (s.d.)		
	Intervention (n=19)	Control (n=22)	Difference (95% CI), p	Intervention (n=19)	Control (n=22)	Difference (95% CI), p	Intervention (n=19)	Control (n=22)	Difference (95% CI), p
ICECAP-O	0.79 (0.12)	0.78 (0.17)	0.01 (-0.07 to 0.10) <sup>a</sup> , 0.95	0.82 (0.12)	0.75 (0.20)	0.07 (-0.02 to 0.17) <sup>a</sup> , 0.26	0.78 (0.12)	0.79 (0.14)	-0.01 (-0.08 to 0.07) <sup>a</sup> , 0.95
EQ-5D index	0.82 (0.12)	0.79 (0.23)	0.03 (-0.15 to 0.09), 0.62	0.84 (0.09)	0.73 (0.24)	0.10 (-0.22 to 0.01), 0.07	0.83 (0.13)	0.76 (0.26)	0.07 (-0.20 to 0.07), 0.32
EQ-VAS	76.16 (14.67)	75.27 (20.15)	0.89 (-9.55 to 11.55) <sup>a</sup> , 0.92	76.84 (15.67)	74.18 (16.83)	2.66 (-6.94 to 11.92) <sup>a</sup> , 0.44	74.11 (14.65)	75.50 (16.11)	-1.40 (-10.88 to 7.70) <sup>a</sup> , 0.90
Quality of Care-giving relationship warmth	33.26 (4.93)	33.43 <sup>1</sup> (4.65)	-0.17 (-2.90 to 3.23), 0.91	31.89 (5.10)	32.35 <sup>2</sup> (4.45)	-0.46 (-2.65 to 3.56), 0.77	33.17 <sup>3</sup> (5.02)	31.57 <sup>1</sup> (6.07)	1.60 (-5.25 to 2.06), 0.38
Quality of Care-giving relationship negativity	20.63 (4.46)	21.10 <sup>1</sup> (4.55)	-0.46 (-2.43 to 3.35), 0.75	21.05 (3.67)	21.10 <sup>1</sup> (4.61)	-0.04 (-2.65 to 2.73), 0.98	21.26 (4.23)	20.90 <sup>1</sup> (4.23)	0.36 (-3.07 to 2.35), 0.79
Quality of Care-giving relationship total	53.89 (8.08)	54.05 (8.66)	-0.15 (-5.17 to 5.47), 0.96	52.95 (7.95)	52.90 <sup>1</sup> (8.89)	0.04 (-5.46 to 5.38), 0.99	54.28 <sup>3</sup> (8.30)	52.48 <sup>1</sup> (9.68)	1.80 (-7.71 to 4.10), 0.54
HADS anxiety sub-scale	6.89 (4.65)	6.00 (3.63)	0.90 (-3.51 to 1.72), 0.49	6.89 (3.93)	6.09 (3.46)	0.80 (-3.14 to 1.53), 0.49	6.84 (3.89)	6.50 (4.23)	0.34 (-2.92 to 2.24), 0.79
HADS depression sub-scale	4.68 (2.89)	4.14 (3.04)	0.55 (-1.11 to 2.44) <sup>a</sup> , 0.57	4.42 (2.55)	4.41 (3.10)	0.01 (-1.60 to 1.66) <sup>a</sup> , 0.97	5.05 (3.10)	4.68 (2.55)	0.37 (-1.43 to 1.99) <sup>a</sup> , 0.70
RSS emotional distress	9.95 (4.37)	9.05 <sup>1</sup> (4.14)	0.90 (-3.62 to 1.82), 0.51	9.89 (4.32)	10.50 (3.86)	-0.61 (-1.98 to 3.19), 0.64	10.11 (4.48)	10.09 (5.07)	0.01 (-3.06 to 3.03), 0.99
RSS social distress	7.16 (3.80)	7.33 <sup>1</sup> (3.77)	-0.18 (-2.25 to 2.60), 0.88	7.84 (3.98)	9.05 (4.05)	-1.20 (-1.34 to 3.75), 0.35	8.63 (4.15)	8.05 (4.02)	0.59 (-3.17 to 2.00), 0.65
RSS negative feelings	4.11 (1.91)	4.64 (1.99)	-0.53 (-0.71 to 1.77), 0.39	4.42 (2.59)	5.32 (2.32)	-0.90 (-0.65 to 2.45), 0.25	4.95 (3.05)	5.14 (2.49)	-0.19 (-1.56 to 1.94), 0.83
RSS total	21.21 (8.63)	21.14 <sup>1</sup> (8.15)	0.07 (-5.44 to 5.30), 0.98	22.16 (9.22)	24.86 (9.19)	-2.71 (-3.12 to 8.54), 0.35	23.68 (10.17)	23.27 (9.94)	0.41 (-6.77 to 5.95), 0.90
GHQ somatic feelings	6.63 (4.22)	5.82 (4.43)	0.81 (-3.56 to 1.93), 0.55	5.95 (3.22)	5.36 (2.85)	0.58 (-2.50 to 1.34), 0.54	6.63 (3.70)	6.19 <sup>1</sup> (3.03)	0.44 (-2.60 to 1.71), 0.68
GHQ anxiety	6.84 (5.27)	6.59 (4.09)	0.25 (-3.21 to 2.71), 0.87	6.28 <sup>3</sup> (3.36)	6.73 (3.24)	-0.45 (-1.67 to 2.57), 0.67	7.59 <sup>4</sup> (4.23)	7.09 (4.09)	0.50 (-3.21 to 2.22), 0.71
GHQ social dysfunction	6.72 <sup>3</sup> (2.68)	7.59 (2.28)	-0.87 (-0.72 to 2.46), 0.28	6.74 (2.75)	7.50 (1.82)	-0.76 (-0.69 to 2.22), 0.30	7.81 <sup>5</sup> (2.97)	8.36 (2.06)	-0.55 (-1.10 to 2.20), 0.50
GHQ depression	1.44 <sup>3</sup> (2.33)	2.64 (3.71)	-1.19 (-0.85 to 3.23), 0.24	1.47 (1.95)	2.14 (2.46)	-0.66 (-0.76 to 2.08), 0.35	1.95 (2.44)	1.76 <sup>1</sup> (2.72)	0.19 (-1.85 to 1.47), 0.82
GHQ-28 total	21.65 <sup>4</sup> (11.64)	22.64 (12.61)	-0.99 (-6.70 to 8.97), 0.80	20.83 <sup>3</sup> (7.99)	21.73 (8.23)	-0.89 (-4.33 to 6.12), 0.73	23.81 <sup>5</sup> (11.66)	23.25 <sup>2</sup> (9.88)	0.56 (-7.86 to 6.73), 0.87

<sup>1</sup>n=21, <sup>2</sup>n=20, <sup>3</sup>n=18, <sup>4</sup>n=17, <sup>5</sup>n=16; <sup>a</sup> bootstrapped 95% confidence interval presented due to non-normal distribution of the data. Note: EQ-VAS = EQ-5D Visual Analogue Scale, RSS = relatives Stress Scale, HADS = Hospital Anxiety and Depression Scale, GHQ = General Health Questionnaire. HADS and GHQ are scored such that a lower score indicates a better outcome

**Figure 6.4:** Mean scores for the carer outcome measures at each time point



Note: HADS and GHQ-28 are scored such that a lower score indicates a better outcome

Table 6.4 shows the mean scores for paired samples, that is, carers who completed the measure at both baseline and final follow-up. Paired samples t-tests were used to detect whether mean scores for either group were significantly different at the 10 month follow-up compared to baseline on all measures apart from the ICECAP-O, EQ-VAS and HADS depression scale. For these non-normally distributed measures I used a related samples Wilcoxon Signed Ranks test. The tests on the outcome data were carried out before bootstrapping. While it is necessary to bootstrap and take account of the non normal distribution of the cost data for the cost-effectiveness analysis (to avoid bias from highly skewed cost data and addressing uncertainty around point estimates of cost-effectiveness ratios; Briggs et al., 1997); it was deemed adequate to use t-tests/related samples Wilcoxon Signed Ranks tests on non bootstrapped outcome data as most variables had been shown to be normally distributed. Bootstrapped confidence intervals are listed for the non-normally distributed measures.

For the intervention group the only significant difference was seen on the social distress subscale of the Relatives Stress Scale, which increased significantly from 7.16 (s.d.  $\pm 3.80$ ) to 8.63 (s.d.  $\pm 4.15$ ) (mean difference 1.47, 95% CI of 0.45 to 2.49) between baseline and 10 months. For the control group the only significant difference was seen on the warmth subscale of the Quality of Care-giving Relationship, which decreased significantly from 33.43 (s.d.  $\pm 4.65$ ) to 31.57 (s.d.  $\pm 6.07$ ) (mean difference -1.86, 95% CI of -3.61 to -0.10) between baseline and 10 months.

**Table 6.4:** Mean treatment group values for paired samples

	Intervention group				Control group			
	Paired n	Baseline Mean (s.d.)	10 month Mean (s.d.)	Mean difference (95% CI), p	Paired n	Baseline Mean (s.d.)	10 month Mean (s.d.)	Mean difference (95% CI), p
ICECAP-O	19	0.79 (0.12)	0.78 (0.12)	-0.01 (-0.06 to 0.08) <sup>a</sup> , 0.55	22	0.78 (0.17)	0.79 (0.14)	0.01 (-0.10 to 0.08) <sup>a</sup> , 0.27
EQ-5D index	19	0.82 (0.12)	0.83 (0.13)	0.01 (-0.07 to 0.09), 0.80	22	0.79 (0.23)	0.76 (0.26)	-0.03 (-0.09 to 0.04), 0.41
EQ-VAS	19	76.16 (14.67)	74.11 (14.65)	-2.05 (-6.63 to 10.53) <sup>a</sup> , 0.86	22	75.27 (20.15)	75.50 (16.11)	0.23 (-11.02 to 10.52) <sup>a</sup> , 0.86
Quality of Care-giving relationship warmth	18	33.61 (4.83)	33.17 (5.02)	-0.44 (-2.46 to 1.57), 0.65	21	33.43 (4.65)	31.57 (6.07)	-1.86 (-3.61 to -0.10), 0.04
Quality of Care-giving relationship negativity	19	20.63 (4.46)	21.26 (4.23)	0.63 (-1.58 to 2.84), 0.56	21	21.10 (4.55)	20.90 (4.23)	-0.19 (-2.01 to 1.63), 0.83
Quality of Care-giving relationship total	18	54.17(8.22)	54.28 (8.30)	0.11 (-3.48 to 3.70), 0.95	21	54.52 (8.57)	52.48 (9.68)	-2.05 (-5.19 to 1.09), 0.19
HADS anxiety sub-scale	19	6.89 (4.65)	6.84 (3.89)	-0.05 (-1.34 to 1.24), 0.93	22	6.00 (3.63)	6.50 (4.23)	0.50 (-0.81 to 1.81), 0.44
HADS depression sub-scale	19	4.68 (2.89)	5.05 (3.10)	0.37 (-2.21 to 1.37) <sup>a</sup> , 0.18	22	4.14 (3.04)	4.68 (2.55)	0.55 (2.05 to 1.11) <sup>a</sup> , 0.41
RSS emotional distress	19	9.95 (4.37)	10.11 (4.48)	0.16 (-1.59 to 1.91), 0.85	21	9.05 (4.14)	9.86 (5.07)	0.81 (-0.88 to 2.50), 0.33
RSS social distress	19	7.16 (3.80)	8.63 (4.15)	1.47 (0.45 to 2.49), 0.007	21	7.33 (3.77)	8.10 (4.11)	0.76 (-0.48 to 2.01), 0.22
RSS negative feelings	19	4.11 (1.91)	4.95 (3.05)	0.84 (-0.34 to 2.03), 0.15	22	4.64 (1.99)	5.14 (2.49)	0.50 (-0.44 to 1.44), 0.28
RSS total	19	21.21 (8.63)	23.68 (10.17)	2.47 (-0.58 to 5.52), 0.11	21	21.14 (8.15)	23.00 (10.10)	1.86 (-1.18 to 4.90), 0.22
GHQ somatic feelings	19	6.63 (4.22)	6.63 (3.70)	0.00 (-1.796 to 1.796), 1.00	21	5.76 (4.53)	6.19 (3.03)	0.43 (-1.30 to 2.16), 0.61
GHQ anxiety	17	6.29 (5.07)	7.59 (4.23)	1.29 (-0.86 to 3.44), 0.22	22	6.59 (4.09)	7.09 (4.09)	0.50 (-1.01 to 2.01), 0.50
GHQ social dysfunction	16	6.56 (2.39)	7.81 (2.97)	1.25 (-0.03 to 2.53), 0.06	22	7.59 (2.28)	8.36 (2.06)	0.77 (-0.40 to 1.94), 0.18
GHQ depression	18	1.44 (2.33)	1.83 (2.46)	0.39 (-0.91 to 1.69), 0.54	21	2.19 (3.14)	1.76 (2.72)	-0.43 (-2.06 to 1.20), 0.59
GHQ-28 total	15	21.00 (11.86)	23.80 (12.07)	2.80 (-2.74 to 1.08), 0.30	20	21.55 (12.27)	23.25 (9.88)	1.70 (-3.71 to 7.11), 0.52

<sup>a</sup> bootstrapped 95% confidence interval presented due to non-normal distribution of the data

Note: HADS scales and GHQ scales are scored such that a lower score indicates a better outcome

### *Resource use and associated costs*

Service use data was not normally distributed and was skewed towards lower costs so a Mann Whitney U test was used to detect significant differences between groups. Although the intervention group had higher service use costs in the three months before baseline, this was not a significant difference. No significant differences between groups were detected on baseline outcome data either. No adjustments were made for baseline cost or outcome values.

The mean cost of resource use at 10 months was £2,004 (s.d.  $\pm$ £1,585) for the joint RT group (including the cost of the intervention, at £964 per dyad) and £540 (s.d.  $\pm$ £585) for the control group, a significant difference ( $p=0.01$ ). Post-hoc tests indicated that there was no significant difference on any individual service use cost category but there was a trend for higher costs overall in the intervention group (for example, mean day hospital cost was £399 per carer in the intervention group and £6 per carer in the control group; mean medical inpatient cost was £160 per carer in the intervention group and £0 in the control group).

### *Cost-effectiveness findings*

As recommended in Briggs and O'Brien (2001) and Dakin and Wordsworth (2013), cost-effectiveness analysis (CEA) was undertaken to estimate the uncertainty surrounding joint density of cost and effects, regardless of whether statistically significant differences were observed on the effectiveness measures. Cost-effectiveness was evaluated in terms of the primary outcome measure, the ICECAP-O. Bootstrapping was used to address uncertainty around the point estimates of costs and effects. Bootstrapped estimates on a cost-effectiveness plane are presented for the ICECAP-O, QALY as calculated using the EQ-5D and QCGR, along with cost effectiveness acceptability curves (Figures 6.5 to 6.10).

Table 6.5 shows the results of the CEA. The mean difference in cost between groups was £1,464 (bootstrapped 95% CI of £758 to £2,313) and the mean difference in ICECAP-O score was -0.02 (bootstrapped 95% CI of -0.105 to 0.066) at 10 months, which would result in RT being dominated by usual care. Where the intervention was more costly and less effective than the control condition, resulting in a negative incremental cost effectiveness ratio (ICER), 'Control condition dominant' is stated. Point estimate ICERs are presented for the other measures. While a widely accepted threshold of £20,000-£30,000 per QALY exists (NICE, 2013), as yet there are no societal thresholds for the willingness to pay for a unit improvement in the other outcome measures. The interpretation of what a unit increase represents for these measures is discussed in Part 3, page 140.

**Table 6.5:** Cost-effectiveness summary

Cost-effectiveness analysis	N (I,C)	Mean Cost		Mean difference (bootstrapped 95% CI)	Mean change between baseline and 10 months		Mean difference (bootstrapped 95% CI)	ICER point estimate	Probability intervention is cost effective at a threshold of...	
		I	C		I	C			£20,000 per unit increase	£30,000 per unit increase
ICECAP-O	41 (19, 22)	£2,004	£540	£1,464 (£758 to £2,313)	-0.01	0.01	-0.02 (-0.105 to 0.066)	Control condition dominant	2%	8%
QCGR total	39 (18, 21)	£1,929	£520	£1,409 (£745 to £2,183)	0.11	-2.05	2.16 (-2.060 to 6.397)	£652	83%	84%
HADS anxiety	41 (19, 22)	£2,004	£540	£1,464 (£721 to £2,237)	0.05	-0.50	0.55 (-1.166 to 2.289)	£2,662	71%	72%
HADS depression	41 (19, 22)	£2,004	£540	£1,464 (£721 to £2,247)	-0.37	-0.55	0.18 (-1.818 to 0.705)	£8,133	56%	57%
RSS total	40 (19, 21)	£2,004	£533	£1,471 (£814 to £2,319)	-2.47	-1.86	-0.61 (-4.406 to 3.231)	Control condition dominant	34%	34%
GHQ-28 total	35 (15, 20)	£1,808	£520	£1,288 (£625 to £2,184)	-2.80	-1.70	-1.10 (-11.795 to 6.833)	Control condition dominant	29%	29%
Cost-utility analysis	N (I,C)	Mean Cost		Mean difference (bootstrapped 95% CI)	Mean QALY at 10 months		Mean difference (bootstrapped 95% CI)	ICER point estimate	Probability intervention is cost effective at a threshold of...	
		I	C		I	C			£20,000 per QALY	£30,000 per QALY
EQ-5D QALY	41 (19, 22)	£2,004	£540	£1,464 (£731 to £2,178)	0.69	0.63	0.06 (-0.017 to 0.182)	£24,400	53%	71%
EQ-VAS	41 (19, 22)	£2,004	£540	£1,464 (£764 to £2,296)	-0.021	0.002	-0.023 (-0.131 to 0.086)	Control condition dominant	33%	33%

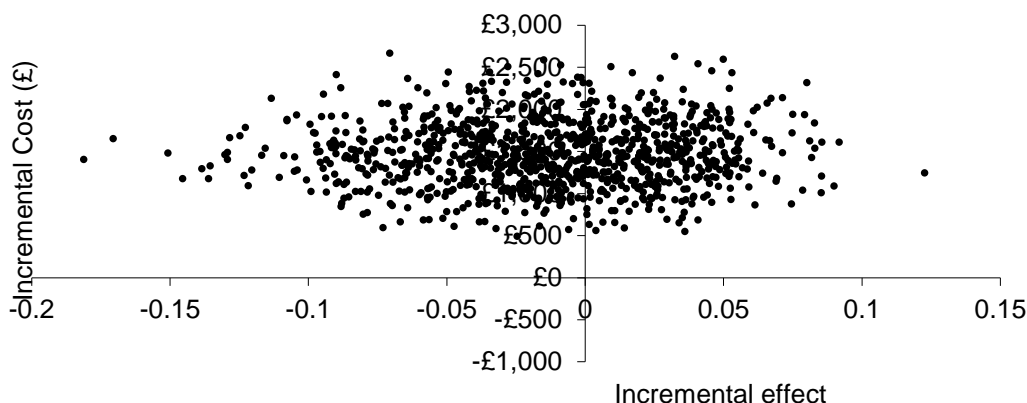
Key: I= Intervention, C= Control

Notes:

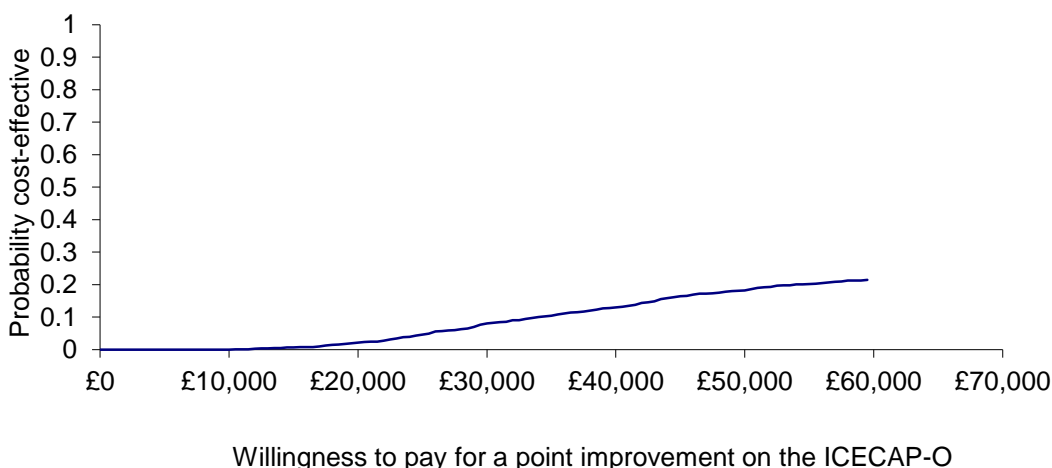
1. Although ‘Control condition dominant’ is stated, it is worth noting that no statistically significant difference was found on any of the effects.
2. The small differences in confidence intervals for the cost are due to random variation from recalculation of the bootstrap each time.
3. In Table 6.5 instruments have been reverse scored where necessary, so that a negative score indicates a worse outcome.
4. EQ-VAS change scores have been divided by 100 to scale them to be the same as the EQ-5D index, enabling comparison.

When the ICECAP-O was the measure of effect, the control condition was dominant. This can be seen in Figures 6.5, where the majority of the plots (60%) fell in the North West quadrant, indicating that the intervention is more costly and less effective than the control condition. Figure 6.6 demonstrates that there was a 2% probability that the intervention was cost-effective when setting the cost-effectiveness threshold at £20,000 per unit increase in the ICECAP-O and an 8% probability that the intervention was cost-effective when setting the threshold at £30,000 per unit of increase in the ICECAP-O.

**Figure 6.5:** Cost-effectiveness plane for the ICECAP-O with 1,000 bootstrapped ICER estimates



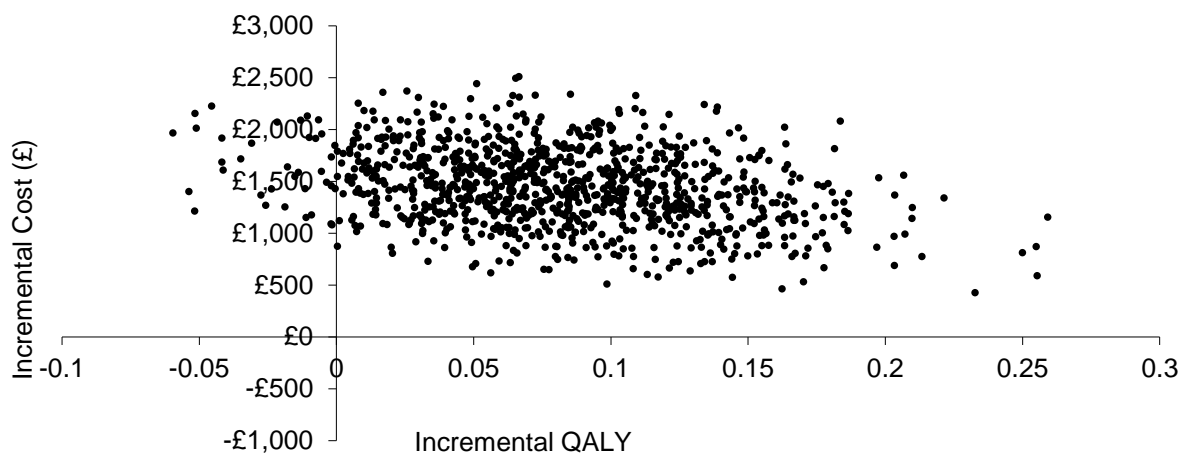
**Figure 6.6:** Cost-effectiveness acceptability curve for the ICECAP-O



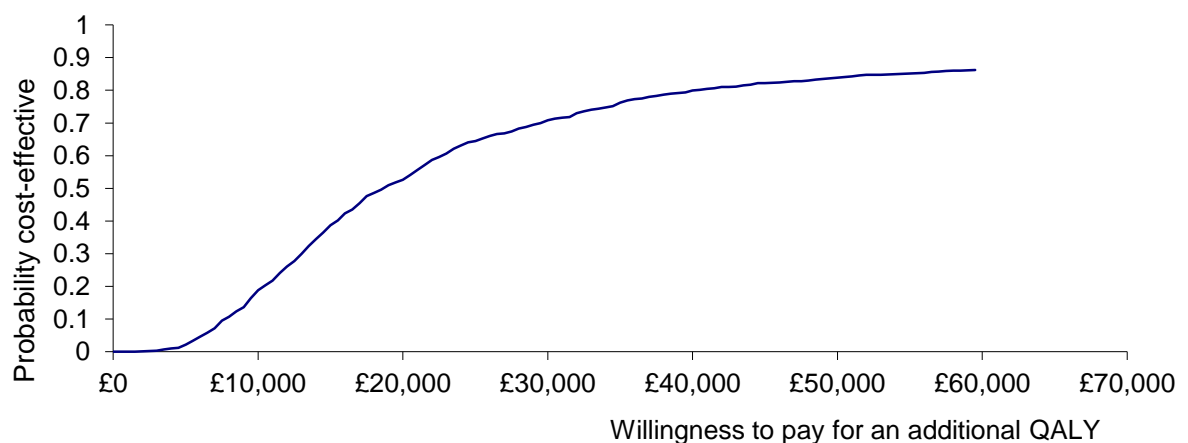


When the QALY (calculated using the EQ-5D index) was the unit of effect, 96% of plots fell in the North East quadrant (Figure 6.7), indicating that the intervention is more costly and more effective than the control condition. There was a 53% probability that the intervention was cost-effective at a willingness to pay threshold of £20,000 per QALY gained and a 71% probability that the intervention was cost-effective at a threshold of £30,000 per QALY gained (Figure 6.8).

