THE EFFECT OF DISEASE ADAPTATION INFORMATION ON GENERAL POPULATION VALUES: A CASE STUDY USING RHEUMATOID ARTHRITIS STATES

by

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Abstract

Objective: The Washington Panel on Cost-effectiveness in Health and Medicine recommends that economic evaluation of healthcare technologies uses values for health states elicited from the general population rather than patients. However, general population respondents do not necessarily recognize the possibility of adapting to the impaired state. This thesis examines how informing the general population about disease adaptation influences their values. Rheumatoid arthritis (RA) states are used as an illustration.

Methods: This work employed a sequential mixed-methods design using three components. First, Rasch and cluster analyses were used to construct RA states. Simultaneously, a novel adaptation exercise consisting of audio-recordings of patients discussing disease adaptation was developed. Second, semi-structured interviews were conducted with 12 general population respondents to identify the effect of adaptation information on their perceptions of RA. Finally, the influence of this information on health state values from a random sample of the general population ($n = 200$) was assessed quantitatively and the factors contributing to this change were identified.

Results: The first component of this study defined three RA states. In the second, the qualitative interviews revealed that the adaptation exercise encouraged the general population to empathize with the messages in the audio-recordings. Finally, the third component showed that the adaptation exercise was effective at changing health state values; for example, for the severe RA state, a mean (standard deviation) change of 0.17 (0.34) ($p < 0.01$) was observed. Individuals who were younger, were healthier, recognized the importance of coping strategies, and comprehended the valuation task were more likely to increase their values.

Conclusions: The results from this thesis demonstrate a novel method of informing the general population about disease adaptation. After undertaking the adaptation exercise, most respondents increased their values for the given health states. Thus, important contributions are made to an emerging field of developing better informed general population values.

Table of Contents

List of Tables

List of Figures

Nomenclature

ANOVA = Analysis of variance
CEA = Cost-effectiveness analysis
DF = Degrees of freedom
DIF = Differential item functioning
EQ-5D = EuroQol-5D©
HAQ = Stanford Health Assessment Questionnaire©
ICER = Incremental cost-effectiveness ratio
KMO = Kaiser-Meyer-Olkin
MD = Mean difference
MVH = Measurement and Valuation of Health study
NDB = National Data Bank for Rheumatic Diseases
NHS = National Health Services
NICE = National Institute for Health and Clinical Excellence
PCA = Principal components analysis
PSI = Person separation index
QALY = Quality-adjusted life year
QOL = Quality of life
QWB = Quality of Well-being
RA = Rheumatoid arthritis
RCQ = Reasons to Change Questionnaire
RS = Rating scale
SD = Standard deviation
SE = Standard error
SG = Standard gamble
TTO = Time trade-off
UK = United Kingdom
US = United States
VAS = Visual analogue scale

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Chapter One

Introduction

Recent advances in healthcare technologies offer ever-improving levels of care. The financial implications of these treatments are becoming increasingly burdensome on publicly funded healthcare systems; available resources are never sufficient to meet all demands (Drummond et al., 2005). As a result, decision-makers are faced with the challenge of allocating these limited resources in the fairest and most efficient manner possible.

A publicly funded healthcare system exists in the United Kingdom (UK). Through general taxation, the country's National Health Service (NHS) aims to provide healthcare for members of the general population on a fixed budget. To ensure that healthcare resources are utilized efficiently, the NHS may follow guidance from the National Institute for Health and Clinical Excellence (NICE). NICE is an independent health technology assessment organization, whose main aims are to promote good health and to prevent and treat ill health (National Institute for Health and Clinical Excellence, 2008). NICE's guidance promotes the application of economic evaluation to healthcare technologies to ensure that informed, transparent, and consistent decisions are made.

The preferred methodology to be used in economic evaluation is cost-effectiveness analysis, in which the benefits of health interventions are quantified using quality-adjusted life years (QALYs) (National Institute for Health and Clinical Excellence, 2008). The QALY is a measure that combines information regarding both duration and quality of life (QOL) into a single index; this is discussed further in Chapter Two. While the use of the QALY is accepted, as demonstrated later, there are debates as to who should be providing the information on QOL to incorporate into cost-effectiveness analysis. This normative debate focuses on whether patients or members of the general population are the better choice.