**Figure 6.7:** Cost-effectiveness plane for the QALY with 1,000 bootstrapped ICER estimates

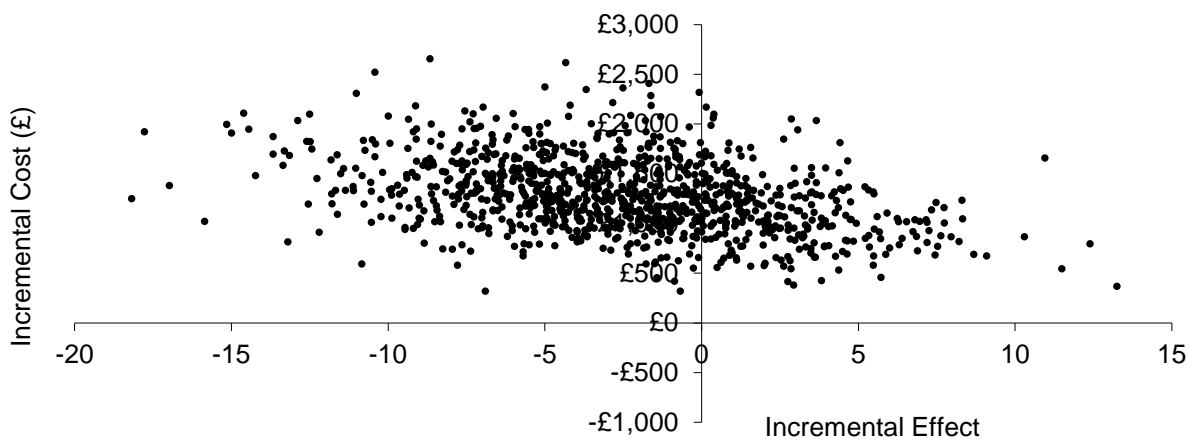


**Figure 6.8:** Cost-effectiveness acceptability curve for the QALY

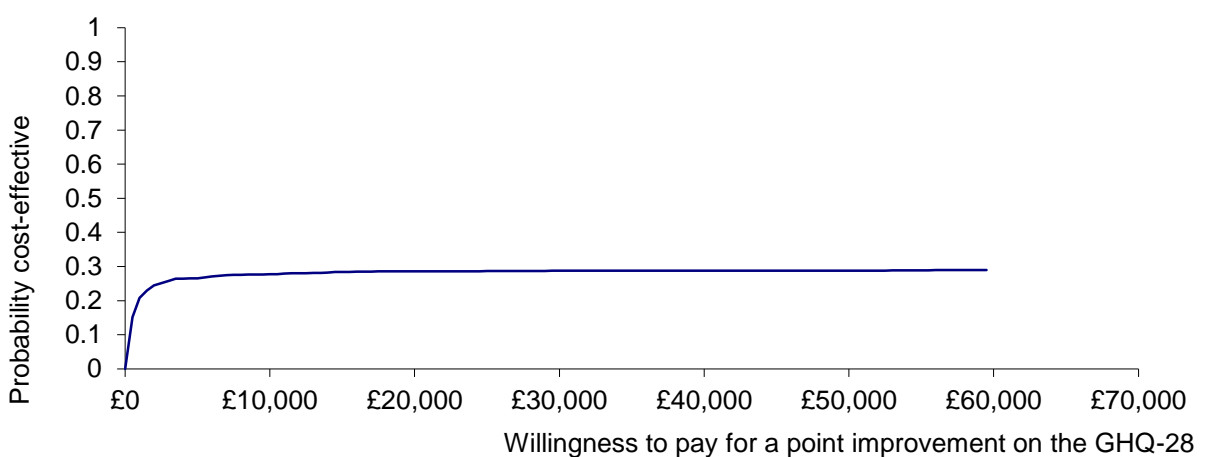


The cost-effectiveness plane for the GHQ-28 in Figure 6.9 shows that 73% of plots fell in the North West quadrant, indicating that the intervention is more costly and less effective than the control condition. There was a 29% probability that the intervention was cost-effective at both the £20,000 and £30,000 willingness to pay per point improvement on the GHQ-28 threshold (Figure 6.10).

**Figure 6.9:** Cost-effectiveness plane for the GHQ-28 with 1,000 bootstrapped ICER estimates



**Figure 6.10:** Cost-effectiveness acceptability curve for the GHQ-28



### *Exploratory subgroup analysis*

Out of the 19 carers in the control group 12 attended 6 or more of the 12 weekly joint RT sessions. These carers were used in a pre planned exploratory subgroup analysis to explore the effect of fidelity to the intervention. Carers in the intervention arm of the subgroup analysis had a slightly higher improvement in ICECAP-O scores at 10 months than the control group, in contrast to carers in the main ICECAP-O analysis who were worse off than the control group at 10 months. The ICER point estimate was £1,623,333 and there was only a 7% probability that the intervention was cost effective at the £20,000 per point improvement threshold and a 16% probability that the intervention was cost-effective at the £30,000 per point improvement threshold.

A secondary pre planned exploratory subgroup analysis was conducted on the thirteen carers who were aged 65 or over at baseline. This analysis was conducted because the primary outcome measure for effectiveness was the ICECAP-O, which was designed for older members (65+) of the general population. Carers in the control group had an improvement in their ICECAP-O score, but carers in the intervention group had a slight decline. RT was dominated by the control condition in this exploratory sub group analysis.

### **Conclusion**

The sub study did not find RT to be effective or cost-effective for carers of people with dementia when the ICECAP-O was the measure of effect. When the QALY was the measure of effect the ICER was £24,400 (53% probability of RT being cost-effective at a maximum willingness to pay threshold of £20,000 per QALY gained and a 71% probability of RT being cost-effective at a maximum threshold of £30,000 per QALY gained).

**Table 6.6:** Subgroup analysis

	N (I,C)	Mean Cost		Mean difference (95% CI)	Mean change between baseline and 10 months		Mean difference (95% CI)	ICER point estimate	Probability intervention is cost effective at a threshold of...	
		I	C		I	C			£20,000 per unit increase	£20,000 per unit increase
ICECAP-O 6 or more sessions	34 (12, 22)	£2,001	£540	£1,461 (£588 to £2,470)	0.01	0.01	0.0009 (-0.104 to 0.080)	£1,623,333	7%	16%
ICECAP-O Aged 65+	35 (13, 22)	£1,885	£540	£1,345 (£563 to £2,276)	-0.01	0.01	-0.0191 (-0.127 to 0.078)	Control condition dominant	5%	12%

I= Intervention, C= Control

### **Part 3: Comparing findings and discussion**

#### *Main findings*

Despite anecdotal evidence that dyads who participate in joint RT groups find the sessions therapeutic and beneficial, we accepted the null hypothesis at 95% that there is no effect of joint RT compared to usual care when the ICECAP-O is the unit of effect.

The primary outcome measure for carers in the main REMCARE trial was the GHQ-28. The main trial reported no significant differences in the total GHQ-28 score at 10 months; however, carers receiving joint RT recorded significantly higher values on the GHQ-28 Anxiety sub-scale than carers in the control group. No significant differences were found between people with dementia in the intervention group and control group either. Economic analysis indicated that joint RT was unlikely to be cost-effective.

The sample of carers in the ICECAP-O sub study was thought to be representative of the carers in the main REMCARE economic analysis sample; however no firm conclusion about this can be made with such a small sample. No significant differences were found between carers in the intervention group and the control group at either the 3 month or 10 month follow-up. Under the primary outcome measure for this sub study, the ICECAP-O, the intervention was found to be neither effective nor cost-effective. A non-statistically significant difference of -0.02 (bootstrapped 95% CI of -0.105 to 0.066) was found between groups on the ICECAP-O at 10 months.

Including the cost of the intervention, mean costs of service use were significantly higher for carers in the intervention group compared to the control group in the ICECAP-O sample (mean difference £1,464, bootstrapped 95% CI £758 to £2,313). In the main economic analysis sample the mean difference in cost of service use was £1,136 (bootstrapped 95% CI of £322 to £1,941). The differences in costs between the intervention group and control group in the ICECAP-O sample and main REMCARE economic analysis sample were not due to any particular service use category, but were due to overall costs differing between groups. At baseline, the only significant difference in service usage between the ICECAP-O sample and main REMCARE economic analysis sample was seen in lunch club use. At 10 months, the significant difference in lunch club use remained (ICECAP-O sample mean use 3.02 (s.d.  $\pm$ 15.5, main economic sample mean use 0.62, s.d.  $\pm$ 3.84,  $p=0.026$ ). No significant differences in other service use were seen across the two samples. When comparing service use across intervention groups in both samples and then control groups in both samples, no significant

differences were seen. However, there were three carers in the intervention arm of the main economic analysis sample who were heavy users of outpatient service (28, 31 and 45 visits over the 10 months between baseline and second follow-up). There was no evidence to suggest that joint RT leads to a reduction in health and social care service use.

A cost-effectiveness analysis was undertaken despite there being no statistically significant differences observed in the effectiveness data for the ICECAP-O or EQ-5D. This is in line with Briggs and O'Brien (2001), who advocate conducting a CEA with presentation of cost-effectiveness planes and cost-effectiveness acceptability curves to quantify uncertainty surrounding ICER point estimates. The CEA showed RT to be dominated by the control condition when the measure of effect was the ICECAP-O, EQ-VAS, RSS and GHQ-28. An ICER of £24,400 (53% probability of the intervention being cost-effective at a maximum willingness to pay threshold of £20,000 per QALY gained and a 71% probability of the intervention being cost effective at a maximum willingness to pay threshold of £30,000 per QALY gained) was estimated when the QALY was the unit of effect, with 96% of plots falling in the North East quadrant on the cost effectiveness plane.

A widely accepted threshold of £20,000-£30,000 per QALY exists (NICE, 2013), but as yet there are no societal thresholds for the maximum willingness to pay for a point improvement on the ICECAP-O. In Table 6.5 the probabilities that the intervention is cost-effective at a threshold of £20,000 and £30,000 per point improvement are presented for the range of carer outcome measures used, and hence the uncertainty around the estimates are shown. A mean difference between groups of 0.02 (bootstrapped 95% CI of -0.105 to 0.066) was found on the ICECAP-O. Given that the ICECAP-O capability index is anchored between 0 and 1, a difference of -0.02 can be considered negligible. The QCGR is summed on a scale of 8-40, so the mean difference between groups of 2.16 (bootstrapped 95% confidence interval of -2.060 to 6.397) is again negligible. An ICER of £652 per point improvement on the QCGR was estimated. Each sub scale of the HADS is scored between 0-21, so the mean differences between groups of 0.55 (bootstrapped 95% confidence interval of -1.166 to 2.289) on the anxiety sub scale and 0.18 (bootstrapped 95% confidence interval of -1.818 to 0.705) on the depression sub scale is again minimal. An ICER of £2,662 per point improvement on the HADS anxiety sub scale and an ICER of £8,133 per point improvement on the HADS depression sub scale was estimated. The RSS is scored between 0-60, so the mean difference between groups of 0.61 (bootstrapped 95% confidence interval of -4.406 to 3.231) is once

again small. The GHQ-28 is scored between 0-28, so the mean difference of 1.10 between groups (bootstrapped 95% confidence interval of -11.795 to 6.833).

Drop-out rates for the carers in the bolt-on study were 38%, compared to 28% in the main trial. No cause for the higher rate of attrition is known. 63% of carers in this sample attended at least 6 of the 12 weekly joint RT sessions, in the main sample the figure was 71%. Subgroup analysis of carers attending 6+ sessions in the main REMCARE trial revealed a near significant difference on the Relatives Stress Scale between carers in the intervention group and control group.

#### *Comparison with other studies*

The major strength of the REMCARE trial was that it is one of the first economic evaluations conducted alongside a randomised controlled trial of joint RT. Findings from the main REMCARE analysis and this bolt-on study echo the findings of earlier systematic reviews; evidence for the effectiveness of RT for people with dementia and their carers is still inconclusive (Cotelli, Manenti, & Zanetti, 2012; Chin, 2007; Woods, Spector, Jones, Orrell, & Davies, 2005). The reviews only found one previous trial which recorded outcome measures for carers as well as people with dementia (Thorgrimsen, Schweitzer, & Orrell, 2002). That study was in fact a pilot study of the ‘Remembering Yesterday, Caring Today’ model (Schweitzer & Bruce, 2008) which was subsequently used in the REMCARE trial. The pilot study found no statistically significant differences between the people with dementia in the intervention group (7 dyads) and the control group (4 dyads) at the final follow-up. Carers in the intervention group had significant improvements in their level of strain (measured by the GHQ-12 and RSS) however, there was no statistically significant difference between the intervention and control group at the final follow-up.

To our knowledge, the ICECAP-O has not been used as an outcome measure in a trial involving carers of people with dementia before and this bolt-on study adds to the evidence base of its acceptability in different populations. The use of the ICECAP-O as a valid outcome measure for carers of people with dementia is explored in Chapter 5. To date, studies which have used the ICECAP-O include the validation of it as a proxy measure in care homes comparing restrained versus unrestrained clients (Makai, Brouwer, Koopmanschap, & Nieboer, 2012); the validation of it in a clinical rehabilitation clinic setting and Australian national transition care (Ratcliffe, Laver, Couzner, Quinn, & Crotty, 2011); the use of it in patients with moderate to severe arthritis (Mitchell, Roberts, Barton, & Coast, 2012) and the

use of it in a trial of telehealth for patients with long term conditions (Henderson et al., 2013). The Henderson et al (2013) paper is the first to publish cost-effectiveness results using the ICECAP-O as an outcome measure. The authors note that no societal threshold has been set for the willingness to pay for a point improvement on the ICECAP-O (a movement from no capability to full capability) and findings must be interpreted with caution. The estimated ICER for the telehealth intervention compared to usual care was similar when both the QALY as calculated using the EQ-5D and the ICECAP-O was the unit of effect (approximately £90,000 per unit change in both cases).

### *Study challenges*

The main limitation to the bolt on study using the ICECAP-O is the number of participants. As the REMCARE trial was already underway when the sub-study was conceived, ICECAP-O data at all three time-points was only available for 41 carers. Loss of power due to small numbers will limit the robustness of the findings; to collect more data it will be useful to include the ICECAP-O as an outcome measure at the onset of future trials involving carers of people with dementia. The small sample size also leads to uncertainty surrounding the confidence intervals around differences in costs and outcomes generated using bootstrapping, as typically sample sizes of over 30 are used for this technique (Mooney & Duval, 1993). Future research with higher numbers of carers than was achieved in this study is required to establish the use of the ICECAP-O as a potential outcome measure in research involving carers of people with dementia.

Psychosocial interventions are complex, often conducted in populations with long term conditions and targeting a number of outcomes. The impact can go beyond the health and social care sector so a wide societal perspective is required to evaluate complex interventions. For reminiscence therapy to be successful the participants have to be willing and able to engage with facilitators and fellow group members. In older populations such as people with dementia a number of reasons could affect initial uptake of the intervention and subsequent participation. Although participants could withdraw from the study at any time without giving a reason, it is known that attrition occurred for a number of reasons, including: death, illnesses, medical appointments conflicting with session times, logistical reasons (although transport to and from sessions was offered to participants) and participants or their carers simply deciding that RT was not suitable for them. In the main REMCARE trial 11% of participants randomised to receive joint RT did not attend a single session.



The control group condition of ‘usual care’ is also problematic. The trial was conducted across 8 sites in England and Wales. The health and social care services available in each area can vary greatly so there is no standardised ‘usual care’ across the United Kingdom. The variability of services meant that costing service usage was time consuming, with researchers needing to find and assign unit costs for a large number of secondary services.

Data collection points were baseline, 3 months and 10 months. The 10 month point allowed the medium term effects of RT to be collected. Data on drug use was collected; however, it is unclear whether changes in the frequency of drug use resulting from receiving RT would be observed in 10 months or whether a longer follow-up period is required.

### **Conclusion**

As discussed in Chapter 3, the choice of outcome measure used in an economic evaluation alongside a trial is important as it will affect the effectiveness and hence cost-effectiveness results. The purpose of the bolt on study was to investigate whether the ICECAP-O, an instrument developed from the capability approach, was more appropriate than the EQ-5D, an instrument focusing on physical health, in a study involving carers of people with dementia. The findings of the bolt on study re-enforce the findings of the main REMCARE study; joint-RT was not found to be effective or cost-effective for carers of people with dementia. Cost-effectiveness planes and cost-effectiveness acceptability curves were presented to demonstrate the uncertainty surrounding the cost-effectiveness estimates. The choice of instrument did not have an effect on our conclusion about the cost-effectiveness of RT.

Randomised controlled trials of RT are primarily focused on the person with dementia; further rigorous research is needed to establish the effect on carers receiving joint RT. Carers did not report any problems with completing the ICECAP-O; however, its suitability as an outcome measure for research involving carers of people with dementia also needs further exploration, preferably through a study involving higher numbers of participants.

### **Acknowledgements**

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## Chapter 7: Discussion

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## **Purpose of thesis restated**

Current NICE guidance on the assessment of technologies and interventions calls for the use of generic preference based utility instruments, such as the EQ-5D, for measuring the quality of life component of the QALY (NICE, 2013). In the introduction chapter I argued that in certain populations, such as carers of people with dementia, the EQ-5D might not be sufficient as it focuses on physical health functioning, whereas interventions involving carers are often complex and target multiple outcomes. I proposed an alternative approach to measuring quality of life in carers of people with dementia, namely the capability approach. The purpose of this thesis was to explore the theoretical suitability, validity and application of a capability based instrument, the ICECAP-O, to interventions involving carers of people with dementia.

This chapter reviews the findings of the thesis by chapter, and how these findings relate to the research questions posed in the introduction. I discuss the theoretical and applied implications of the findings for government policy on dementia carer research, funding bodies of dementia research and finally commissioners of local services. Strengths and limitations of this thesis are presented. Finally, recommendations for future research directions are considered.

## **Findings by chapter**

### *Chapter 1- Introduction*

Chapter 1 provided a general introduction to both the thesis and the topic being explored. The methodological perspective underpinning the choice of health economics instruments was outlined, along with an overview of the data sources used in this thesis and the thesis structure.

### *Chapter 2- Systematic review of interventions supporting informal carers*

Chapter 2 was a systematic review of the cost-effectiveness of interventions for supporting informal carers of people with dementia living in the community. As over half of the estimated £23 billion annual cost of dementia to the United Kingdom economy is borne by informal carers (Luengo-Fernandez et al., 2010), there is a clear economic argument for health and social care interventions to target informal carers as well as people with dementia. This chapter sought to establish what cost-effectiveness evidence existed for interventions involving carers of people with dementia living at home, and what types of interventions carers are typically invited to participate in. To be included in the systematic review, a study had to include both a cost and a health-related outcome measure for an informal carer of a

person with dementia living in the community. The interventions could target the person with dementia, their carer, or both as a dyad.

Only twelve studies which included both costs and a health-related outcome measure for the carer were found. Four studies were on pharmacological interventions, primarily targeting the person with dementia. Four studies were of psychosocial interventions. Four studies were on service delivery interventions, which involved a change in the management or delivery of existing services to people with dementia or their carer. Of the psychosocial interventions; one targeted the quality of life of the carer through a befriending scheme (Charlesworth et al., 2008), two targeted the skills of the carer (Graff et al., 2008; Roberts et al., 1999) and one study targeted the carer and person with dementia as a dyad (Martikainen et al., 2004).

The review found four out of the twelve studies reported significant differences in outcome measures for the carers. Two of the studies were of psychosocial interventions and two were of service delivery interventions. Graff and colleagues (2006) reported a trial of an in-home/telephone based occupational therapy programme for carers compared to usual care. At the end of the intervention, carers in the intervention group reported a higher sense of competence, as measured by the Sense of Competence Scale, compared to those in the control group. Roberts and colleagues (1999) reported improved coping skills, as measured by the Indices of Coping scale, for carers in the intervention group who received one-to-one problem-solving therapy and counselling sessions, compared to carers who received usual care. Challis and colleagues. (2002) reported lower perceived burden for carers in the intervention group of a case management study compared to those in the control group. Engelhardt and colleagues (2008) reported an increase in nursing knowledge for carers receiving the intervention of a telephone support service, compared to carers receiving usual care. Although nursing knowledge is not in itself a health outcome, the ability of a carer to cope is correlated to stress levels (Matson, 1994). Two of the studies reporting significant effects for carers in the intervention group compared to the control group also reported significant differences in costs between groups (Roberts et al., 1999; Engelhardt et al., 2008). Neither study used a preference based utility instrument to measure carer quality of life, nor were the costs and consequences aggregated to form an incremental cost-effectiveness ratio. The main finding of Chapter 2 was that economic evidence of interventions to support carers of people with dementia living at home is sparse; no evidence of cost-effectiveness as measured by the QALY was found. An edited version of Chapter 2 is published in *International Psychogeriatrics* (Jones, Edwards & Hounscome, 2012a).

### *Chapter 3- Systematic review of outcome measures used with carers*

Chapter 3 presented the results of a systematic review of the instruments used most frequently in interventions involving carers of people with dementia. With the need to use appropriate outcome measures in economic evaluations discussed earlier, the focus of this chapter was to determine which instruments researchers currently include in carer interventions, and how applicable they are for health economic evaluations. Broad search terms for carer, randomised controlled trial and dementia were used, after exclusions were applied 362 studies using 228 outcome measures for carers remained. Using the categories developed by Moniz-Cook and colleagues (2008), these 228 outcome measures were classified into: burden; mastery; mood; quality of life; social support and relationships; and staff competency and morale measures. The review found that burden and mood measures were used most frequently in research involving carers of people with dementia. The least frequently used category of measure was staff competency and morale. While health economists do not typically measure the utility of paid carers, I included outcome measures in this category to give an indication of the aspects of health and social care which are typically measured for formal carers.

For the purpose of this thesis, the outcome measures found in the quality of life (QoL) category are of the most interest. Thirty-two quality of life measures were identified. The most popular QoL measures were the Short Form-36 (SF-36), the EQ-5D, the World Health Organization Quality of Life-brief (WHOQoL-BREF) and the Health Utilities Index (HUI). Preference based utility scores are available for all of the above apart from the WHOQOL-BREF. As preference based utility scores are available for the SF-36, EQ-5D and HUI, it is feasible for all three to be used to produce QALYs as part of cost-utility analysis.

The suitability of the EQ-5D for measuring quality of life in carers of people with dementia has been discussed previously, so I will limit this section to listing the domains again: mobility; self-care; usual activities; pain and discomfort; and anxiety and depression. The SF-36 was developed in America, while the SF-6D, the version used for calculating utility scores, was developed in the United Kingdom. Domains of the SF-6D are: physical functioning; role limitation; social functioning; pain; mental health; and vitality. The SF-6D contains more of a balance between physical functioning and mental health than the EQ-5D. Finally, the domains of the HUI3 are: vision; hearing; speech; ambulation; dexterity; emotion; cognition; and pain. The focus of the HUI3 is toward physical health functioning. The review found only 29 studies reporting cost information, and only three studies conducted a cost-utility analysis.

The EQ-5D, HUI2 and the Caregiver Quality of Life Instrument were used in these cost-utility analyses.

Chapter 3 concluded by proposing the ICECAP index of capability as a potential outcome measure for use in research involving family carers of people with dementia. An edited version of Chapter 3 is published in *Health and Quality of Life Outcomes* (Jones, Edwards & Hounsome, 2012b).

#### *Chapter 4- Qualitative exploration of carer quality of life*

Having identified the types of outcome measures frequently used with carers of people with dementia and introduced the ICECAP-O in the previous chapter, Chapter 4 presented the results of qualitative research exploring the suitability of capability based instruments for family carers of people with dementia. I conducted semi-structured interviews lasting up to one hour with eight family carers, typically in the home of the carer, although one interview was conducted at the carer's place of work. Questions revolved around the experiences of the carer, and how they perceived the impact of caring on their quality of life. Interviews were transcribed verbatim and I used framework analysis to code and analyse the qualitative data.

Four themes emerged from the interviews: social network and relationships; interactions with agencies; recognition of role; and time for oneself. Social support was important to carers as they sometimes found it hard to communicate effectively with the person being cared for. Interactions with agencies, such as Social Services or voluntary organisations such as the Alzheimer's Society were seen as a source of frustration as well as support. Frustration could be caused by the timeliness of a dementia diagnosis; however, carers were keen to praise staff and services who gave practical support and information. Recognition of role and feeling valued was important to the carers. Five of the carers were retired; none of the carers interviewed had decreased their paid employment hours to provide informal care to their family member. The final theme of time for oneself acknowledged the need carers felt for time away from caring activities. This time was not necessarily leisure time, as often it was used to catch up with chores which were not possible to do while looking after the person with dementia, for example shopping for groceries.

The four identified themes had a high degree of overlap with the capabilities based instruments (the ICECAP-O, ASCOT and CES). The EQ-5D had very little overlap with the themes. The highest degree of overlap was with the ICECAP-O and CES. The overlap with

the CES is unsurprising as it is a scale developed to measure the caring experience. The overlap with the ASCOT was good, although three domains were not mentioned by the carers: dignity; food and drink; and personal safety. This could be because the ASCOT was developed to measure the quality of care experienced by the care recipient, rather than the carer. Chapter 4 concluded that based on the small sample of carers interviewed, for research targeting the caring experience, the CES would be the preferred capability based instrument. However, as NICE require a generic preference based utility instrument for QALY calculations, there would still be the need to include EQ-5D as an outcome measure for carers.

#### *Chapter 5- Validity of the ICECAP-O for use with informal carers*

Chapter 5 presented an assessment of the feasibility and construct validity of the ICECAP-O index of capability for use with carers of people with dementia. The ICECAP-O has been validated for use with members of the general population aged 65+ (Coast et al., 2008c), but as it is a fairly recent development in the capability field there is a need to validate it for use with different populations in different settings.

Data for Chapter 5 came from two primary sources. The first source was baseline data for 157 carers in the Challenge FamCare cohort study of people with dementia who exhibited challenging behaviours. The ICECAP-O was included as an additional instrument in Challenge FamCare following an ethics amendment which I assisted with. To increase the numbers for analysis, I designed and set up an online survey (through SurveyMonkey) for carers of people with dementia to collect additional data. This support group sample yielded data for an additional 151 carers.

Feasibility of using the ICECAP-O with carers of people with dementia was high, with 99% of Challenge FamCare and 96% of the support group carers completing all five items of the ICECAP-O. Carers in the support group sample were given the choice of completing the survey online or by postal copy; 70% of carers completed the survey online, indicating that the population involved in this study was reasonably computer literate and it is feasible to complete the ICECAP-O online. Ceiling effects were present on three of the five ICECAP-O domains: love and friendship; doing things that make you feel valued; and independence. Floor effects were present on the item 'thinking about the future without concern'. Internal consistency was good, with Cronbach's alpha of 0.77 for Challenge FamCare data and 0.84 for the support group data. The correlation between the ICECAP-O and other outcome measures was weak to moderate; however, relationships were in the expected direction.

The results of Chapter 5 suggest that the ICECAP-O is feasible and valid for use with carers of people with dementia; however, a larger sample size is needed for a definitive answer, along with a test-retest reliability study.

#### *Chapter 6- Cost-effectiveness analysis of REMCARE*

Chapter 6 presented the results of a cost-effectiveness analysis for a sub sample of carers enrolled in the REMCARE joint-reminiscence therapy for people with dementia and their carers. The ICECAP-O was included as an additional instrument following an ethics amendment in November 2009. Of the 336 carers used in the main economic analysis of the REMCARE trial, 41 carers completed the ICECAP-O at all three time-points and were included in this analysis.

In the main REMCARE analysis, no significant differences were found between carers in the intervention group and control group on the General Health Questionnaire-28 (GHQ-28), EQ-5D, Relatives Stress Scale (RSS), Hospital Anxiety and Depression Scale (HADS) or Quality of Care-giving Relationship (QCGR) scale at any time point. In my sub sample analysis of 41 carers described in this thesis, no significant differences were found between groups on the ICECAP-O, GHQ-28, EQ-5D, RSS, HADS or QCGR.

In the main REMCARE analysis, costs in the intervention group were significantly higher than the control group at 10 months (£2,495 compared to £1,359). This result was also found in my sub sample, where mean costs per carer were significantly higher in the intervention group compared to the control group at 10 months (£2,004 compared to £540). The intervention group had higher costs than the control group at baseline, but this was not a statistically significant difference and was not adjusted for. Post-hoc tests indicated the significant difference in costs was not due to any individual service use category, but there was an overall trend for higher costs for carers in the intervention group.

The mean difference in ICECAP-O scores between groups was -0.02 (bootstrapped 95% CI of -0.105 to 0.066) at 10 months (the intervention group being lower than the control group). The ICECAP-O is scored on a scale of 0 to 1, so a difference of 0.02 is negligible. The mean difference in costs was £1,464 (bootstrapped 95% CI of £758 to £2,313), which would result in RT being dominated by usual care. The cost-effectiveness acceptability curve indicated a 2% probability that RT was cost-effective at a willingness to pay threshold of £20,000 per



point improvement and an 8% probability that RT was cost-effective at a threshold of £30,000 per point improvement on the ICECAP-O.

Chapter 6 results supported the findings from the main REMCARE economic analysis; joint-remembrance therapy compared to usual care for people with dementia and their carers was not found to be effective or cost-effective.

### **Research questions revisited**

This thesis set out to answer two overarching research questions. To do this, I defined five research sub-questions and dedicated a chapter to exploring each sub-question (see Figure 7.1).

**Figure 7.1:** Research questions addressed by this thesis

**Overarching research questions addressed by this thesis:**

1. To what extent is the capability approach appropriate for measuring quality of life in informal carers of people with dementia?
2. What implications does this have for future economic evaluations of interventions to support family carers of people with dementia?

**Research sub-questions:**

1. What existing literature is there on the effectiveness and cost-effectiveness of interventions to support informal carers of people with dementia?
2. What outcome measures are used most frequently in interventions involving carers of people with dementia, and how useful are these measures for economic evaluation?
3. What do family carers of people with dementia perceive as affecting quality of life, and how well do capability based instruments capture these aspects of quality of life?
4. To what extent is the ICECAP-O valid for use with carers of people with dementia?
5. To what extent does the ICECAP-O capture the effect of a psychosocial intervention of joint reminiscence therapy for carers of people with dementia?

I will address the research sub-questions first:

1. Evidence on the effectiveness of interventions to support informal carers of people with dementia living in the community is negligible, with only four studies found which demonstrated a significant outcome for carers. Cost-effectiveness of these interventions was not established in the literature.
2. The systematic review in Chapter 3 found that burden and mood instruments are used the most frequently as outcome measures in research involving carers of people with dementia. The most frequently used quality of life measures which could be used to calculate QALYs are the SF-36, EQ-5D and HUI.
3. The four themes identified as affecting quality of life for family carers by the qualitative research in Chapter 4 are: social network and relationships; interactions with agencies; recognition of role; and time for oneself. The domains of the Carer Experience Scale and ICECAP-O had a high degree of overlap with these themes, indicating that these capabilities based instruments are a potentially useful alternative to instruments focused on physical functioning, such as the EQ-5D.
4. Chapter 5 found that the ICECAP-O is both feasible and valid for use with informal carers of people with dementia.
5. The ICECAP-O produced effectiveness results which supported the EQ-5D in the sub sample analysis of the REMCARE trial of joint reminiscence therapy compared to usual care; however, due to the small sample used in this analysis the answer to this question is inconclusive at present.

I will address the overarching research questions posed at the start of the thesis in the Discussion section which follows.

## **Discussion**

This thesis set out to explore the application of the capability approach, in particular through the use of the ICECAP-O, to research involving informal carers of people with dementia. A variety of research methods (systematic reviewing, qualitative techniques and quantitative techniques) were employed to answer the research questions set out in the introduction. The main finding of this thesis is that the capability approach is appropriate for use with informal carers of people with dementia, and the ICECAP-O is both feasible and valid for use with this population.

An emerging pattern from the two systematic review chapters is that very little economic evidence exists surrounding the cost-effectiveness of interventions to support informal carers of people with dementia. While screening papers for inclusion in Chapter 2, I found many studies focusing on the clinical effectiveness of interventions targeting people with dementia. Instruments measuring the effects on informal carers were included as secondary outcomes; few trials were primarily targeted at improving quality of life for the carer. Of the studies which did include carer outcome measures, insufficient cost data were available to be able to undertake cost-effectiveness analyses. The review of outcome measures used with carers of people with dementia in Chapter 3 found that only 29 studies had collected cost data. Of these, only 3 had used preference based utility measures for carers, enabling cost-utility analysis consistent with NICE guidance (NICE, 2013).

This paucity of evidence on cost-effectiveness can be attributed to two reasons. First, as reported by Luengo-Fernandez et al. (2010), the amount of funding given to dementia research is not proportional to the burden of dementia to the economy. In 2008, dementia research funding in the United Kingdom was £50 million, and the economic burden was £23 billion. In comparison, £600 million was spent on cancer research; however, the economic burden of cancer was £12 billion, which is considerably lower than that of dementia. The lack of funding for dementia research limits the amount of large scale randomised controlled trials that can be undertaken to establish evidence on treatments that are both clinically-effective and cost-effective for people with dementia and their carers. The second reason for a lack of cost-effectiveness evidence in this area is that health economics data is not routinely collected as part of clinical-effectiveness trials. This was shown in Chapter 3, where <1% of studies included a cost-utility analysis and a further <1% included a cost-effectiveness analysis.

The first overarching research question was 'To what extent is the capability approach appropriate for measuring quality of life in informal carers of people with dementia?' The argument for needing to include instruments capable of measuring what an intervention is targeting has been presented in this thesis. Physical effects can be direct, such as back pain caused by lifting; or indirect, such as increased blood pressure from stress. The extra-welfarism framework of maximising health output through maximising QALYs is appropriate for measuring the physical effects of caring; the EQ-5D domains of mobility, self-care, usual activity and pain cover physical functioning well. However, caring for a person with dementia impacts on the mental health as well as the physical health of the carer. Schoenmakers and colleagues (2010) found that up to 30% of carers looking after people with dementia had

depression. The EQ-5D only contains one question on mental health (anxiety and depression). I argue that the capability approach is a more appropriate paradigm to measure carer quality of life than extra-welfarism, and additional capability-based instruments need to be included in future dementia carer research to capture a broader aspect of quality of life than physical functioning alone. This thesis has demonstrated that the ICECAP-O is both feasible and valid for use with informal carers of people with dementia. The qualitative research reported in Chapter 4, although only using a small sample size, suggests that the ICECAP-O is also *appropriate* for use with this population as the domains of the ICECAP-O are areas that carers identified as affecting their quality of life: love and friendship; thinking about the future; doing things that make you feel valued; enjoyment and pleasure; independence.

The second overarching research question was 'What implications does this have for future economic evaluations of interventions to support family carers of people with dementia?' As NICE guidance currently calls for the inclusion of the EQ-5D (NICE, 2013), interventions which target the mental health of carers need to include additional instruments which can detect changes in this aspect of quality of life alongside the EQ-5D, which focuses on physical functioning. The BECCA HTA trial of befriending for carers compared to usual care (Charlesworth et al., 2008) is an example of a psychosocial intervention primarily targeting the mental health of carers. The following instruments were included in the trial: EQ-5D, Hospital Anxiety and Depression Scale (HADS), Positive and Negative Affect Schedule (PANAS), Carers Assessment of Difficulties Index (CADI), Brief Coping Orientation for Problem Experience (COPE). Despite the intervention not targeting physical functioning, the EQ-5D was included to meet NICE cost-utility guidelines, but instruments focusing on mental health and carer efficacy were also included. This trial is a good example of a balanced researchers' tool-kit, aiming to measure clinical-effectiveness as well as cost-effectiveness.

The main implication for the ICECAP-O being both feasible and valid for use with informal carers of people with dementia is, quite simply, that it can now be included in future economic evaluations of interventions involving this population, and the conclusions drawn from the results will be meaningful. At present, the ICECAP-O does not have a preference-based utility scoring index. Respondents rate their level on the five ICECAP-O domains on a four point scale. Results are converted into an index score of between 0 and 1, with 0 representing no capability and 1 full capability. The algorithm to calculate the index score was developed using best-worst scaling with a sample of the UK population aged 65+ (Coast et al., 2008b). Best-worst scaling asks individuals to select the best and worst option in a given

set of samples (Finn and Louviere, 1992), revealing the preference for different options. However, as Coast et al. (2008b) note, while best-worst scaling can be seen as a choice experiment it is not strictly eliciting preferences as people are not asked to trade options, only select the best and worst from a set. Coast et al. (2008b) argue that best-worst scaling is closer to satisfying Sen's interpretation of how to elicit values than other methods commonly used, and also has the advantage of being less cognitively demanding.

To be able to elicit preference based scores for the ICECAP-O the measure would need to be anchored on an interval scale with 0 representing dead and 1 representing full health. A valuation exercise using a method such as standard gamble, time trade-off or a visual analogue scale would be required (Brazier et al., 2007). In contrast to the ICECAP-O, the EQ-5D scoring algorithm was developed using preference based tariffs for the different health states elicited using the time trade-off method with a selection of the UK population. The preference based scoring enables comparisons to be made between the EQ-5D index scores obtained in different studies and to population norms (Kind et al., 1999). As the ICECAP-O does not have preference based scores, it cannot replace the EQ-5D as a generic outcome measure. However, the inclusion of the ICECAP-O as an additional instrument, rather than as an EQ-5D replacement, in future research involving informal carers of people with dementia offers the opportunity for researchers to collect information on the changes to capability that a carer experiences as a result of an intervention. I have touched on the notion that many health care interventions have effects on other sectors, e.g. the social care sector. The capability framework allows outcomes other than health to be considered in economic evaluations, making the use of instruments such as the ICECAP-O ideal for complex interventions, which have the potential to have an impact beyond the health care sector.

### **Strengths and unique contribution of the thesis**

The annual economic burden of dementia to the United Kingdom economy is £23 billion, of which £12 billion is attributed to costs of informal care (Luengo-Fernandez et al., 2010). The need to support informal carers of people with dementia in their role is clear, and to do this high quality evidence of interventions which are both clinically-effective and cost-effective are required. The EQ-5D is the NICE favoured health-related quality of life outcome measure; however, this thesis explores alternative outcome measures with foundations in the capability framework. Before this current body of work, the extent of the appropriateness of using the capability approach as an alternative to the extra-welfarism approach of maximising health output for research involving informal carers of people with dementia was unknown. This

thesis offers a substantial increase in the current knowledge base surrounding the use of the capability approach and the ICECAP-O for informal carers of people with dementia.

This thesis presents, to my knowledge, the first results of using the ICECAP-O in a randomised controlled trial involving informal carers of people with dementia. Although the sample size used was small, the ICECAP-O performed comparably to the EQ-5D in terms of clinical-effectiveness results, which is an encouraging finding.

I have also explored the validation of the ICECAP-O for this population, something which to date has only been done for the general population aged 65 and over (Coast et al., 2008c) and in a Dutch nursing home setting (Makai et al., 2012). Validation is a requirement of all instruments used in research; if an instrument has not been validated for a particular population or research setting then the conclusion derived from an intervention is not particularly meaningful.

### **Implications for government policy**

The National Dementia Strategy in England (Department of Health, 2009a) proposed to improve service provision in dementia care by increasing awareness of dementia, striving to diagnose dementia earlier and offering a higher quality of care once a diagnosis had been made. On the face of it, these three aims all target the person with dementia; however, an earlier diagnosis of dementia can reduce the carer stress associated with the uncertainty of not knowing why a loved one is behaving differently; and improved quality of care can be applicable to the carer as well as the person with dementia. Improved quality of care for carers can include respite breaks or offering the carer information on how to cope with caring. To identify clinically- effective and cost-effective ways of increasing quality of care for carers, it is necessary to conduct research, ideally through randomised controlled trials as they are the gold standard for economic evaluations. While the research presented in this thesis does not have a direct implication for government policy, it serves to highlight the economic need to consider the role of informal carers in the United Kingdom. By emphasising the need to offer interventions that support carers, as well as the person with dementia, this thesis aims to encourage increased awareness of dementia which is in line with current government policy.

NICE is a non-departmental public body of the Department of Health with a remit of providing guidance to healthcare professionals and others about quality and value for money in healthcare. NICE must be encouraged to broaden their horizons about ways to measure quality of life and other benefits arising from interventions. In 2010 a cost impact project was

set up to develop NICE's approaches to assessing the cost effectiveness and cost impact of public health interventions. The key findings were that a three stage approach should be taken to determine the return on investment of public health interventions (NICE, 2011). Stage one involves presenting a cost consequence analysis with natural units, stage two involves the currently favoured cost-utility analysis with costs per QALY shown, and stage three involves presenting the first two stages to local decision-makers and allowing them to include information about implementation costs and population size to determine which interventions are a priority. By looking beyond the cost-per QALY methods traditionally favoured by NICE, this thesis and its investigation into the capability framework supports NICE's move towards exploring alternative methods of assessing cost-effectiveness.

### **Implications for funding bodies**

As highlighted by Luengo-Fernandez and colleagues (2010), funding for dementia research is relatively low compared to its economic burden. This thesis emphasises the need to consider the appropriateness of existing outcome measures used in research involving carers of people with dementia. When funding bodies are allocating their limited funds, I argue that they need to be vigilant that research proposals include outcome measures which are appropriate for the type of intervention and population being investigated.

The strong message arising from this thesis is that focusing on instruments that measure physical functioning is not always appropriate when attempting to measure quality of life; capability based instruments that bridge the gap between functioning and a wider quality of life should be considered. While this thesis was focused on investigating the suitability of the capability approach for carers of people with dementia, I believe that instruments such as the ICECAP-O offer the potential to explore quality of life in other areas. For example, the Lifelong Health and Wellbeing Programme launched in 2008 as a major cross-council initiative to support multi-disciplinary research into factors addressing healthy ageing and wellbeing in later life. The Arts and Humanities Council is a collaborator in the project, but using an instrument such as the EQ-5D to measure the benefits of an arts and humanities project is unlikely to demonstrate a benefit. The capability approach may be more suitable for measuring benefits in more diverse subject areas.

### **Implications for commissioners of local services**

The NHS Commissioning Board Health Authority was established in October 2011 with the aim of improving health outcomes for patients in England through making the NHS inclusive,

evidence-based and transparent. It is anticipated that the commissioning board will take over NHS strategic planning and delivery responsibility from the Department of Health, as well as many of the commissioning responsibilities currently undertaken by primary care trusts (PCTs) by April 2013 (NHS, 2012). The objective of a high quality of care and patient experience can be monitored through building routine data collection into audit and service evaluation. This thesis discusses the suitability of capability based instruments, and the qualitative research chapter found that the ASCOT offers real potential to contribute to the measurement of quality of care for people with dementia and their carers. The ASCOT measures social care related quality of life; however, the division between which services should be offered by the NHS and which should be offered by Social Services is often unclear. The ASCOT also benefits from having preference-based index scores, meeting one of the NICE requirements for cost-utility analysis. As the ASCOT was not developed for use with a specific population, it can be used to measure quality of care for a range of patients and results can be easily compared. Use of measures such as the ASCOT will support evidence-based decision making and commissioning in the use of public resources to support family carers of people with dementia.

### **Limitations of this thesis**

The unique contribution of this thesis has been stated; however, despite having confidence in the overall findings and significance of this work it is also necessary to consider the limitations.

Due to the selection of the ICECAP-O as the primary outcome measure for the validation work and the cost-effectiveness analysis of the sub-sample of carers in the REMCARE trial; findings can only be generalised for carers aged 65+. The last United Kingdom census found the most common age category for carers (of all types) to be ages 50-59 (The Office of National Statistics, 2002). However, as dementia is an illness that predominantly affects older people it is possible that carers of people with dementia, particularly spousal carers, are likely to be older than the average carer. The team that developed the ICECAP-O subsequently developed the ICECAP-A, which is suitable for use with adults aged 18+ (Al-Janabi et al., 2012). Had the ICECAP-A been available at the start of the REMCARE and Challenge FamCare trials it would have been included as the capability based outcome measure instead. As the purpose of this thesis was to explore the suitability of different capability based outcome measures for carers it would have been preferable to have included an instrument which could be used with a wider population than the more restrictive aged 65+ version.



Furthermore, the carers who participated in the REMCARE, Challenge FamCare and the qualitative research might not be fully representative of carers in the general population. Carers have many competing demands on their time so those experiencing extremely high levels of burden and stress are unlikely to take part in research. Unfortunately, it is the carers who are at breaking point who are most in need of support in their role. Experiences of those who are unwilling or unable to participate in research might be very different to those who do. Potential participants for the REMCARE trial were approached via a number of methods, including Community Mental Health Teams (CMHTs), day services, and relevant voluntary sector organisations. This recruitment strategy ensured that a good cross-section of carers from the general population would have been approached to take part in the trial; however, it is not possible to control which carers chose to take part.

A major limitation of this thesis is the sample size for the cost-effectiveness analysis undertaken in Chapter 6. Recruitment for the REMCARE trial of joint-remembrance therapy for people with dementia and their carer, as compared to usual care, began in May 2008. The inclusion of the ICECAP-O as an outcome measure for carers was approved as a bolt-on study in late 2009. The effect of this was that of the 336 carers whose data were used for the main economic analysis, only 41 had also completed the ICECAP-O at all three time-points. The small sample size limits the confidence that can be had in the findings of this particular aspect of the work; bootstrapping of cost and outcome data can be unstable when resampling is done from a small sample size (Mooney & Duval, 1993). However, the sub sample of carers used in this analysis was representative of carers in the main REMCARE sample and the clinical-effectiveness data yielded similar results i.e. no significant differences were found between intervention and control group at any time-point. It would be prudent to include the ICECAP-O as an outcome measure from the beginning of future trials to allow maximum data collection.

While this thesis has the advantage of using data from a variety of sources, I recognise that it only represents an in depth study into a small aspect of the capability field and its potential application in economic evaluations involving informal carers.

#### **Areas for further research**

- *Aim 1:* Investigate the ICECAP-O in a larger trial population to be able to effectively compare its performance to other outcome measures.

*Method:* Include the ICECAP-O as an outcome measure from the initial planning stage of a trial. As the REMCARE trial did not establish the effectiveness or cost-effectiveness of reminiscence therapy compared to usual care for people with dementia and their carers to be either clinically-effective or cost-effective, I would hesitate to recommend replicating the trial and would instead include the ICECAP-O in a different trial of a psychosocial intervention for older carers.