1.1 Whose Health State Values Should Be Used in Healthcare Decision-Making?

The Washington Panel on Cost-effectiveness in Health and Medicine advocates that information on QOL, in the form of health state values[1], should be obtained from *informed* members of the general population[2] rather than from patients (Gold et al., 1996). Respondents are asked to envision what life in the impaired health state would be like; the QALYs are calculated from their resulting health state values and used to inform health policy decisions. This method follows the concept that, in a publicly funded healthcare system, the main objective is to meet societal preferences for maximizing health. A principal drawback to this approach is that the general population respondents may not fully comprehend life with an impaired health condition nor consider the possibility of adapting to the impaired state; this will depend on the health state being valued. Thus, the use of general population values may lead to inaccurate valuations and, subsequently, decisions which ultimately do not maximize health benefits.

Alternatively, it can also be argued that patients, due to their direct experience, are better suited to value their own health (Nord et al., 1999). However, there is concern that patients' self-reports of their QOL may be distorted (Ubel et al., 2003) in an attempt to modify their health state values to please their doctor or family members. Using only patients' or only members of the general population's values for health states leads to different outcomes when used to evaluate various types of healthcare technologies; this is discussed in greater detail in Chapter Two. Using *informed* general population values may be considered to be a better approach in guiding healthcare decisions, as it combines the impartiality of the general population with some knowledge similar to that of patients (Gold et al., 1996; Fryback, 2003; Ubel et al., 2003; Brazier et al., 2005); this is described further in Chapter Two.

The advantage of using values provided by informed general population respondents in a healthcare decision-making context is further exemplified by the fact that previous comparative valuation studies have shown that discrepancies between patient and general population values do, in fact, exist (e.g., Sackett and Torrance, 1978). Most such studies have found that patients provided higher health state values. This suggests that the respondents from the general population do not

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[1] Due to interchangeable use of the terms *health state value*, *utility*, *quality of life value*, and *quality-adjusted weights* found in the scientific literature, the term *health state value* will be used consistently throughout this thesis to avoid any potential for confusion.

[2] The term 'general population' is used to describe a heterogeneous group which may contain patients. On average, the current health states of the respondents from the general population are better than the health states that they are asked to imagine experiencing in most valuation studies.

understand how valuable life can be for people with disabilities, do not consider that adaptation to the impaired health state can occur over time (Hadorn, 1992; Ubel et al., 2003) or, perhaps, do not consider a combination of both aspects. However, there are also instances showing the opposite trend, with the members of the general population valuing some diseases higher than do patients (e.g., Stein et al., 2003; Stolk and Busschbach, 2003). The results from these studies are described in greater detail in Chapter Three.

1.2 Research Needs and Justification

Previous work has highlighted that health state values provided by patients do differ from those provided by the general population. The reasons for this divergence have not been empirically examined. Researchers have proposed that the main reasons for this difference are either that patients and members of the general population interpret the health states differently or that they incorporate disease adaptation into their valuation differently (Ubel et al., 2003; Brazier et al., 2005; Stiggelbout and de Voel-Voogt, 2008). While descriptions of health states can be optimized to ensure similar interpretations by patients and the general population, the fact remains that respondents from the general population fail to consider that, over time, adaptation to the new health state is possible; the concept of adaptation is discussed in more detail in Chapter Two. The incorporation of adaptation into health state valuations, or the lack thereof, could significantly impact the outcome of a cost-effectiveness analysis; furthermore, this impact is expected to vary between disease states.

While it is speculated that informing the general population about disease adaptation may alleviate the differences observed between the health state values, there is a lack of empirical information in the scientific literature about informed general population values. Specifically, there is a need to determine how members of the general population can be informed about disease adaptation and a need to understand the effect that information pertaining to adaptation may have on their values for health states. By addressing these issues, a greater understanding of informed general population values can be gained.

1.2.1 A Case Study: Valuing Rheumatoid Arthritis States

Rheumatoid arthritis (RA) was chosen as the case study disease for this Ph.D. thesis. It is a chronic autoimmune inflammatory disorder that afflicts 0.8% of the population in the UK (Symmons, 2005). RA usually begins in the small joints of the patient's hands and feet (Symmons et al., 2000). Individuals with RA suffer with painful, tender, and swollen joints, and may experience a loss of functioning in everyday activities. While the health of an individual with RA usually deteriorates over the longer term, the condition itself may also fluctuate in severity. RA is a systemic disease; for patients suffering from a severe form of RA, the condition can affect the whole body and internal organs (National Rheumatoid Arthritis Society, 2008).