- *Aim 2:* Investigate the validity of the Carer Experience Scale for carers of people with dementia. In Chapter 4 the Carer Experience Scale was noted as having a good conceptual overlap with aspects of quality of life that were important to carers of people with dementia. It was not possible to investigate the construct validity of the Carer Experience Scale for carers of people with dementia as part of this thesis because the instrument had not been included in the Challenge FamCare study. Therefore, I propose the exploration of its validity with this population as future work.

*Method:* The methods used in Chapter 5 would be replicated; however it would be preferable to collect the data from researcher administered interviews because the Carer Experience Scale has not yet been validated for online completion.

- *Aim 3:* Investigate the suitability of the capability approach in a trial involving carers of people with a different chronic illness.

*Method:* As with Aim 1, this would involve the inclusion of a capability based instrument in a full scale trial. The ICECAP-A has been developed for adults aged 18+ so this would be the preferred instrument of choice over the ICECAP-O in a trial involving a wider age range of carers. Collecting data on how different interventions affect capability, and by extension quality of life, for different populations would enable comparisons to be made between groups/interventions and hence aid decision-making when allocating scarce resources.

## **Conclusion**

NICE has helped reinforce the idea of economic evaluation as an essential and mainstream component when determining the cost-effectiveness of health care interventions. Given the future economic burden of dementia care faced by the United Kingdom, now is the time to focus on the role played by informal carers, and on commissioning services to support carers. This thesis has a strong message that there is a need to use appropriate and valid outcome

measures in economic evaluations of health care research. This issue has already been identified in published literature (Payne et al., 2012; Kelly et al., 2005; Craig et al., 2008), and is particularly relevant when discussing complex interventions. Currently, generic preference based utility measures, such as the EQ-5D, are the favoured instruments for measuring quality of life in cost-utility analyses. By suggesting the use of capability based instruments, such as the ICECAP-O, to collect additional data on the clinical-effectiveness of interventions, this thesis encourages further debate surrounding how effects on carers are currently measured.

The current knowledge base surrounding the cost-effectiveness of interventions to support informal carers of people with dementia is insufficient. Consideration needs to be given to the outcome measures used in research with this population, as the effect of caring on quality of life is not limited to physical functioning, as measured by the NICE favoured EQ-5D, but also includes effects on mental health. The capability approach offers an opportunity to go beyond the traditional extra-welfarism framework of maximising health output, and instead consider a broader framework for measuring quality of life.

I set out to explore the suitability of the capability approach for use with informal carers of people with dementia. This thesis presents evidence that the ICECAP-O is feasible, valid and appropriate for use in research involving informal carers of people with dementia. The implication of this for future health economic evaluations is that the ICECAP-O can be included as an outcome measure, alongside the EQ-5D, with this population and the results will be meaningful.

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## Appendices

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## Appendix 1: EQ-5D

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By placing a tick in one box in each group below, please indicate which statements best describe your own health state today.

### **Mobility**

- I have no problems in walking about
- I have some problems in walking about
- I am confined to bed

### **Self-Care**

- I have no problems with self-care
- I have some problems washing or dressing myself
- I am unable to wash or dress myself

### **Usual Activities** (e.g. work, study, housework, family or leisure activities)

- I have no problems with performing my usual activities
- I have some problems with performing my usual activities
- I am unable to perform my usual activities

### **Pain/Discomfort**

- I have no pain or discomfort
- I have moderate pain or discomfort
- I have extreme pain or discomfort

### **Anxiety/Depression**

- I am not anxious or depressed
- I am moderately anxious or depressed
- I am extremely anxious or depressed

## Appendix 2: EQ-VAS

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To help people say how good or bad a health state is, we have drawn a scale (rather like a thermometer) on which the best state you can imagine is marked 100 and the worst state you can imagine is marked 0.

We would like you to indicate on this scale how good or bad your own health is today, in your opinion. Please do this by drawing a line from the box below to whichever point on the scale indicates how good or bad your health state is today.

**Your own health state today**

Best imaginable health state

100

90

80

70

60

50

40

30

20

10

0

Worst imaginable health state

## Appendix 3: ICECAP-O

© Joanna Coast and Terry Flynn

ABOUT YOUR QUALITY OF LIFE	
By placing a tick (✓) in ONE box in EACH group below, please indicate which statement best describes your quality of life at the moment.	
<b>1. Love and Friendship</b>	
I can have all of the love and friendship that I want	<input type="checkbox"/> 4
I can have a lot of the love and friendship that I want	<input type="checkbox"/> 3
I can have a little of the love and friendship that I want	<input type="checkbox"/> 2
I cannot have any of the love and friendship that I want	<input type="checkbox"/> 1
<b>2. Thinking about the future</b>	
I can think about the future without any concern	<input type="checkbox"/> 4
I can think about the future with only a little concern	<input type="checkbox"/> 3
I can only think about the future with some concern	<input type="checkbox"/> 2
I can only think about the future with a lot of concern	<input type="checkbox"/> 1
<b>3. Doing things that make you feel valued</b>	
I am able to do all of the things that make me feel valued	<input type="checkbox"/> 4
I am able to do many of the things that make me feel valued	<input type="checkbox"/> 3
I am able to do a few of the things that make me feel valued	<input type="checkbox"/> 2
I am unable to do any of the things that make me feel valued	<input type="checkbox"/> 1
<b>4. Enjoyment and pleasure</b>	
I can have all of the enjoyment and pleasure that I want	<input type="checkbox"/> 4
I can have a lot of the enjoyment and pleasure that I want	<input type="checkbox"/> 3
I can have a little of the enjoyment and pleasure that I want	<input type="checkbox"/> 2
I cannot have any of the enjoyment and pleasure that I want	<input type="checkbox"/> 1
<b>5. Independence</b>	
I am able to be completely independent	<input type="checkbox"/> 4
I am able to be independent in many things	<input type="checkbox"/> 3
I am able to be independent in a few things	<input type="checkbox"/> 2
I am unable to be at all independent	<input type="checkbox"/> 1

Tick one box only in each section



## Appendix 4: Carer Experience Scale

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**PLEASE TICK ONE BOX FOR EACH GROUP to indicate which statement best describes your current caring situation.**

**1. Activities outside caring** (*Socialising, physical activity and spending time on hobbies, leisure or study*)

- You can do most of the other things you want to do outside caring .....  1  
You can do some of the other things you want to do outside caring .....  2  
You can do few of the other things you want to do outside caring .....  3

**2. Support from family and friends** (*Personal help in caring and/or emotional support from family, friends, neighbours or work colleagues*)

- You get a lot of support from family and friends .....  1  
You get some support from family and friends .....  2  
You get little support from family and friends .....  3

**3. Assistance from organisations and the Government** (*Help from public, private or voluntary groups in terms of benefits, respite and practical information*)

- You get a lot of assistance from organisations and the Government .....  1  
You get some assistance from organisations and the Government .....  2  
You get little assistance from organisations and the Government .....  3

**4. Fulfilment from caring** (*Positive feelings from providing care, which may come from: making the person you care for happy, maintaining their dignity, being appreciated, fulfilling your responsibility, gaining new skills or contributing to the care of the person you look after*)

- You mostly find caring fulfilling .....  1  
You sometimes find caring fulfilling .....  2  
You rarely find caring fulfilling .....  3

**5. Control over the caring** (*Your ability to influence the overall care of the person you look after*)

- You are in control of most aspects of the caring .....  1  
You are in control of some aspects of the caring .....  2  
You are in control of few aspects of the caring .....  3

**6. Getting on with the person you care for** (*Being able to talk with the person you look after, and discuss things without arguing*)

- You mostly get on with the person you care for .....  1  
You sometimes get on with the person you care for .....  2  
You rarely get on with the person you care for .....  3

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## Appendix 5: Adult Social Care Outcomes Toolkit

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### four-level self-completion questionnaire (SCT4)

**1. Which of the following statements best describes how much control you have over your daily life?**

*By 'control over daily life' we mean having the choice to do things or have things done for you as you like and when you want.*

Please tick (☑) one box

- |   |                          |
|---|--------------------------|
| I have as much control over my daily life as I want   | <input type="checkbox"/> |
| I have adequate control over my daily life            | <input type="checkbox"/> |
| I have some control over my daily life but not enough | <input type="checkbox"/> |
| I have no control over my daily life                  | <input type="checkbox"/> |

**2. Thinking about keeping clean and presentable in appearance, which of the following statements best describes your situation?**

Please tick (☑) one box

- |   |                          |
|---|--------------------------|
| I feel clean and am able to present myself the way I like | <input type="checkbox"/> |
| I feel adequately clean and presentable                   | <input type="checkbox"/> |
| I feel less than adequately clean or presentable          | <input type="checkbox"/> |
| I don't feel at all clean or presentable                  | <input type="checkbox"/> |

**3. Thinking about the food and drink you get, which of the following statements best describes your situation?**

Please tick (☑) one box

- |  |                          |
|--|--------------------------|
| I get all the food and drink I like when I want  | <input type="checkbox"/> |
| I get adequate food and drink at OK times  | <input type="checkbox"/> |
| I don't always get adequate or timely food and drink   | <input type="checkbox"/> |
| I don't always get adequate or timely food and drink, and I think there is a risk to my health | <input type="checkbox"/> |

## four-level self-completion questionnaire (SCT4)

### 4. Which of the following statements best describes how safe you feel?

*By feeling safe we mean how safe you feel both inside and outside the home. This includes fear of abuse, falling or other physical harm.*

Please tick (☑) one

- |   |                          |
|---|--------------------------|
| I feel as safe as I want  | <input type="checkbox"/> |
| Generally I feel adequately safe, but not as safe as I would like | <input type="checkbox"/> |
| I feel less than adequately safe                                  | <input type="checkbox"/> |
| I don't feel at all safe  | <input type="checkbox"/> |

### 5. Thinking about how much contact you've had with people you like, which of the following statements best describes your social situation?

Please tick (☑) one

- |   |                          |
|---|--------------------------|
| I have as much social contact as I want with people I like          | <input type="checkbox"/> |
| I have adequate social contact with people                          | <input type="checkbox"/> |
| I have some social contact with people, but not enough              | <input type="checkbox"/> |
| I have little social contact with people and feel socially isolated | <input type="checkbox"/> |

### 6. Which of the following statements best describes how you spend your time?

*When you are thinking about how you spend your time, please include anything you value or enjoy including leisure activities, formal employment, voluntary or unpaid work and caring for others.*

Please tick (☑) one

- |  |                          |
|--|--------------------------|
| I'm able to spend my time as I want, doing things I value or enjoy   | <input type="checkbox"/> |
| I'm able to do enough of the things I value or enjoy with my time    | <input type="checkbox"/> |
| I do some of the things I value or enjoy with my time but not enough | <input type="checkbox"/> |
| I don't do anything I value or enjoy with my time                    | <input type="checkbox"/> |

## four-level self-completion questionnaire (SCT4)

**7. Which of the following statements best describes how clean and comfortable your home is?**

*Please tick (☑) one box*

- |  |                          |
|--|--------------------------|
| My home is as clean and comfortable as I want    | <input type="checkbox"/> |
| My home is adequately clean and comfortable      | <input type="checkbox"/> |
| My home is not quite clean or comfortable enough | <input type="checkbox"/> |
| My home is not at all clean or comfortable       | <input type="checkbox"/> |

**8. Which of these statements best describes how having help to do things makes you think and feel about yourself?**

*Please tick (☑) one box*

- |   |                          |
|---|--------------------------|
| Having help makes me think and feel better about myself                 | <input type="checkbox"/> |
| Having help does not affect the way I think or feel about myself        | <input type="checkbox"/> |
| Having help sometimes undermines the way I think and feel about myself  | <input type="checkbox"/> |
| Having help completely undermines the way I think and feel about myself | <input type="checkbox"/> |

**9. Which of these statements best describes how the way you are helped and treated makes you think and feel about yourself?**

*Please tick (☑) one box*

- |  |                          |
|--|--------------------------|
| The way I'm helped and treated makes me think and feel better about myself                 | <input type="checkbox"/> |
| The way I'm helped and treated does not affect the way I think or feel about myself        | <input type="checkbox"/> |
| The way I'm helped and treated sometimes undermines the way I think and feel about myself  | <input type="checkbox"/> |
| The way I'm helped and treated completely undermines the way I think and feel about myself | <input type="checkbox"/> |

This questionnaire has been created by the Personal Social Services Research Unit (PSSRU) at the University of Kent with funding from HM Government, and the University of Kent is the sole owner of the copyright in those materials. The University of Kent authorises non-commercial use of this questionnaire on the condition that anyone who uses it contact the ASCOT team ([ascot@kent.ac.uk](mailto:ascot@kent.ac.uk)) to discuss this use and enable the PSSRU at University of Kent to track authorised non-commercial use. The University of Kent does not authorise commercial use of the questionnaire. Anyone wishing to obtain a licence for commercial use of any of the ASCOT materials should contact the ASCOT team, who will put them in touch with Kent Innovation & Enterprise.

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## Appendix 6: Search strategy used in Chapter 2

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### **Cinahl via EBSCO**

All major headings apart from cost utility analysis, cost effectiveness analysis

1. TX All text: Dementia or Alzheimer's Disease
2. TX All text: Caregiver\* or care\*
3. TX All text: costs and cost analysis or cost benefit analysis or cost utility analysis or cost effectiveness analysis

#1 and #2 and #3

(((TX+(alzheimer's+disease))+or+(TX+(dementia)))+AND+((TX+(caregiver\*))+or+(TX+(care\*))) AND+((TX+(cost+benefit+analysis))+or+(TX+(costs+AND+cost+analysis))))

### **Web of Knowledge (Web of Science, Biosis and Medline)**

Topic= (dementia) or topic=(alzheimer) or topic= (alzheimer's disease)

1. Topic=(costs and cost analysis) OR Topic=(cost-benefit analysis) OR Topic=(cost benefit analysis) OR Topic=(cost utility analysis) OR Topic=(cost-effectiveness analysis) OR Topic=(cost effectiveness analysis) OR Topic=(cost-utility analysis)
2. Topic=(care\*) OR Topic=(caregiver\*)

### **PubMed Central (archive of PubMed, contains Medline)**

1. (dementia or alzheimer's disease)
2. (carer\* or caregiver\*)
3. (costs and cost analysis) or (cost benefit analysis) or (cost utility analysis) or (cost effectiveness analysis)

#1 and #2 and #3

### **PubMed**

1. (dementia or alzheimer's disease)
2. (carer\* or caregiver\*)
3. (costs and cost analysis) or (cost benefit analysis) or (cost utility analysis) or (cost effectiveness analysis)

#1 and #2 and #3

### **Science Direct**

Alzheimer, dementia, family caregiver, caregiver are topics

1. Dementia or Alzheimer or “alzheimer’s disease”
2. Caregiver\* or care\* or “family caregiver”
3. “costs and cost analysis” or “cost-benefit analysis” or “cost-effective analysis” or “cost-utility analysis”

#1 and #2 and #3

### **Cochrane Library inc NHS EED**

All mesh terms apart from cost-utility and cost-effectiveness

1. All text: Dementia or “Alzheimer Disease”
2. All text: Caregiver\* or care\*
3. All text: “costs and cost analysis” or “cost-benefit analysis” or “cost-utility analysis” or “cost-effectiveness analysis”

#1 and #2 and #3

### **CSA (contains Psychlit)**

No mesh terms, the below is the natural science breakdown

1. Anywhere: Dementia or Alzheimer’s Disease
2. Anywhere: Caregiver\* or care\*
3. Anywhere: costs and cost analysis or cost-benefit analysis or cost utility analysis or cost effectiveness analysis

#1 and #2 and #3

(dementia or alzheimer's disease) and(caregiver\* or care\*) and(((costs and cost analysis) or (cost-benefit analysis) or (cost effectiveness analysis)) or (cost utility analysis))

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## Appendix 7: Search strategy used in Chapter 3

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### **Pubmed/Medline:**

((("caregivers"[MeSH Terms] OR "care\*" [title] OR "care\*" [text]) AND ("dementia"[mesh terms] or "dementia"[title] or "dementia"[text] or "Alzheimer Disease"[Mesh terms] or "alzheimer\*" [title] or alzheimer\* [text]) AND ("clinical trial"[publication type] or "clinical trials as topic"[mesh terms] or "Randomized controlled trial"[text] or "randomized controlled trial"[title] or "clinical trial" [title] or "clinical trial" [text]))

### **CINAHL**

((caregivers) and ("randomized controlled trials" or "clinical trials") and (dementia or "Alzheimer's disease"))

[http://0-](http://0-search.ebscohost.com.unicat.bangor.ac.uk/login.aspx?direct=true&db=c8h&bquery=((caregivers)+AND+(%e2%80%9crandomized+controlled+trials%e2%80%9d+OR+%22clinical+trials%22)+AND+(dementia+OR+%e2%80%9cAlzheimer%e2%80%99s+disease%e2%80%9d))&cli0=LA1&clv0=Y&type=0&site=ehost-live)

search.ebscohost.com.unicat.bangor.ac.uk/login.aspx?direct=true&db=c8h&bquery=((caregivers)+AND+(%e2%80%9crandomized+controlled+trials%e2%80%9d+OR+%22clinical+trials%22)+AND+(dementia+OR+%e2%80%9cAlzheimer%e2%80%99s+disease%e2%80%9d))&cli0=LA1&clv0=Y&type=0&site=ehost-live

### **Centre for Reviews and Dissemination (NHS EED/ HTA/ DARE)**

(caregiver\*) or (carer\*)

(dementia) or (Alzheimer\*)

(clinical trial) or (trial)

#1 AND #2 AND #3

### **PsycINFO**

((Title:trial) OR (Subject:trial) OR (FullText:trial)) AND ((Title:dementia) OR (Subject:dementia) OR (FullText:dementia) OR (Title:alzheimer\*) OR (Subject:alzheimer\*) OR (FullText:alzheimer\*)) AND ((Title:carer\*) OR (Subject:carer\*) OR (FullText:carer\*) OR (Title:caregiver\*) OR (Subject:caregiver\*) OR (FullText:caregiver\*))

## Appendix 8: Full list of carer outcome measures extracted in Chapter 3

Category of measure	Instrument	# of studies	% of studies	Earliest date	Latest date
<b>Burden</b>	Zarit Burden Interview	76	21.1%	1994	2012
	Revised Memory and Behavior Problems Checklist (RMBPC)	44	12.2%	1994	2012
	Unspecified burden, behavioural problems and affect measure	37	10.2%	-	-
	Relatives Stress Scale	13	3.6%	1998	2011
	Novak Caregiver Burden Inventory	11	3.0%	2005	2011
	Perceived Stress Scale	11	3.0%	1996	2010
	Screen for Caregiver burden	11	3.0%	2000	2009
	(Revised) Caregiver Burden Scale	6	1.7%	1991	2009
	Caregiver Stress Scale	4	1.1%	2003	2007
	Lawton Caregiver Appraisal Measure	3	0.8%	1999	2010
	Unspecified stress measure	3	0.8%	-	-
	Bradburn Affect Scale	2	0.6%	1989	2003
	Burden Scale for Family caregivers (Grassel)	2	0.6%	2009	2010
	Carer Strain Index	2	0.6%	2003	2006
	Decisional Conflict scale	2	0.6%	2005	2011
	Family Caregiver Conflict scale	2	0.6%	2010	2011
	Gilleard Strain Scale	2	0.6%	1999	2002
	Modified Conflict Tactics Scale	2	0.6%	2006	2010
	Nursing home hassles scale	2	0.6%	2007	2011
	Apparent Affect rating scale	1	0.3%	2009	2009
	Behavior Management Scale–Revised	1	0.3%	2007	2007
	Burnout Measure	1	0.3%	1994	1994
	Caregiver Assessment of Functional Dependence and Caregiver Upset	1	0.3%	2003	2003
	Caregiver Bathing Behavior Rating Scale	1	0.3%	2006	2006
	Caregiver Burden caused by Behavioral and Psychological Symptoms of Dementia (CBBD)	1	0.3%	2007	2007
	Caregiver Strain (Rose)	1	0.3%	2003	2003
	Caregiver stress inventory	1	0.3%	2011	2011
	Caregiving Hassle Scale	1	0.3%	2001	2001
	Carers Assessment of Difficulties Index (CADI)	1	0.3%	2008	2008
	Cook-Medley Hostility Scale	1	0.3%	2010	2010



	Hassles During Bathing Scale	1	0.3%	2006	2006
	Home Care scale	1	0.3%	2005	2005
	Interpersonal Conflict Scale	1	0.3%	2007	2007
	Minimum Data Set for Home Care (MDS-HC)	1	0.3%	2008	2008
	Montgomery Borgotta Caregiver Burden scale	1	0.3%	2011	2011
	Multiple Affect Adjective Checklist (MAACL) Hostility and Depression subscales	1	0.3%	2003	2003
	Negative Reactions to Care Recipient Behaviour (Kinney)	1	0.3%	2011	2011
	NIVEL Scale for Perceived Problems in Dementia Care (NSPP-DC)	1	0.3%	2005	2005
	NIVEL Scale for Perceived Problems with Specific Behaviors of patients with dementia (NSPP-SB)	1	0.3%	2005	2005
	Organization and Stress Scale	1	0.3%	2005	2005
	Philadelphia Geriatric Centre Affect Rating Scale	1	0.3%	2008	2008
	Spielberger State-Trait Anger Inventory	1	0.3%	2010	2010
	Strains in Nursing Care scale	1	0.3%	2007	2007
	Stress Appraisal Measure	1	0.3%	2005	2005
	Whitlatch Burden Interview	1	0.3%	2003	2003
	Zarit stress scale	1	0.3%	1999	1999
<b>Mastery</b>	Unspecified self-efficacy and coping measure	22	6.1%	-	-
	Unspecified knowledge and mastery measure	20	5.5%	-	-
	Unspecified intervention satisfaction measure	17	4.7%	-	-
	Sense of Competence Questionnaire	12	3.3%	2000	2011
	Brief Coping Orientation for Problems Experienced (COPE)	6	1.7%	2006	2011
	Revised Scale for Caregiving Self Efficacy	6	1.7%	2003	2011
	Ways of coping scale	6	1.7%	1999	2007
	Unspecified confidence and self-esteem measure	5	1.4%	-	-
	Alzheimer's Disease Caregiver Preference Questionnaire	3	0.8%	2007	2011
	Alzheimer's Disease Knowledge Test	3	0.8%	1994	2008
	Beliefs about Caregiving Scale	3	0.8%	2001	2005
	Caregiver self-efficacy (Pearlin)	3	0.8%	1997	2012
	Carers' Assessment of Managing Index	2	0.6%	2005	2011
	General self-efficacy scale	2	0.6%	2005	2010
	Jalowiec Coping Scale	2	0.6%	2005	2006
	Philadelphia Geriatric Center Caregiving Appraisal	2	0.6%	2002	2009

	Scale				
	Symptom Management	2	0.6%	2009	2009
	Task Management Strategy Index	2	0.6%	2003	2008
	Agitation Management Self-efficacy Scale	1	0.3%	2003	2003
	Care Effectiveness Scale	1	0.3%	2006	2006
	Caregiving Mastery Index	1	0.3%	2003	2003
	Carers of Older People in Europe Index (COPE-Index)	1	0.3%	2011	2011
	Coping Resources Inventory	1	0.3%	2004	2004
	Coping Responses Inventory	1	0.3%	2010	2010
	Coping Strategies Inventory-Revised	1	0.3%	2000	2000
	Dementia Management Strategies Scale	1	0.3%	2008	2008
	Environmental Mastery scale	1	0.3%	2010	2010
	Expectation of Benefit Index	1	0.3%	2011	2011
	Geriatric Center Morale Scale	1	0.3%	2000	2000
	Goal Attainment Scale	1	0.3%	2011	2011
	Health Specific Family Coping Index	1	0.3%	1989	1989
	Indices of coping	1	0.3%	1999	1999
	Inventory of Geriatric Nursing Self-Efficacy	1	0.3%	2003	2003
	Knowledge of Alzheimer's Test	1	0.3%	1999	1999
	Knowledge of Services scale	1	0.3%	2011	2011
	Life-Events and difficulties	1	0.3%	2003	2003
	Locus of Control Scale	1	0.3%	2006	2006
	MacArthur Competency Assessment Tool for Clinical Research (MacCAT-CR)	1	0.3%	2006	2006
	Management of meaning (Pearlin)	1	0.3%	2010	2010
	Penn State Health Care-giving Questionnaire	1	0.3%	2009	2009
	Planning for Future Care Needs scale	1	0.3%	2011	2011
	Preparedness for Caregiving scale	1	0.3%	2011	2011
	Psychosocial Adjustment to Relatives Illness scale	1	0.3%	1999	1999
	Saunders and Courtney confidence-in-decision making	1	0.3%	1995	1995
	Steinmetz Control Scale	1	0.3%	2003	2003
	The Self-Efficacy of Dementia Care	1	0.3%	2007	2007
	Therapeutic engagement index	1	0.3%	2003	2003
<b>Mood</b>	Center for Epidemiologic Studies Depression Scale (CES-D)	57	15.8%	1989	2012
	General Health Questionnaire (GHQ)	31	8.6%	1987	2011

Neuropsychiatric Inventory-Distress (NPI-D)	30	8.3%	2000	2011
Geriatric Depression Scale	19	5.3%	1995	2011
Beck Depression Inventory (BDI)	17	4.7%	1998	2010
Neuropsychiatric Inventory-Questionnaire (NPI-Q)	12	3.3%	2006	2011
Brief Symptom Inventory	8	2.2%	1994	2007
Hamilton Depression Scale	8	2.2%	1989	2010
Pittsburgh Sleep quality Index	8	2.2%	2002	2011
Unspecified satisfaction with quality of care	8	2.2%	-	-
Hospital Anxiety and Depression Scale	7	1.9%	1998	2011
State-Trait Anxiety Inventory	7	1.9%	1991	2010
Unspecified depression	7	1.9%	-	-
Unspecified satisfaction and positive states of mind	7	1.9%	-	-
Positive and Negative Affect Scale (PANAS)	5	1.4%	1998	2011
Hopkins Symptoms Checklist	4	1.1%	1996	2009
Positive Aspects of Caregiving	4	1.1%	1997	2012
Unspecified life satisfaction	4	1.1%	-	-
Unspecified sleep	4	1.1%	-	-
Caregiver Satisfaction Survey	3	0.8%	1997	2004
Montgomery-Asberg Depression Rating Scale	3	0.8%	2002	2010
Patient Health Questionnaire	3	0.8%	2003	2008
Unspecified distress and upset	3	0.8%	-	-
Comfort Assessment in Dying	2	0.6%	2009	2009
Cornell Scale for Depression in Dementia	2	0.6%	2003	2010
Delusions Symptoms States Inventory/States of Anxiety and Depression (DSSI)	2	0.6%	1987	1990
Profile of Moods States (POMS)	2	0.6%	1999	2002
SATMED-Q	2	0.6%	2009	2009
Self Reporting Questionnaire-20	2	0.6%	2007	2009
Self-Assessing Depression Scale (SADS)	2	0.6%	1997	2009
Unspecified grief	2	0.6%	-	-
Unspecified guilt	2	0.6%	-	-
Befindlichkeits-Skala	1	0.3%	2000	2000
Camberwell Family Interview	1	0.3%	2002	2002
Caregiver Distress About Night-time Activity	1	0.3%	2010	2010
Caregiver Distress Scale	1	0.3%	2003	2003
Carers' Assessment of Satisfaction Index	1	0.3%	2008	2008
Carlsson Visual Analogue Scale	1	0.3%	2011	2011
Depression, Anxiety and Stress Scale (DASS-21)	1	0.3%	2008	2008

	Dysfunctional Thoughts about Caregiving	1	0.3%	2010	2010
	Epworth Sleep Questionnaire	1	0.3%	2010	2010
	Eysenck Personality Questionnaire revised	1	0.3%	2011	2011
	Family caregiver distress (Pearlin)	1	0.3%	2005	2005
	General Sleep Disturbance Scale	1	0.3%	2009	2009
	Ilfeld Psychiatric Symptoms Index	1	0.3%	2003	2003
	Impact of event	1	0.3%	2009	2009
	Inventory of Complicated Grief	1	0.3%	2003	2003
	Kessler distress scale	1	0.3%	2011	2011
	Kessler-6	1	0.3%	2008	2008
	Leisure Time Satisfaction	1	0.3%	2003	2003
	Marwit and Meuser Caregiver Grief Inventory-Short Form (MMCGI-SF)	1	0.3%	2010	2010
	Mental Health Index	1	0.3%	2006	2006
	Morin Daily Sleep Diary	1	0.3%	2006	2006
	NEO Personality Inventory (NEO-PI)	1	0.3%	2011	2011
	Night Time Activity Worry – Scale	1	0.3%	2012	2012
	Ohio Department of Aging Family Satisfaction Instrument	1	0.3%	2011	2011
	Penn State Mental Health Questionnaire-20	1	0.3%	1999	1999
	Penn State Worry Questionnaire	1	0.3%	2009	2009
	Poulshok and Deimling cognitive subscale	1	0.3%	2000	2000
	Prolonged Grief Disorder-12	1	0.3%	2008	2008
	Psychological Distress Index	1	0.3%	2005	2005
	Satisfaction with Care at End-of-Life in Dementia	1	0.3%	2008	2008
	Schedule for the Evaluation of Individualized Quality of Life	1	0.3%	2006	2006
	State-Trait Personality Inventory	1	0.3%	2003	2003
	Swiss health survey	1	0.3%	2006	2006
	Symptom Check List 90 Revised	1	0.3%	1999	1999
	Taylor Manifest Anxiety Scale	1	0.3%	2002	2002
	Texas Revised Inventory of Grief	1	0.3%	2003	2003
	Zung depression scale	1	0.3%	1989	1989
<b>Quality of life</b>	Short Form-36 (SF-36)	32	8.9%	2001	2011
	EuroQoL (EQ-5D)	18	5.0%	2001	2011
	Unspecified health/quality of life	11	3.0%	-	-
	World Health Organization Quality of Life-Bref (WHOLQOL-BREF)	8	2.2%	2007	2011

	Health Utilities Index(HUI)	4	1.1%	1999	2010
	Health Status Questionnaire	3	0.8%	2003	2011
	Caregiver Health and Health Behaviours (Posner)	2	0.6%	2003	2007
	Duke Health Profile	2	0.6%	2005	2010
	Nottingham Health Profile	2	0.6%	2008	2010
	Perceived Change Index	2	0.6%	2003	2010
	Philadelphia Geriatric Centre multilevel assessment	2	0.5%	1989	2001
	Beschwerdeliste	1	0.3%	2009	2009
	Caregiver Quality of Life Instrument (Mohide)	1	0.3%	1991	1991
	CarerQol-7D	1	0.3%	2009	2009
	CQOL (Logsdon)	1	0.3%	2010	2010
	Cumulative Illness Rating Scale	1	0.3%	2010	2010
	DQOL	1	0.3%	2008	2008
	General Medical Health Rating	1	0.3%	2003	2003
	General Well-being Adjustment Scale (Brook)	1	0.3%	2001	2001
	General Well-Being scale	1	0.3%	2003	2003
	General well-being schedule	1	0.3%	2011	2011
	Giessen physical health	1	0.3%	2005	2005
	Health Assessment Scale	1	0.3%	2000	2000
	Norderstedt	1	0.3%	2004	2004
	Personal Well-being Index	1	0.3%	2010	2010
	Physical Activity Question Score (Voorrips)	1	0.3%	2011	2011
	Quality of Life questionnaire (Ruiz)	1	0.3%	2002	2002
	Rapid Assessment of Physical Activity scale	1	0.3%	2011	2011
	Ruiz and Baca's Questionnaire-39	1	0.3%	2010	2010
	Siegrist Profil der Lebensqualität chronisch (PLC)	1	0.3%	2005	2005
	Symptom Questionnaire	1	0.3%	2004	2004
	Well-Being Scale	1	0.3%	2001	2001
	World Health Organisation's Alcohol Use Disorders Identification Test	1	0.3%	2011	2011
<b>Social support and relationships</b>	Unspecified social support and relationships measure	20	5.5%	-	-
	Social Support Questionnaire	7	1.9%	1999	2011
	Stokes Social Support network List	4	1.1%	2006	2010
	Inventory of Socially Supportive Behaviours	3	0.8%	2003	2011
	Multidimensional scale of perceived social support	3	0.8%	2008	2011
	Quality of the carer-patient relationship	3	0.8%	2009	2011
	Unspecified loneliness measure	3	0.8%	-	-
	Duke Social support questionnaire	2	0.6%	1999	2010

	Perceived Social Support scale (Pearlin)	2	0.6%	2002	2002
	Practitioner Assessment of Network Type	2	0.6%	2008	2011
	Social Provisions Scale	2	0.6%	2002	2011
	Clifton Assessment Procedures for Elderly Behavioural Rating Scale (CAPE-BRS)	1	0.3%	1999	1999
	Family APGAR questionnaire	1	0.3%	2010	2010
	Family Involvement scale	1	0.3%	2007	2007
	Family Support Services Index	1	0.3%	2011	2011
	Interpersonal Social Evaluation List	1	0.3%	2002	2002
	Lubben Social Network Index	1	0.3%	2003	2003
	Marital Needs Satisfaction Scale	1	0.3%	2000	2000
	Modified Social Support (Barrera)	1	0.3%	2003	2003
	Moses	1	0.3%	2008	2008
	Mutual Communal behaviours Scale	1	0.3%	2008	2008
	Mutuality Scale of the Family Care Inventory	1	0.3%	2011	2011
	Personal Resource Questionnaire (PRQ-85)	1	0.3%	2004	2004
	Positive and Negative Social Exchanges	1	0.3%	2011	2011
	Received Social Support scale	1	0.3%	2007	2007
	Relationship Attribution Measure	1	0.3%	2005	2005
	Sherbourne Social Support	1	0.3%	2007	2007
	Systematic Care Program for Dementia	1	0.3%	2009	2009
	Thomas Assessment of communication inadequacy	1	0.3%	2001	2001
<b>Staff competency and morale</b>	Maslach Burnout Inventory	10	2.6%	2004	2011
	Unspecified job satisfaction and competence measure	7	1.8%	-	-
	Approaches to Dementia Questionnaire	4	1.1%	2005	2012
	Modified Nursing Care Assessment scale M-NCAS	3	0.8%	2004	2011
	Blenkner (relationship between nurses and person with dementia)	1	0.3%	2004	2004
	Canadian occupational performance measure	1	0.3%	2011	2011
	Creative Climate Questionnaire	1	0.3%	1994	1994
	Experience and Assessment of work (VBBA)	1	0.3%	2011	2011
	Family behaviours and empathy scale	1	0.3%	2007	2007
	Formal Caregivers' Attitude toward Feeding Dementia Patient Questionnaire	1	0.3%	2005	2005
	Formal Caregivers' Behaviours toward Feeding Dementia Patients Observation Checklist	1	0.3%	2005	2005
	Formal Caregivers' Knowledge of Feeding Dementia	1	0.3%	2005	2005

Patient Questionnaire				
Iowa Dependent Adult Abuse Nursing Home Questionnaire	1	0.3%	2010	2010
Kazdin's Treatment Evaluation Inventory	1	0.3%	1995	1995
Knowledge and Management of Abuse	1	0.3%	2010	2010
Leiden Quality of Work Questionnaire	1	0.3%	2011	2011
Maastricht Work Satisfaction Scale for Healthcare	1	0.3%	2005	2005
Sense of Competence in Dementia care Staff (SCIDS)	1	0.3%	2012	2012
STAR Staff Feedback Questionnaire (SSFQ)	1	0.3%	2012	2012
Stress screening of human services (nurses' occupational stress)	1	0.3%	2009	2009
Subjective work pressure (Potentialanalyse stationärer Altenpflege - PASTA)	1	0.3%	2010	2010
Work Stress Inventory	1	0.3%	2005	2005

---

## Appendix 9: Interview schedule used in Chapter 4

---

### **Opening**

Please remember that what you tell me today will remain confidential. Your name will not appear on the interview notes that I am making today or in any reports. The information collected will be used as part of a PhD about caregiving for people with dementia.

It would be helpful if we could record this information so that the information we collect is as accurate and complete as possible.

If you have any questions please do not hesitate to ask. You may stop the interview at any time that you wish.

### **Framing**

I would like to ask you some questions about yourself and your experience of caregiving in order to learn more about your role as a partner/child of somebody with dementia and how it affects your daily life.

### **Phase 1 (Uninterrupted)**

1. Please can you start by telling me about your life around the time when your partner/parent was diagnosed, mentioning events and experiences that you found important.

Please begin wherever you like, I won't interrupt you but I will take some notes for after you have finished telling me your story.

### **Phase 2 (Cue phrases)**

I noted down some key points as you were telling me your story and I would like to go back and talk about these some more.

2. You said (...). Can you remember a particular (key word) when it happened?

Key words: situation, event, incident, occasion, time, day, memory of a moment, example.

If the carer can't remember ask 'Can you remember a time when (...) did not happen?'



### **Phase 3 (specific questions)**

You have kindly told me your story, about your life and your experiences. I now have some specific questions that I would like to ask you.

3. Can you tell me about how your life changed once the illness progressed?
4. Moving forward to the present, can you tell me about your life now?
5. Looking to the future, how do you see your life changing, if at all?
6. Can you tell me a little about your experience of health and social services?
7. Based on your experience, can you suggest anything that would improve service provision for you and your partner/parent in the future?

### **Prompts**

Can you tell me a bit more about (...)?

You talked about (...); can you tell me more about that?

Can you think of an example of (...)?

Does anything else come to mind?

### **Closing**

I should have all the information I need, is there anything else you think would be helpful for me to know about caring and how it affects you?





Well, it has been a pleasure finding out more about you. The conversation will remain confidential with no names appearing in the write-up.

I appreciate the time you took for this interview. Thanks again.

---

## Appendix 10: Ethical approval for validity survey used in Chapter 5

---

From Bangor Research Applications <psse09@bangor.ac.uk>★	 Reply	 Reply All	 Forward	 Archive
Subject: <b>Ethics Application Approved</b>	18/05/2011 15:41			
To: mhpa01@bangor.ac.uk★	Other Actions ▾			

Dear Carys Llywela,

2011-3088 Assessing the validity of the ICECAP-O measure for use with informal carers of people with dementia

Your research proposal number 2011-3088 has been reviewed by the School of Psychology Ethics and Research Committee and the committee are now able to confirm ethical and governance approval for the above research on the basis described in the application form, protocol and supporting documentation. This approval lasts for a maximum of five years from this date.

Ethical approval is granted for the study as it was explicitly described in the application

If you wish to make any non-trivial modifications to the research project, please submit an amendment form to the committee, and copies of any of the original documents reviewed which have been altered as a result of the amendment. Please also inform the committee immediately if participants experience any unanticipated harm as a result of taking part in your research, or if any adverse reactions are reported in subsequent literature using the same technique elsewhere.

.

Governance approval is granted for the study as it was explicitly described in the application and we are happy to confirm that this study is now covered by the University's indemnity policy.

If any new researchers join the study, or any changes are made to the way the study is funded, or changes that alter the risks associated with the study, then please submit an amendment form to the committee.

Yours sincerely

Everil McQuarrie

---

## Appendix 11: Validity survey used in Chapter 5, postal version

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### Information Sheet

You are being invited to take part in a research survey, the results of which will be used as part of a PhD project. Before you decide, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully. Ask if there is anything that is not clear or if you would like more information.

What is the purpose of this study?

Family caregivers are known to be a vital source of support for people with dementia who live at home. Health professionals are increasingly recognising the importance of considering the needs of the carer as well as their relative and questionnaires are a commonly used method of measuring health status and quality of life.

This study seeks to assess the strength of a newly developed questionnaire, the ICECAP\_O (ICEpop CAPability measure for Older people). This questionnaire has been developed to measure quality of life for members of the general population aged 65 and over. We are asking you to complete the questionnaire to see how suitable it is for use with carers of people with dementia. The information collected from this survey will form part of a PhD project assessing the use of different quality of life questionnaires in studies involving carers of people with dementia.

The survey will consist of a number of questions relating to your quality of life and living situation. The survey should take no more than 15 minutes to complete. Each person can only complete the survey once, but if you share caregiving with another person they are able to take part in the survey.

All survey responses received by 31/12/2011 will be entered into a prize draw to win a £250 Marks and Spencer voucher.

Participation in the study is entirely voluntary, and you are free to refuse to take part or withdraw at any time, without giving a reason. Data will remain confidential and participants will not be named or identifiable in the notes or subsequent write ups.

If you have any questions, or require further information please contact:

Carys Jones  
Centre for Health Economics and Medicines Evaluation  
FREEPOST BG35  
Institute of Medical and Social Care Research  
Dean Street  
Bangor, Gwynedd  
LL57 1UT

Email: [c.l.jones@bangor.ac.uk](mailto:c.l.jones@bangor.ac.uk) Telephone: 01248 38 2483

The survey can also be completed online: <http://www.surveymonkey.com/s/Carersurvey>

Thank you for considering taking part in this research study.

Consent

Thank you for agreeing to take part in this research study. In the case of any complaints concerning the conduct of research, these should be addressed to Professor Oliver Turnbull, Head of School of Psychology, Brigantia Building, Bangor University, LL57 2AS.

Please tick the boxes below to confirm that you are willing to take part in the survey:

- I agree to participate in this research study.....
- I confirm that I have read and understand the information sheet .....
- I understand that my participation is voluntary and that I am free to withdraw at any time without giving a reason .....
- I understand that if I choose to leave my contact details they will be stored on a confidential database.....
- I understand that all the information I provide will be treated as strictly confidential and I will not be identifiable in any notes or subsequent publishing.....
- I understand that if I have any questions at any point I can contact the researcher via c.l.jones@bangor.ac.uk or the details given on the information sheet.....

Eligibility

The following questions are to determine whether or not you are eligible to take part in the survey. Please circle as applicable.

*I am aged 65 years or over:* YES/ NO

*I would describe myself as a current or former carer of a person with dementia:* I am a current carer/ I am a former carer

Quality of life questionnaire 1

By placing a tick in one box in each group below, please indicate which statements best describe your own health state today.

**Mobility**

- I have no problems in walking about
- I have some problems in walking about
- I am confined to bed

**Self-Care**

- I have no problems with self-care
- I have some problems washing or dressing myself
- I am unable to wash or dress myself

**Usual Activities** (e.g. work, study, housework, family or leisure activities)

- I have no problems with performing my usual activities
- I have some problems with performing my usual activities
- I am unable to perform my usual activities

**Pain/Discomfort**

- I have no pain or discomfort
- I have moderate pain or discomfort
- I have extreme pain or discomfort

**Anxiety/Depression**

- I am not anxious or depressed
- I am moderately anxious or depressed
- I am extremely anxious or depressed

Quality of life questionnaire 2

<b>ABOUT YOUR QUALITY OF LIFE</b>	
By placing a tick (✓) in ONE box in EACH group below, please indicate which statement best describes your quality of life at the moment.	
<b>1. Love and Friendship</b>	
I can have all of the love and friendship that I want	<input type="checkbox"/> 4
I can have a lot of the love and friendship that I want	<input type="checkbox"/> 3
I can have a little of the love and friendship that I want	<input type="checkbox"/> 2
I cannot have any of the love and friendship that I want	<input type="checkbox"/> 1
<b>2. Thinking about the future</b>	
I can think about the future without any concern	<input type="checkbox"/> 4
I can think about the future with only a little concern	<input type="checkbox"/> 3
I can only think about the future with some concern	<input type="checkbox"/> 2
I can only think about the future with a lot of concern	<input type="checkbox"/> 1
<b>3. Doing things that make you feel valued</b>	
I am able to do all of the things that make me feel valued	<input type="checkbox"/> 4
I am able to do many of the things that make me feel valued	<input type="checkbox"/> 3
I am able to do a few of the things that make me feel valued	<input type="checkbox"/> 2
I am unable to do any of the things that make me feel valued	<input type="checkbox"/> 1
<b>4. Enjoyment and pleasure</b>	
I can have all of the enjoyment and pleasure that I want	<input type="checkbox"/> 4
I can have a lot of the enjoyment and pleasure that I want	<input type="checkbox"/> 3
I can have a little of the enjoyment and pleasure that I want	<input type="checkbox"/> 2
I cannot have any of the enjoyment and pleasure that I want	<input type="checkbox"/> 1
<b>5. Independence</b>	
I am able to be completely independent	<input type="checkbox"/> 4
I am able to be independent in many things	<input type="checkbox"/> 3
I am able to be independent in a few things	<input type="checkbox"/> 2
I am unable to be at all independent	<input type="checkbox"/> 1

Tick  
one  
box  
only in  
each  
section

Background information

1. What is your relationship to the person with dementia that you care(d) for?

Spouse/ Son/ Daughter/ Son-in-law/ Daughter-in-law/ Brother/ Sister/ Other relative/  
Friend/ Neighbour

2. What is your gender?

Male/ Female

3. What is the gender of the person that you care(d) for?

Male/ Female

4. What is your date of birth?.....

5. Which of the following best describes your current employment status?

Full time employment/ Full time self-employed/  
Part time employment/ Part time self-employed/  
Unemployed/ Homemaker/ Retired  
Other (please specify).....

6. How would you describe your health in general?

Excellent/ Good/ Fair/ Poor/ Very poor

7. What type of accommodation do you normally live in?

Owner occupied house or flat

Privately rented house or flat

House or flat rented from housing association or local authority

Residential or Nursing home

18. If you are a current carer, does the person that you care for live with you?

YES/ NO/ N/A I am not a current carer

19. What type of accommodation does the person that you care(d) for normally live in?

Owner occupied house or flat

Privately rented house or flat

House or flat rented from housing association or local authority

Residential home

Nursing home

N/A, the person that I cared for is now deceased

20. On a typical day, how much of the time can you leave the person that you care(d) for alone?

Not at all

An hour or two

Up to half a day

Up to a whole day

Overnight

N/A, the person that I cared for is now deceased

21. How many hours do you spend each week performing care tasks for your relative?  
Please leave blank if you are not a current carer.....

22. The questions on page 4 were developed to describe and measure quality of life. Are there any important factors that affect quality of life that you think are missing and should be included? To remind you, questions were asked about love and friendship, thinking about the future, doing things that make you feel valued, enjoyment and pleasure, and independence.....

23. Do you think that any of the quality of life questions were not relevant to you, and should not be included?



24. Were the instructions for the questionnaire clear?

YES/ NO

25. If you have any additional comments please enter them below.

26. Thank you for completing the survey. If you would like to be entered into the prize draw for a £250 Marks and Spencer voucher drawn on 10/01/12 please enter your contact details below.

Name.....

Email.....

Telephone.....

27. Once all responses have been collected and analysed, would you like to receive a brief summary of findings? If yes, please leave your name and email address.

Name.....

Email.....

---

Appendix 12: Press release and cuttings advertising the validity survey used in  
Chapter 5

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Press release:

**University researchers seek feedback from older carers of people with dementia**

The views of people aged 65 and over who have experience of looking after somebody with dementia are being sought by researchers at Bangor University. All people responding to the survey by 31/12/11 will be entered into a prize draw to win a £250 Marks and Spencer voucher.

Professor Rhiannon Tudor Edwards and colleagues are carrying out a short survey as part of a project that aims to add to the evidence validating a quality of life measure for use in economic evaluations.

She said: “This survey will help healthcare professionals improve understanding of what affects quality of life for friends and family carers of people with dementia. Many current measures focus on physical issues, underestimating the effects on carer well-being and leading to underfunding of programmes designed to support carers. We are investigating a measure which has been designed to capture effects on broader well-being, such as companionship and independence.”

“The survey is being conducted across the UK, and we hope as many people as possible will take part.”

If you are aged 65 and over, and are a friend or family member of somebody with dementia you are invited to take part in the survey to share your views and experiences of caring.

Access to the survey is via <http://www.surveymonkey.com/s/Carersurvey> , further information or postal copies are available from Carys Jones on 01248 382483.