According to the UK's National Rheumatoid Arthritis Society (2008), RA tends to affect more women than men. The onset of the condition generally occurs in individuals between 40-60 years of age, although RA is known to occur at any age. In 2008, approximately 12,000 children under the age of 16 suffered from juvenile RA in the UK. Although there is no available cure, early detection and treatment ensures that individuals suffering from RA have a generally good QOL.

RA was chosen to illustrate the effects of informing disease adaptation on general population values for numerous reasons. Firstly, its primary symptoms address aspects of functioning. These symptoms, especially when compared to those of mental health issues, are believed to be more comprehensible to respondents who may not be well informed about the condition under investigation. Secondly, research has shown that individuals living with RA are likely to adapt to their health condition over time (Affleck et al., 1987; DIPEx Health Experiences Research Group, 2008). Thirdly, the disease is such that it appears to strike randomly; therefore, there is no blame or negative stigma attached to it. Finally, as there is no cure currently available for RA, treatment decisions will focus on achieving a given improvement in QOL, rather than a complete cure. As a result, understanding why members of the general population value health states the way they do could help to ensure that treatments are prioritized so that they maximize the improvement in QOL for all individuals living with RA; this issue is exemplified further in Chapter Two.

1.3 Thesis Objectives

The overall aim of this thesis is to examine how informing general population respondents about disease adaptation influenced their valuation for hypothetical health states. The specific project objectives were to:

1. Construct representative RA health states to be valued by respondents from the general population;
2. Develop an intervention for informing general population respondents about disease adaptation;
3. Explore the influence of disease adaptation information on general population respondents' perceptions of RA;

4. Quantify the impact that disease adaptation information has on altering health state values from general population respondents; and
5. Identify the factors that influence the general population respondents to change their initial health state values after being informed about disease adaptation.

By meeting these objectives, a better understanding of how disease adaptation information influences general population values can be achieved. From this, it may be possible to determine how the general population can be informed about disease adaptation and to understand the influences this information may have on their health state values.

1.4 Organization of the Thesis

This thesis is comprised of nine chapters. The current chapter, Chapter One, provides a brief introduction to the use of health state values in a decision-making context and identifies the overall objectives of this research project. Chapter Two introduces some of the pertinent techniques used in the appraisal of healthcare technologies and discusses whose health state values should be considered in an economic evaluation framework. Chapter Three attempts to identify the factors that cause the discrepancies observed between patient and general population values by synthesizing the results from a literature review of published comparative valuation studies. Chapter Four provides an overview of the methodology involved in conducting this research project. Chapter Five describes the first of the empirical results. This chapter specifically discusses the development of the RA health states using patient condition-specific information (Thesis Objective One). Prior to describing the remaining empirical results, Chapter Six presents the key elements that are fundamental to the methodologies for the studies that are described in Chapters Seven and Eight. Chapter Six particularly focuses on the intervention used to inform the respondents about disease adaptation (Thesis Objective Two). Chapter Seven qualitatively explores the impacts disease adaptation information may have on individuals' perceptions of RA (Thesis Objective Three). Chapter Eight quantitatively assesses first whether or not general population respondents change their values for RA health states after being informed about disease adaptation (Thesis Objective Four), and then what the factors are that influence this change (Thesis Objective Five). The final chapter, Chapter Nine, summarizes the key research findings and outlines the contributions the research project has made to the scientific literature. Suggestions for future work are also addressed in this chapter.

Modified versions of Chapters Three, Five, Seven, and Eight are either under review or in preparation for submission to major peer-reviewed journals. The chapters describing the empirical results (i.e., Chapters Five, Seven, and Eight) have been disseminated at various conferences and published as discussion papers (for a discussion of the dissemination of the thesis results, refer to Appendix A.1, page 239). The work presented in this thesis was conducted solely by the Ph.D. candidate.