The screenshot shows a web browser window with the URL <http://www.newswales.co.uk/?section=Health&F=1&id=23420>. The page features the NewsWales logo with a yellow flower and the tagline "Your daily Welsh news briefing". A navigation menu includes categories like HOME, AGRICULTURE, BUSINESS, COMMUNITY, CULTURE, EDUCATION, ENVIRONMENT, HEALTH, LOCAL GOVT., MEDIA, POLITICS, SPORT, TOURISM, and TRANSPORT. A search bar is present on the left. The main article is titled "Bangor survey on dementia" and is dated 17 Dec 2011. The article text discusses a survey by Bangor University on dementia care, mentioning Professor Rhiannon Tudor Edwards and the survey's focus on quality of life measures. A Booking.com advertisement for Howard Johnson Washington District is also visible on the left side of the page.

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## Bangor survey on dementia

Section **Health** | Published on 17 Dec 2011

People aged 65 and over who have experience of looking after somebody with dementia can contribute towards research currently being carried out by academics at Bangor University.

Health economists there are investigating the economic demands of caring for people with dementia. The results of their research will contribute towards shaping health and social care policies across the UK.

All people responding to the survey by 31/12/11 will be entered into a prize draw to win a £250 Marks and Spencer voucher.



PRIFYSGOL BANGOR UNIVERSITY

Professor Rhiannon Tudor Edwards and colleagues are carrying out the short survey as part of a project that aims to assess the use of a new quality of life measure for use in economic evaluations.

She said: "This survey will help healthcare professionals improve understanding of what affects quality of life for friends and family carers of people with dementia.

"Many current measures focus on physical issues, underestimating the effects on carer well-being and leading to underfunding of programmes designed to support carers.

"We are investigating a measure which has been designed to capture effects on broader well-being, such as companionship and independence."

"The survey is being conducted across the UK, and we hope as many people as possible will take part."


People aged 65 and over, who are a friend or family member of somebody with dementia are invited to take part in the survey to share their views and experiences of caring.

Access to the survey is via <http://www.surveymonkey.com/s/Carersurvey>, further information or postal copies are available from Carys Jones on 01248 382483.

Published in the Chronicle newspaper 22/11/2011:



Published in the NEURODEM newsletter 09/02/2012:



*Carys Jones, PhD student*

## FOCUSING ON THE WELL-BEING OF OLDER CARERS

As part of a PhD focusing on supporting quality of life for family carers, Bangor University is currently conducting a short survey to investigate the suitability of well-being measures used in research involving older carers.

Many current measures focus on physical issues, underestimating the effects on well-being and leading to underfunding of programmes designed to support carers. This research study is comparing a brief physical health questionnaire to a questionnaire about broader well-being, such as companionship and security. The results will help healthcare professionals improve understanding of what

affects quality of life for friends and family carers of people with dementia, and aims to increase the evidence to support the view that the mental well-being of carers should be considered as well as physical health.

The study began in June 2011 and we are very pleased with the number of members of the NEURODEM Research Participant Register who have taken part so far. The survey is open until 30/04/2012 and we would like to invite readers who have not already participated to complete the survey. The survey takes approximately 15-20 minutes.

To be eligible to take part

in the study, participants must be aged 65 or over and be a current or former carer for somebody with dementia. The survey can be completed online at <http://www.surveymonkey.com/s/Carersurvey>.

If you are interested in finding out more about our study or want a postal copy, please call us on 01248 382483 or email us at [c.l.jones@bangor.ac.uk](mailto:c.l.jones@bangor.ac.uk)

**Carys Jones,**  
**PhD student, Bangor University**

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Appendix 13: Histograms and detrended normal Q-Q plots for measures used in  
Chapter 6

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Fig 1: Histogram for ICECAP, intervention group (normally distributed)

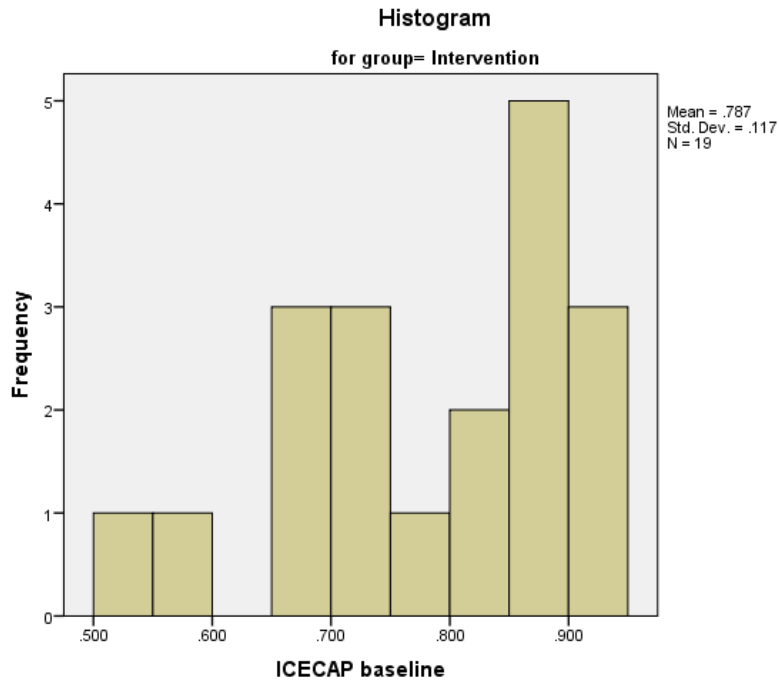


Fig 2: Histogram for ICECAP, control group (not normally distributed)

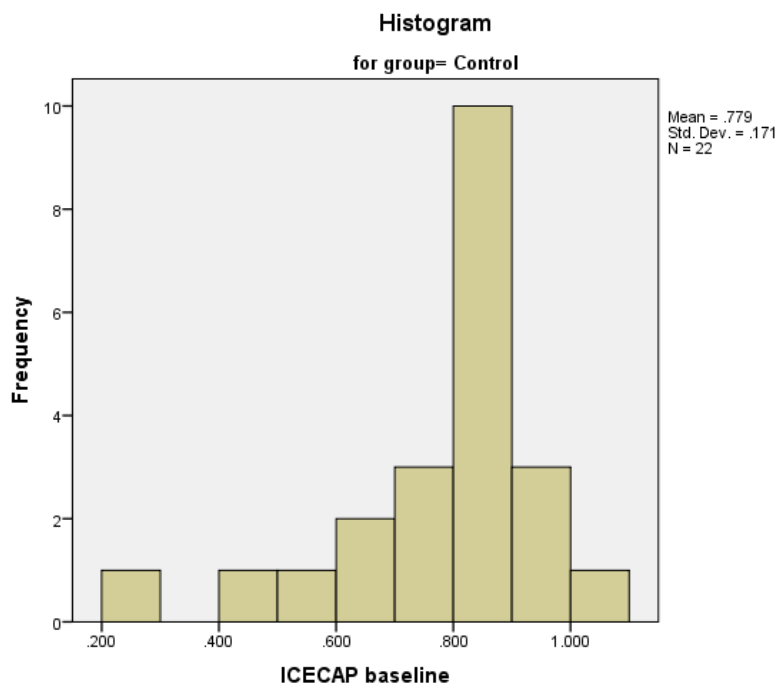


Fig 3: Detrended normal Q-Q plot for ICECAP, intervention group (normally distributed)

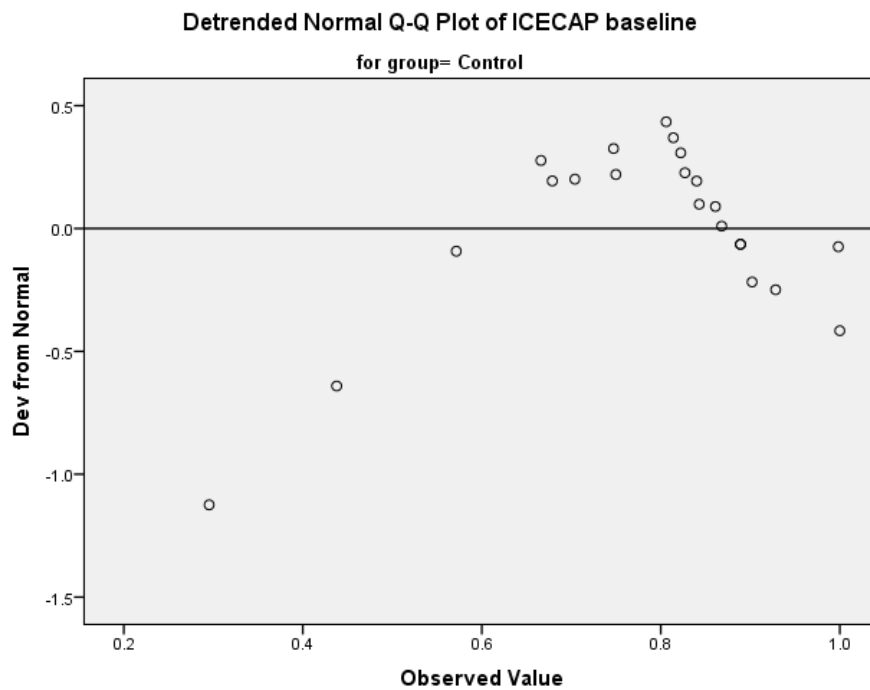


Fig 4: Detrended normal Q-Q plot for ICECAP, control group (not normally distributed)

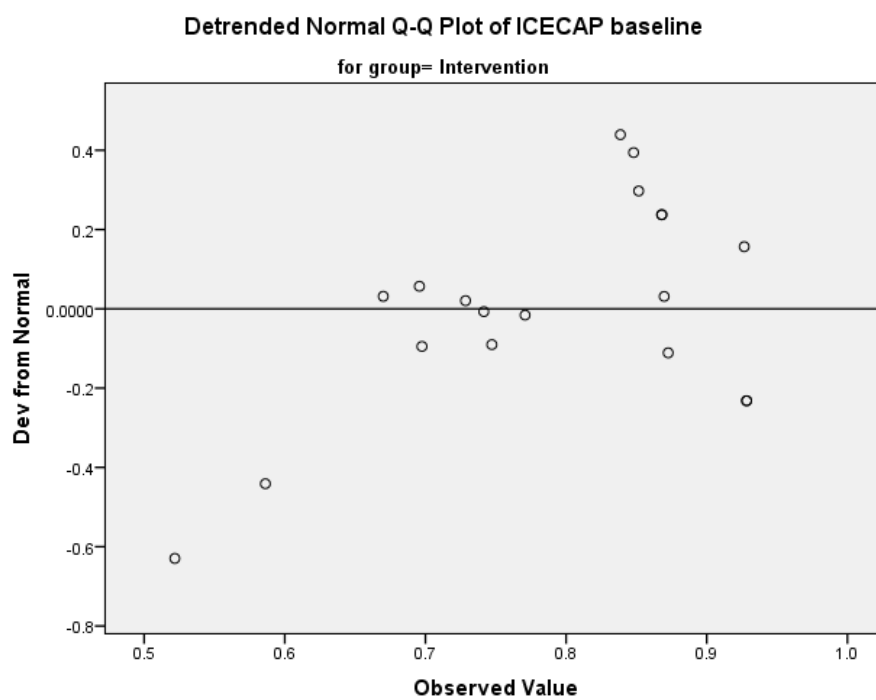


Fig 5: Histogram for EQ-5D, intervention group (normally distributed)

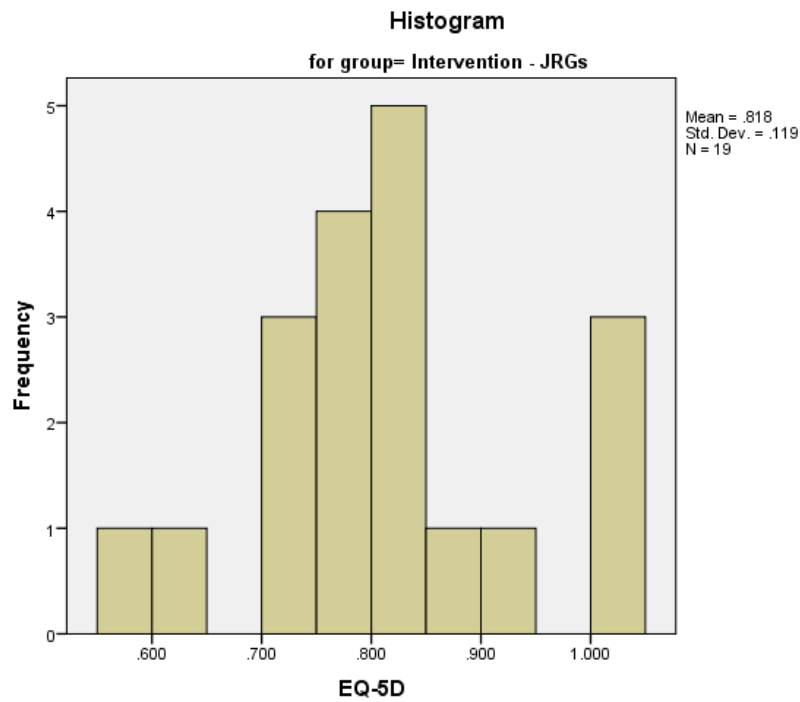


Fig 6: Histogram for EQ-5D, control group (normally distributed)

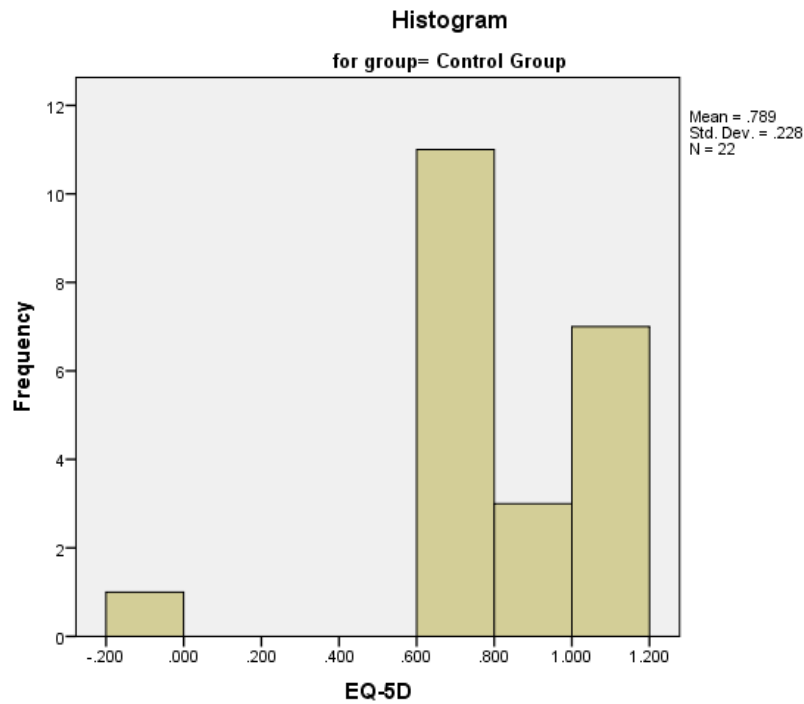


Fig 7: Detrended normal Q-Q plot for EQ-5D, intervention group (normally distributed)

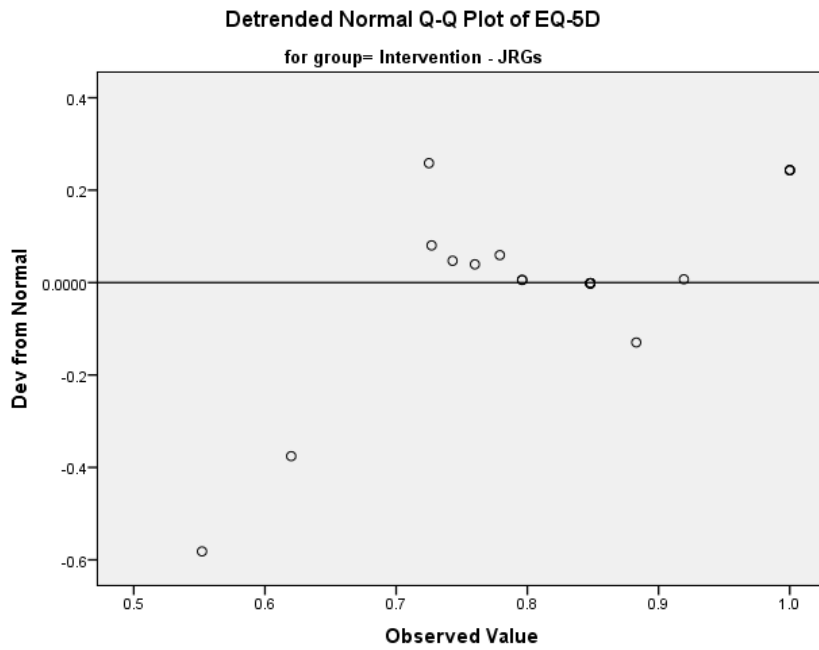


Fig 8: Detrended normal Q-Q plot for EQ-5D, control group (normally distributed)

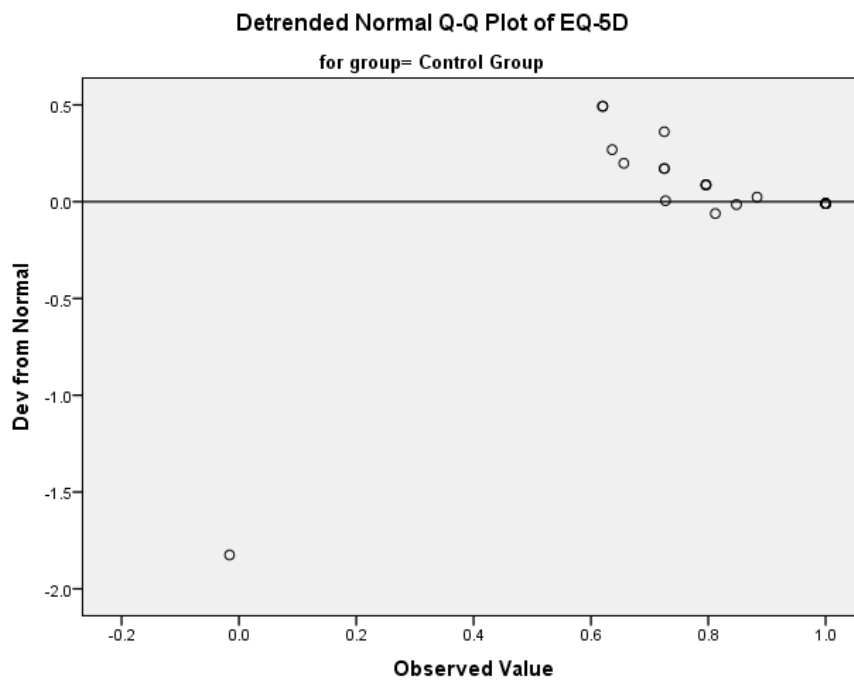




Fig 9: Histogram for EQ-VAS, intervention group (not normally distributed)

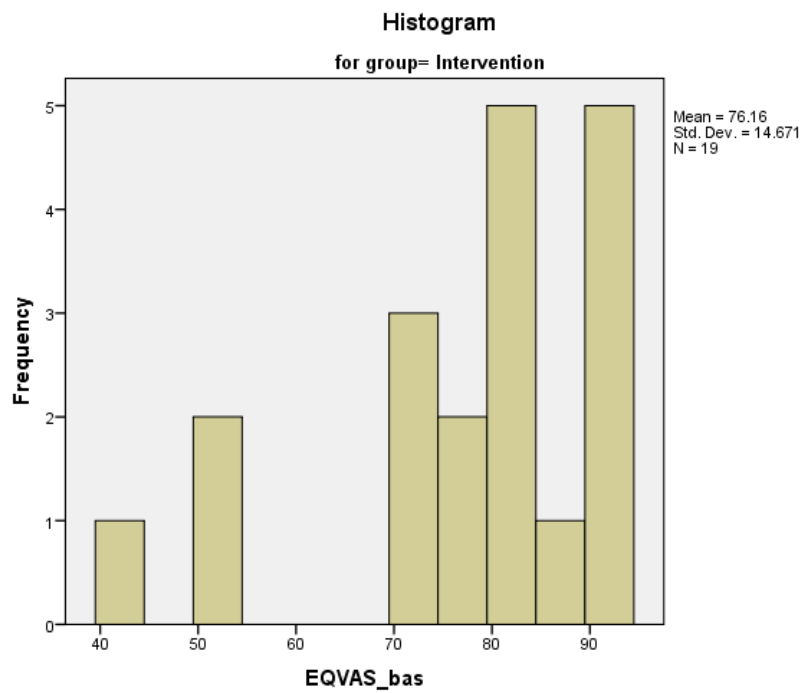


Fig 10: Histogram for EQ-VAS, control group (normally distributed)

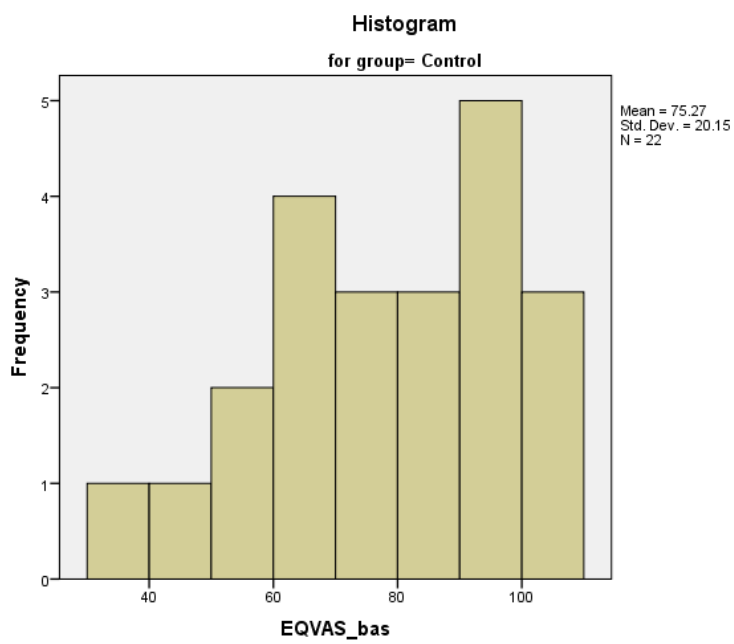


Fig 11: Detrended normal Q-Q plot for EQ-VAS, intervention group (not normally distributed)

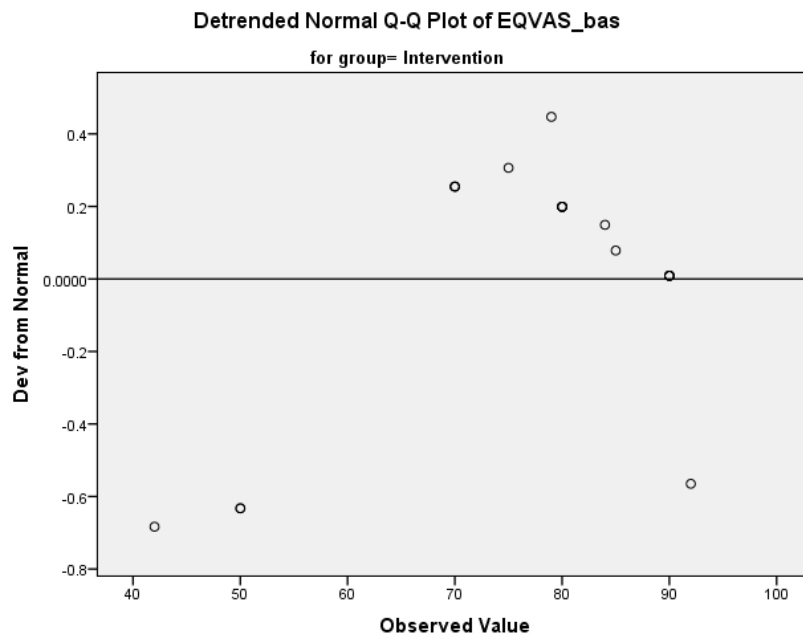


Fig 12: Detrended normal Q-Q plot for EQ-VAS, control group (normally distributed)

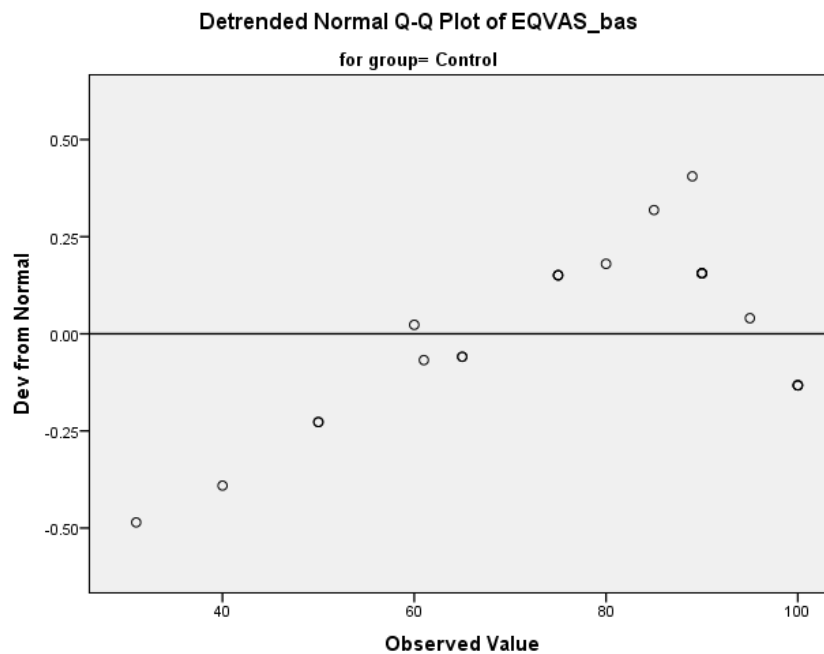


Fig 13: Histogram for HADS-anxiety, intervention group (normally distributed)

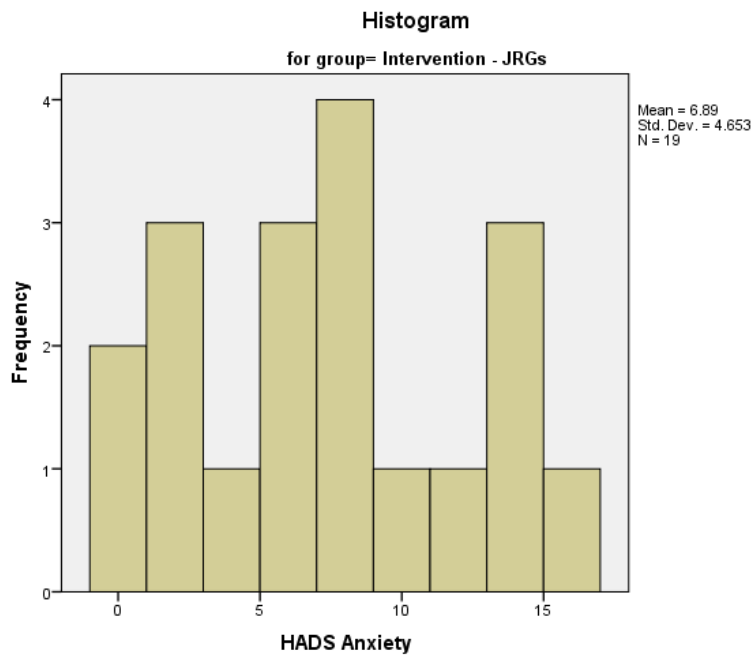


Fig 14: Histogram for HADS-anxiety, control group (normally distributed)

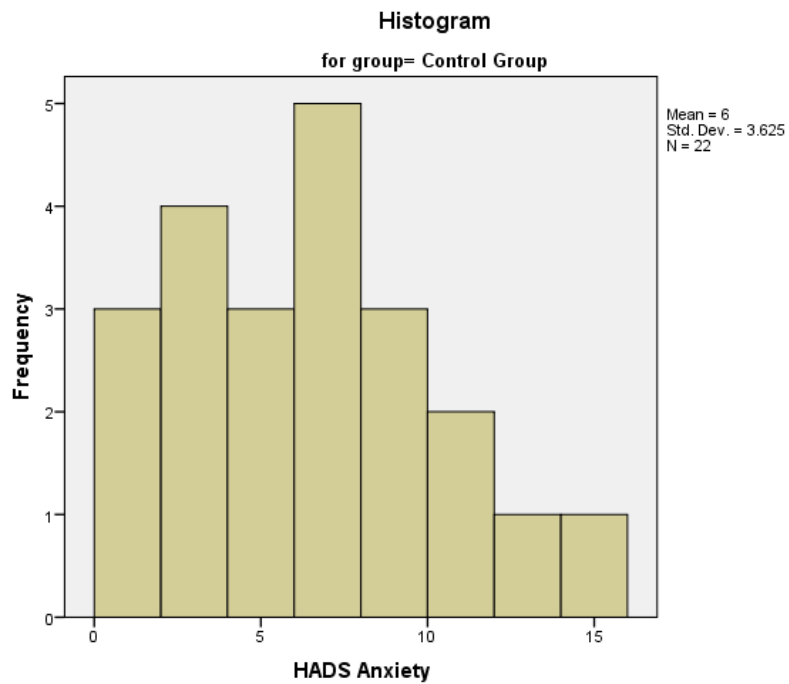


Fig 15: Detrended normal Q-Q plot for HADS-anxiety, intervention group (normally distributed)

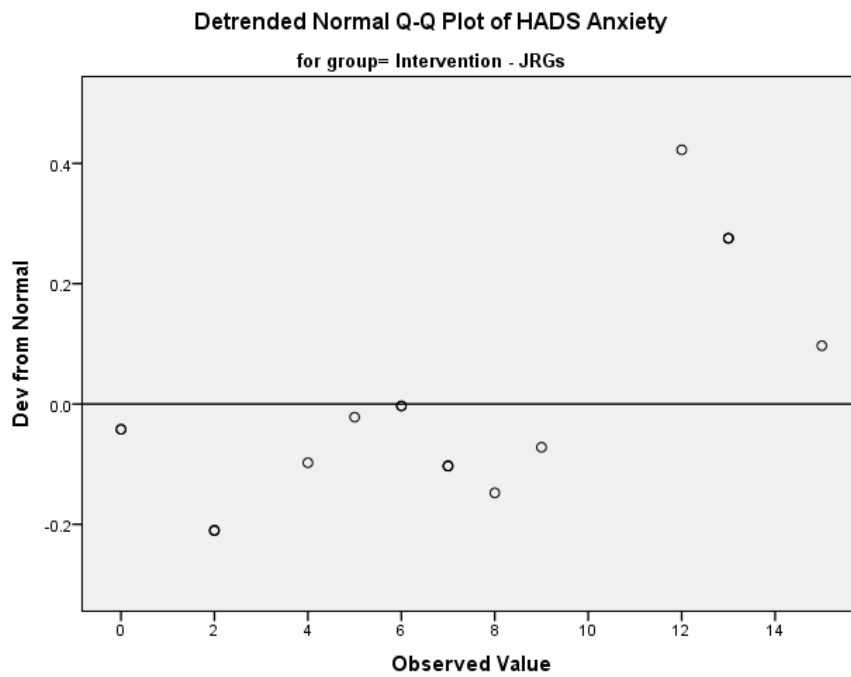


Fig 16: Detrended normal Q-Q plot for HADS-anxiety, control group (normally distributed)

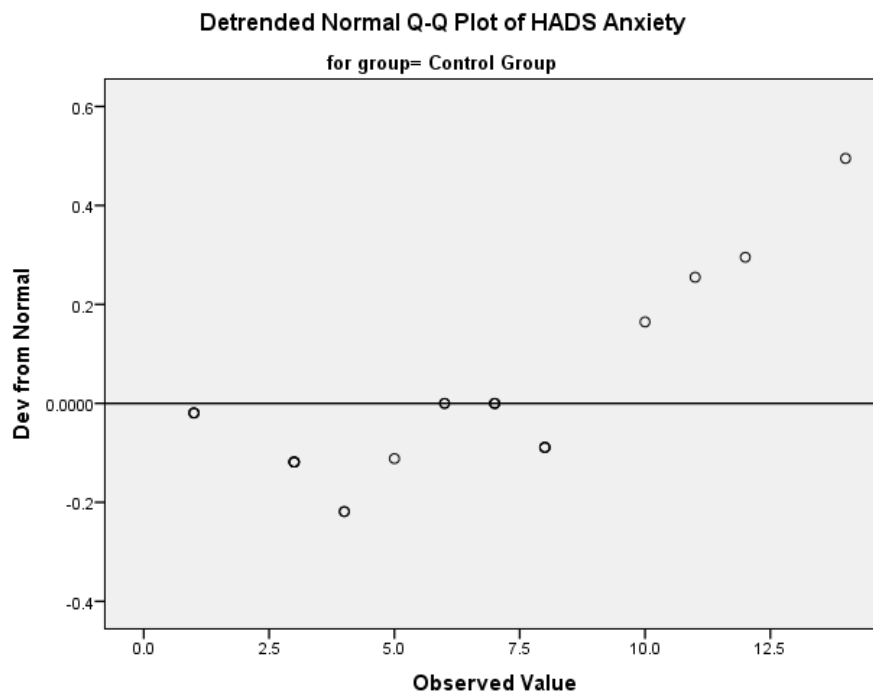


Fig 17: Histogram for HADS-depression, intervention group (not normally distributed)

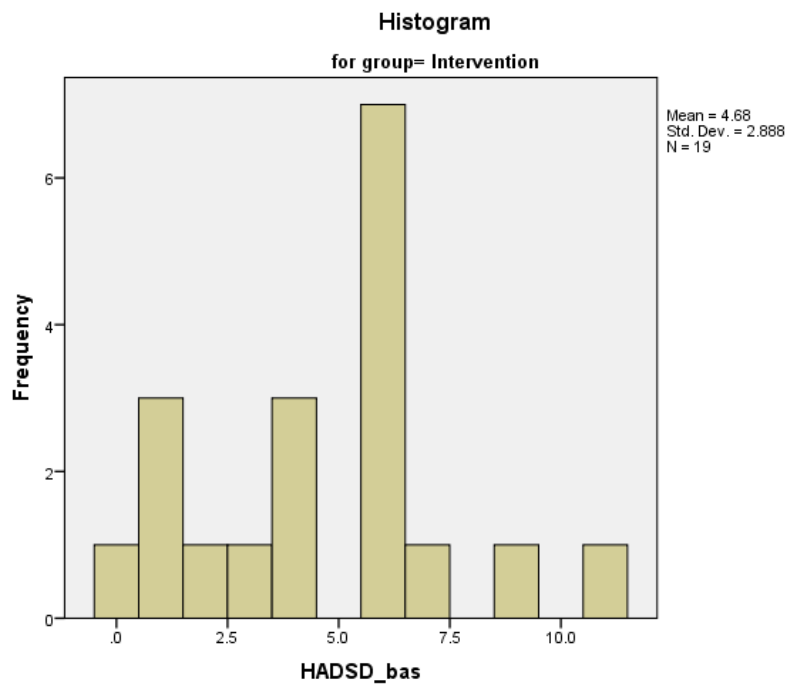


Fig 18: Histogram for HADS-depression, control group (not normally distributed)

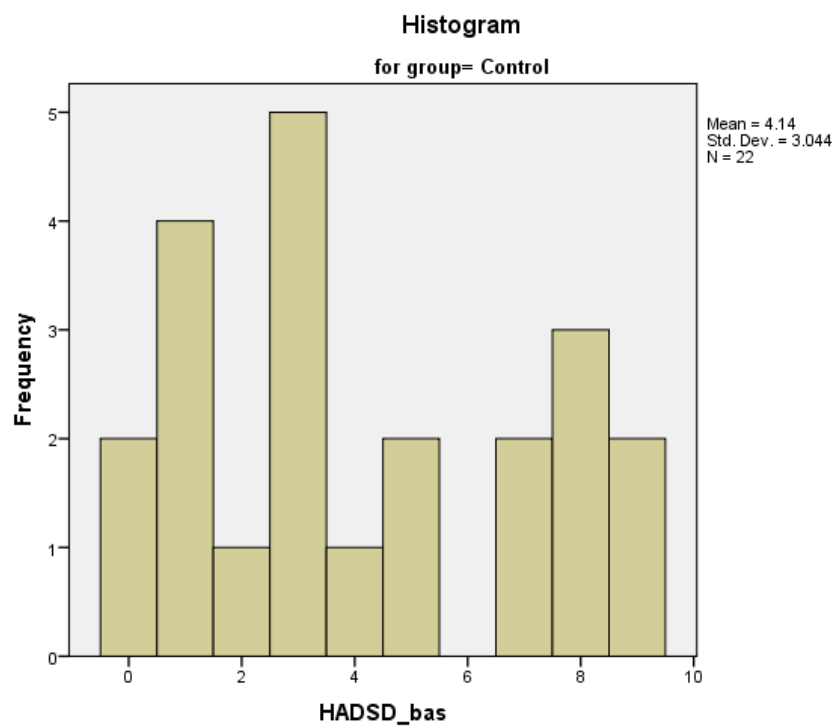


Fig 19: Detrended normal Q-Q plot for HADS-depression, intervention group (not normally distributed)

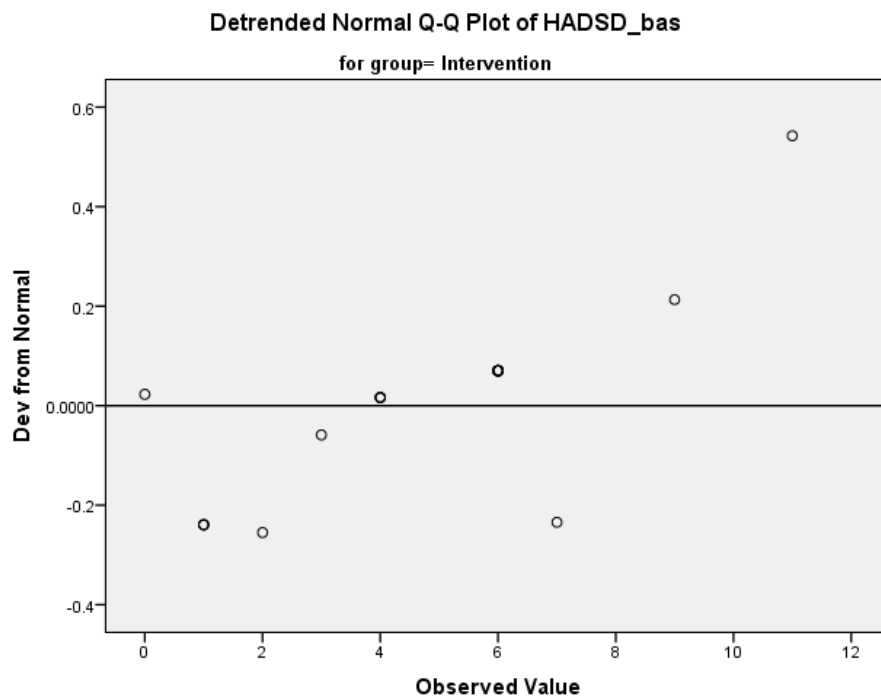


Fig 20: Detrended normal Q-Q plot for HADS-depression, control group (not normally distributed)

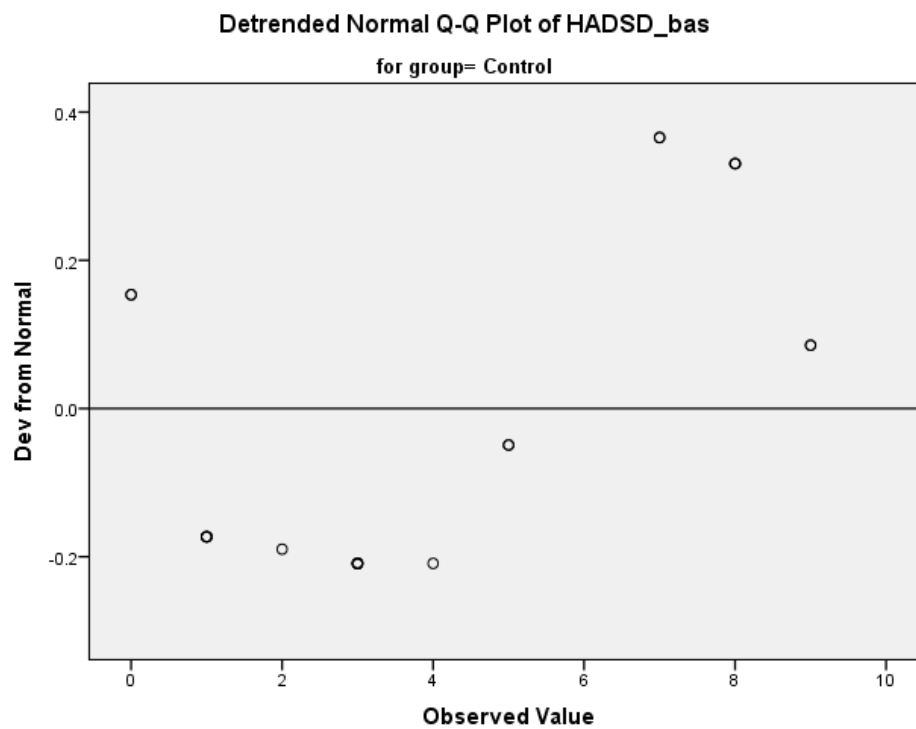


Fig 21: Histogram for Quality of Care giving relationship scale, intervention group (normally distributed)

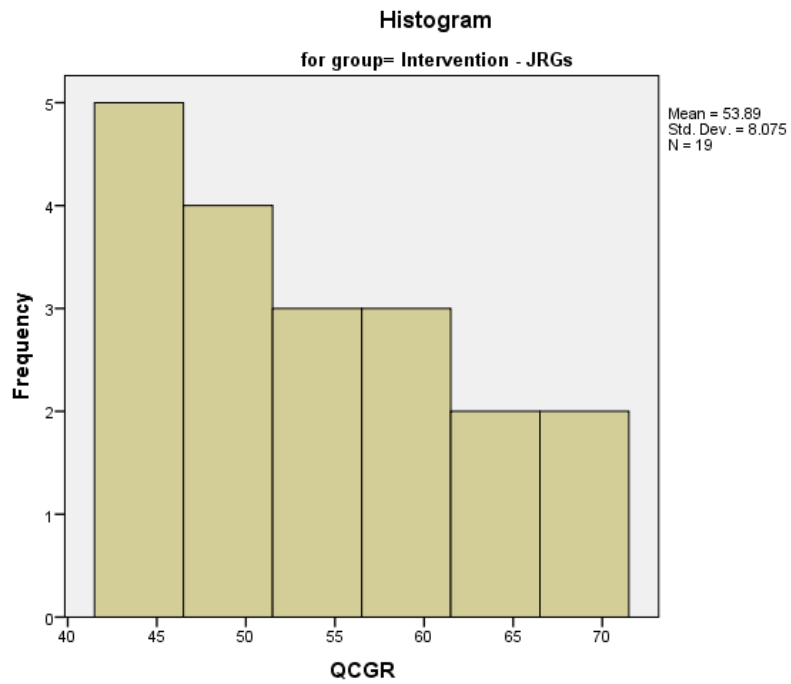


Fig 22: Histogram for Quality of Care giving relationship scale, control group (normally distributed)

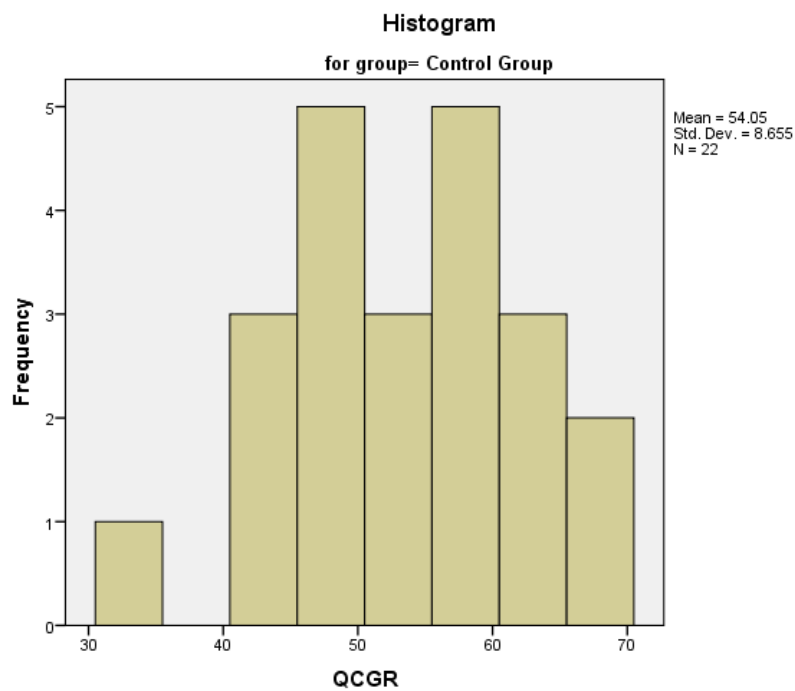


Fig 23: Detrended normal Q-Q plot for Quality of Care giving relationship scale, intervention group (normally distributed)

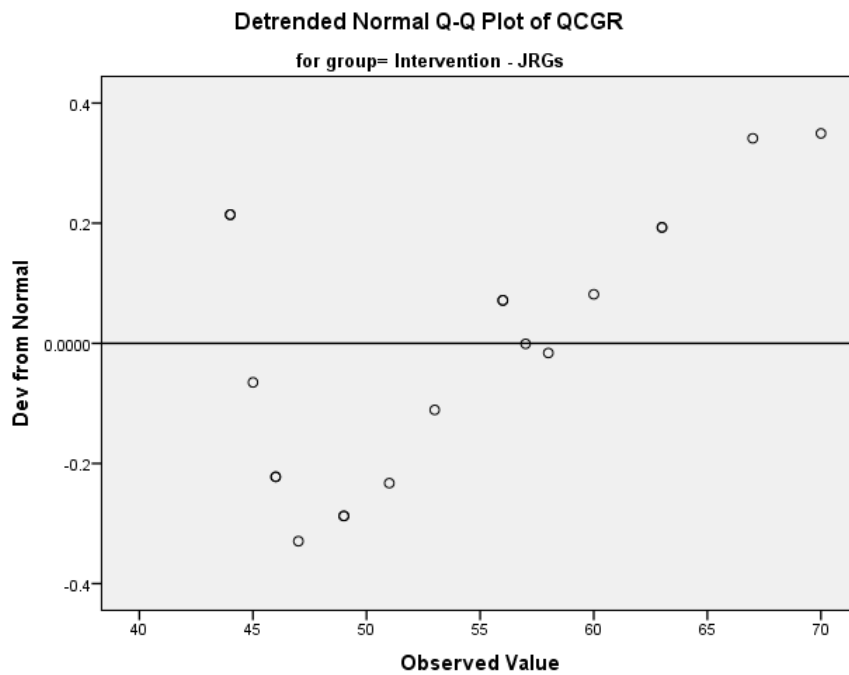


Fig 24: Detrended normal Q-Q plot for Quality of Care giving relationship scale, control group (normally distributed)

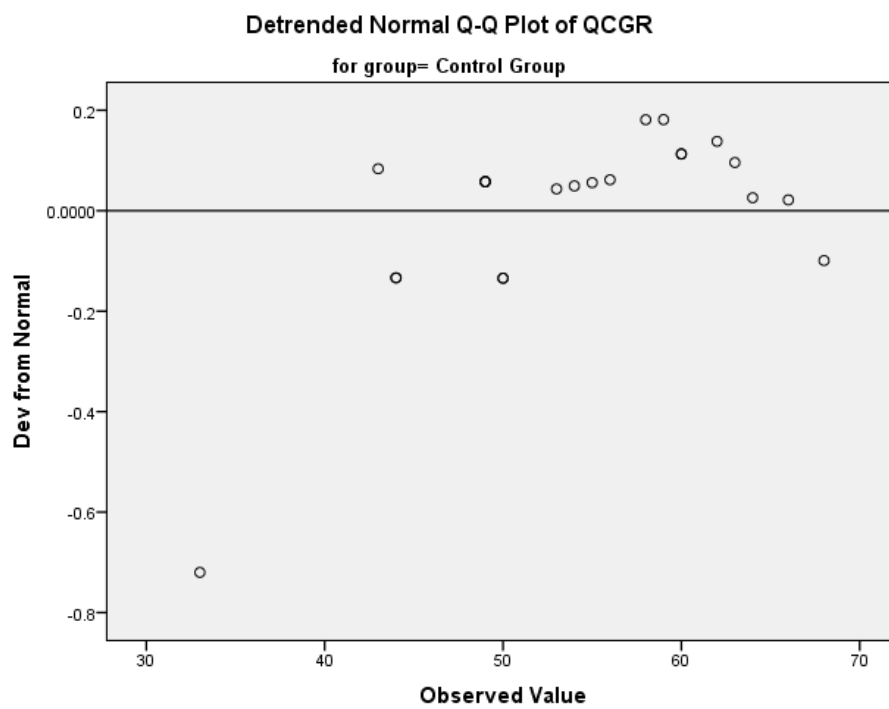




Fig 25: Histogram for Relatives Stress Scale, intervention group (normally distributed)

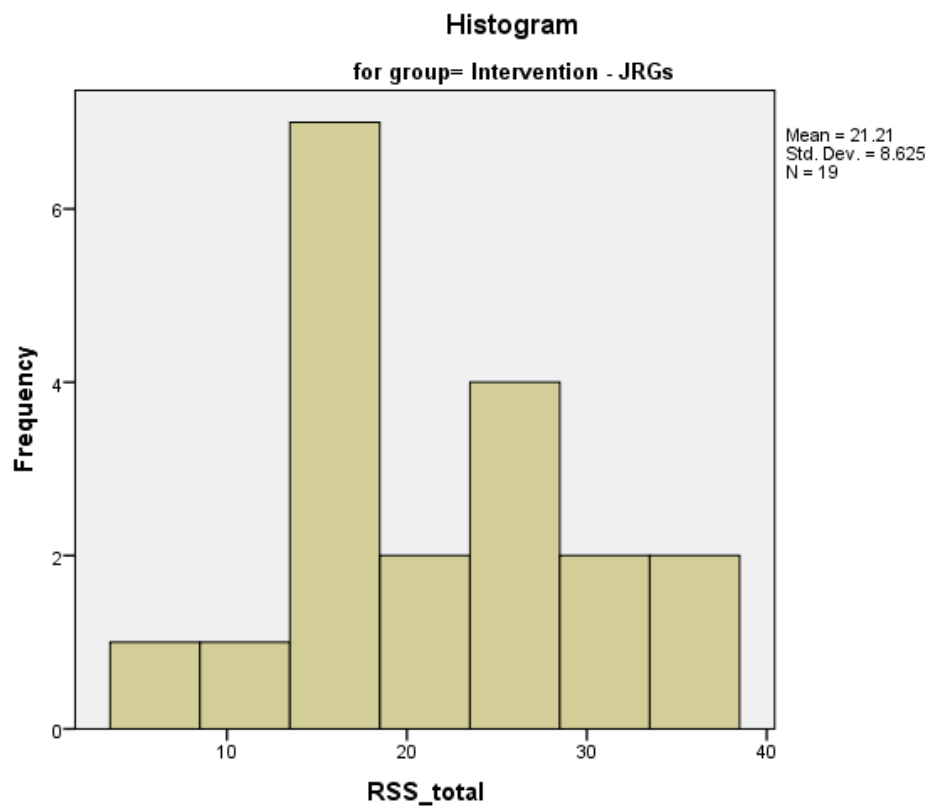


Fig 26: Histogram for Relatives Stress Scale, control group (normally distributed)

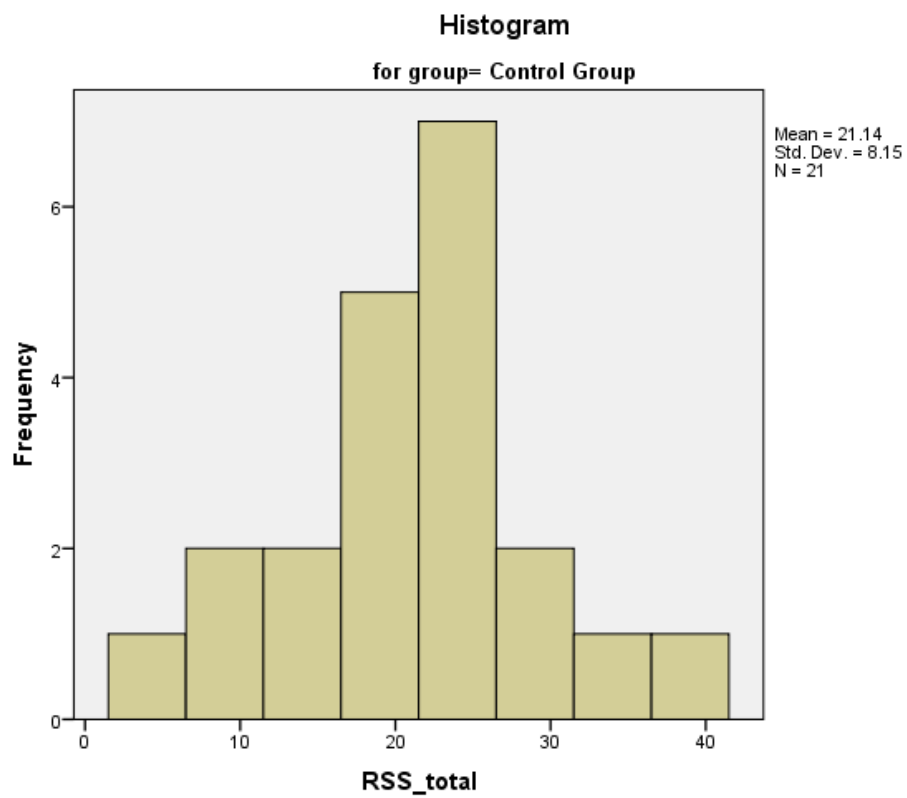


Fig 27: Detrended normal Q-Q plot for Relatives Stress Scale, intervention group (normally distributed)

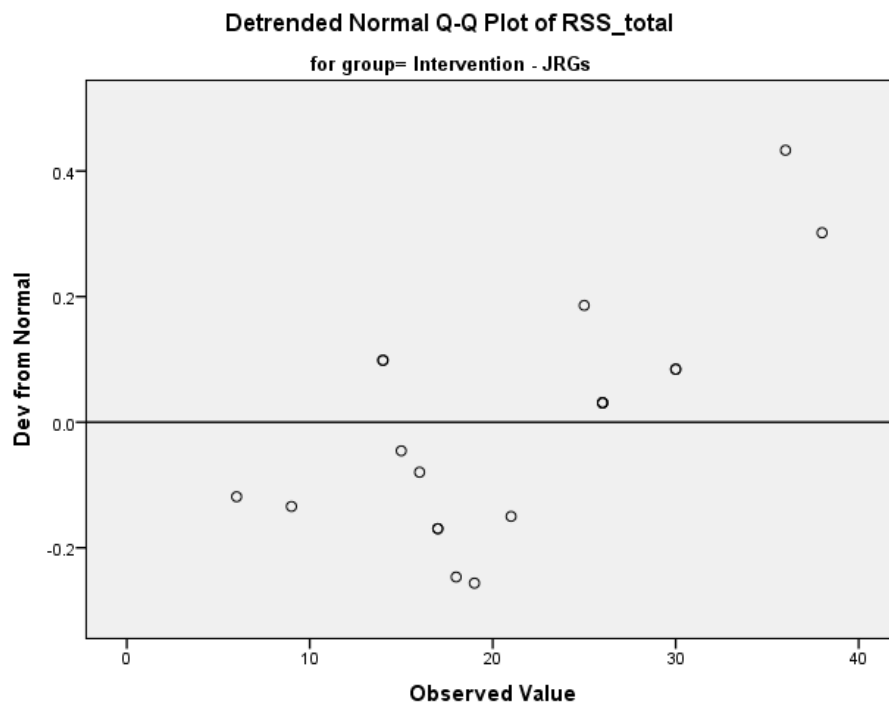


Fig 28: Detrended normal Q-Q plot for Relatives Stress Scale, control group (normally distributed)

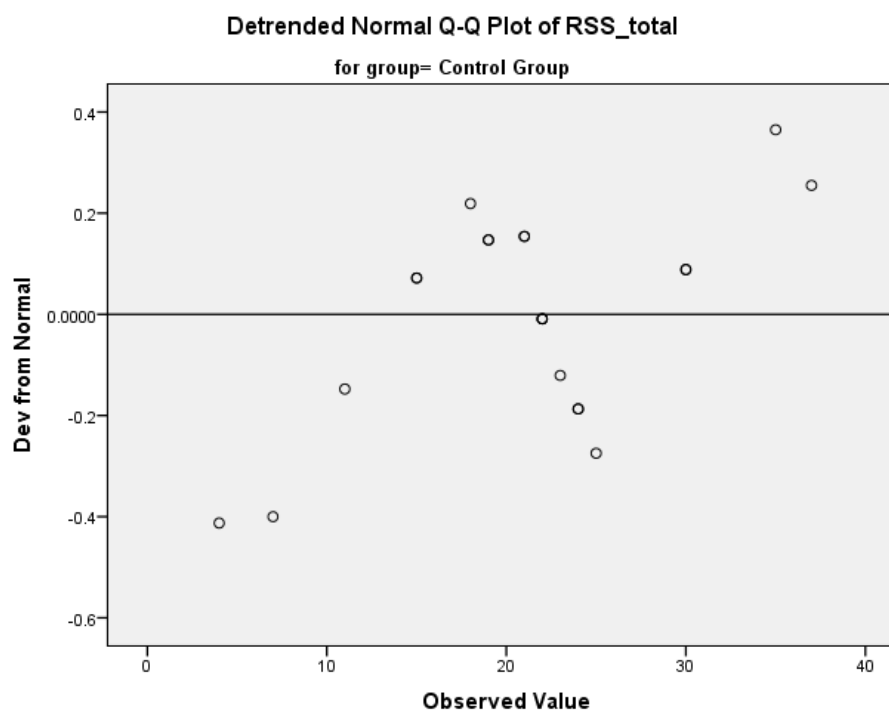


Fig 29: Histogram for GHQ-28, intervention group (normally distributed)

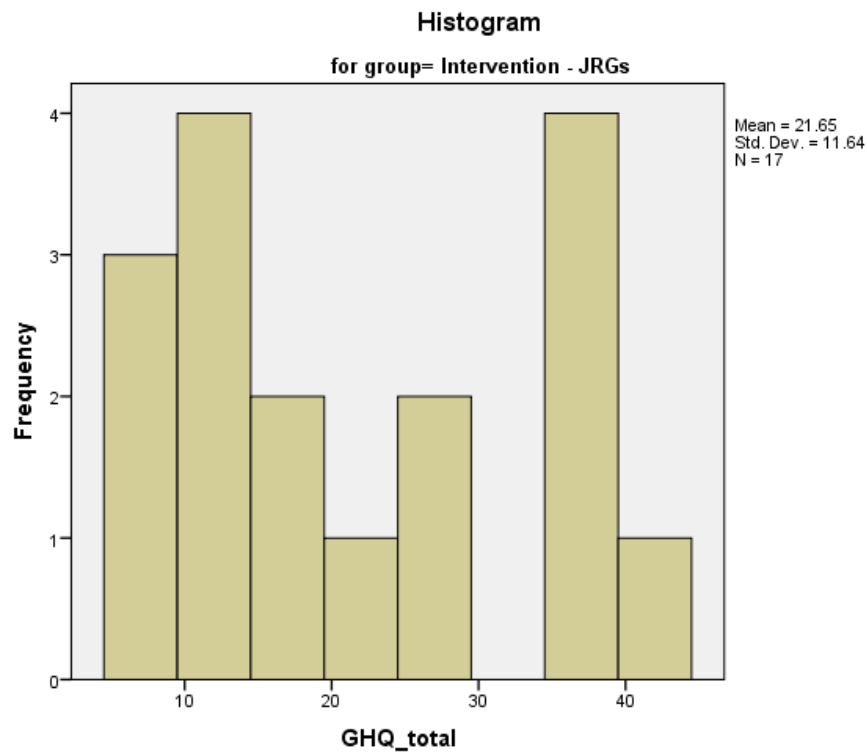


Fig 30: Histogram for GHQ-28, control group (normally distributed)

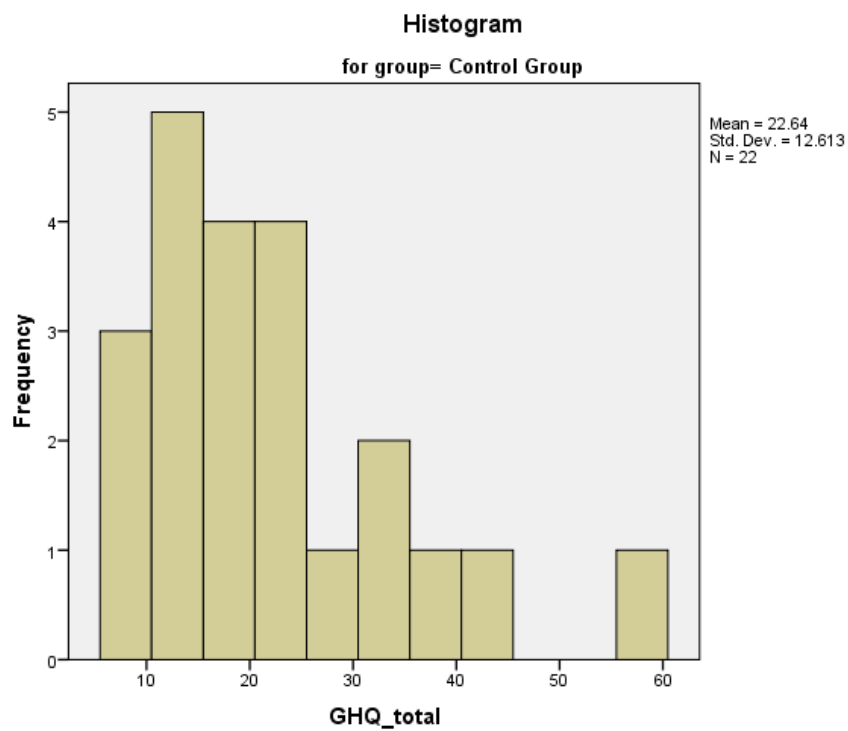


Fig 31: Detrended normal Q-Q plot for GHQ-28, intervention group (normally distributed)

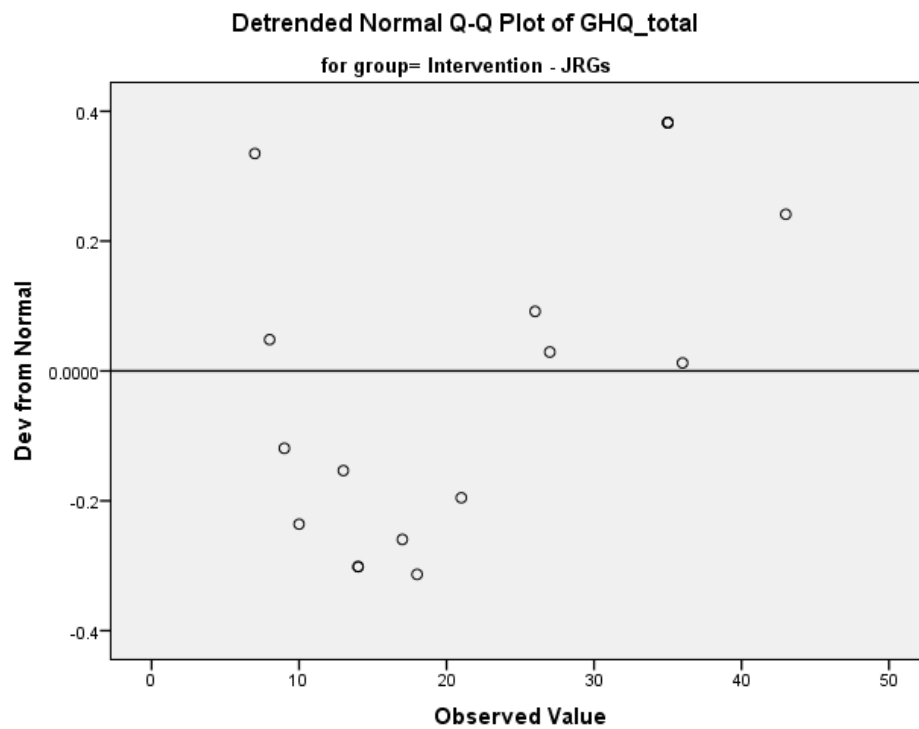


Fig 32: Detrended normal Q-Q plot for GHQ-28, control group (normally distributed)

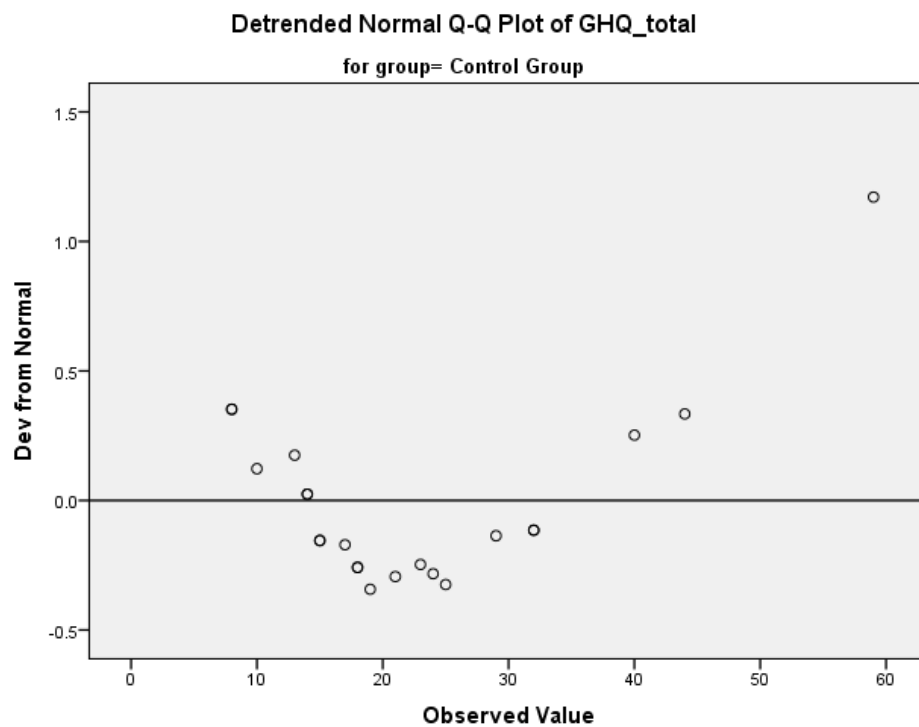


Fig 33: Histogram for costs, intervention group (not normally distributed)

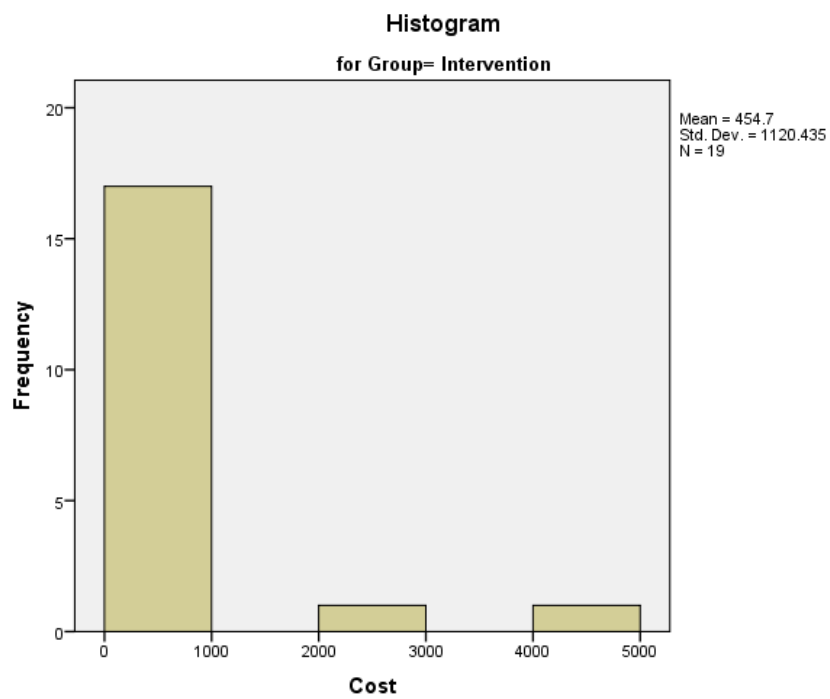


Fig 34: Histogram for costs, control group (not normally distributed)

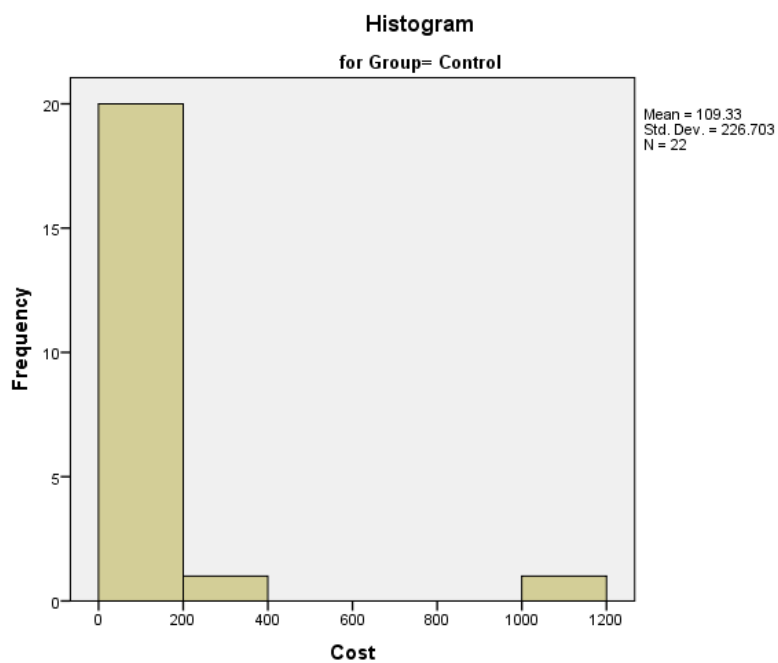


Fig 35: Detrended normal Q-Q plot for costs, intervention group (not normally distributed)

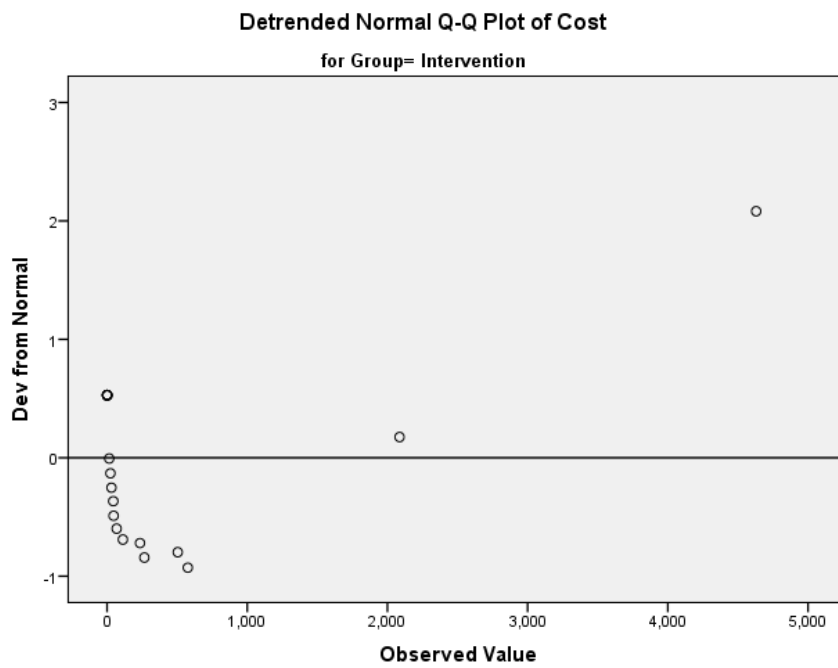


Fig 36: Detrended normal Q-Q plot for costs, control group (not normally distributed)