1.5 Summary

The demand to maximize the benefits of limited healthcare resources requires decision-makers to make choices on how best to allocate them. One widely used approach to inform resource allocation decision is cost-effectiveness analysis; however, there are normative debates as to whose values – patients or members of the general population – should be given the greater weight in assessing health states. While responses from the general population should benefit society at large, patients have a better understanding of the effect of living in an impaired health state and hence will incorporate adaptation into their valuations. Informed general population values have been proposed as an unbiased method for use in healthcare resource allocation decision-making; however, very little is known about how to inform respondents or the impact this may have on their valuations. The primary aim of this thesis is to examine how informing general population respondents about disease adaptation influenced their valuations of health states. Rheumatoid arthritis states are used as an illustration.

Chapter Two

Economic Evaluation in Healthcare: Whose Values Could Be Used?

Whose values – those of patients, or those of members of the general population – should be considered when appraising health states? This question is constantly debated. The decision as to whose voice should be given preference within an economic evaluation framework is primarily a normative choice. Either perspective, depending on the decision-making context, may lead to legitimate outcomes (Williams, 1991).

At the policy level, the obvious solution may be to use general population values when implementing healthcare decisions; this is based on the fundamental premise that meeting societal preferences should maximize health. The issue is that previous work comparing the valuation of health states revealed that values provided by patients and by members of the general population do, in fact, differ; the results of these studies are discussed in greater detail in Chapter Three. It is generally accepted that patients' ability to adapt to the impaired conditions – and, similarly, the general population's unawareness of disease adaptation – make the greatest contribution to the differences observed between the two sets of values (Ubel et al., 2003; Brazier et al., 2005). Understanding these differences is important as the use of either patient or general population values may lead to different outcomes when incorporated into an economic evaluation framework; this ultimately affects decisions that address the allocation of scarce healthcare resources.

This chapter aims to provide a necessary background to understand the implications of adaptation on economic assessment in healthcare. The first objective is to present economic evaluation techniques used to appraise healthcare technologies. The second is to examine the impacts of using solely patient or general population values within an economic evaluation framework. The final objective is to describe the use, and the development, of informed general population values.

2.1 Economic Evaluation Techniques in Technology Appraisal

Globally, there is an insatiable demand for resources that greatly exceeds available supply. As discussed in Chapter One, within the field of publicly funded healthcare, resources available to meet its demands are scare. Decision-makers are therefore faced with the challenge of how to allocate these resources to ensure that fair and efficient decisions are being made.

The National Institute for Health and Clinical Excellence (NICE), an independent health technology assessment organization, recommends that economic evaluation be used to aid decision-making to ensure that resources allocated to healthcare are utilized efficiently (National Institute for Health and Clinical Excellence, 2008). Since the introduction of NICE, the use of economic evaluation to appraise healthcare technologies has increased (Brazier et al., 2007). In addition to the United Kingdom (UK), other countries – for example, Canada (Canadian Agency for Drugs and Technologies in Health, 2008) and Australia (Australian Government Department of Health and Ageing, 2007) – have adopted similar approaches to healthcare decision-making by advocating the use of economic evaluation.

Economic evaluation techniques are used in the appraisal of healthcare technologies to ensure that informed, transparent, and consistent decisions are being made (National Institute for Health and Clinical Excellence, 2008). The key feature of economic evaluation is that it enables healthcare technologies to be compared by considering their costs and benefits (Drummond et al., 2005).

While different techniques can be used in economic evaluation (e.g., cost-benefit, cost-minimization, and cost-consequence analyses) (Drummond et al., 2005), cost-effectiveness analysis (CEA) is the preferred methodology to appraise the comparative impacts of expenditures on different healthcare technologies (National Institute for Health and Clinical Excellence, 2008). A fundamental of CEA is the incremental cost-effectiveness ratio (ICER). The ICER permits the comparison of two alternatives, where one is the healthcare technology under investigation and the other is a suitably chosen alternative (e.g., the technology currently used in practice). This is determined by:

$$ICER = \frac{\Delta Cost}{\Delta Effectiveness}, \qquad (2.1)$$

where $\Delta Cost$ is the difference in cost between the two alternatives and $\Delta Effectiveness$ is a numeric value representing the difference in effectiveness between the two alternatives. The ICER is essentially the incremental price of obtaining a unit of health effect from a given healthcare technology when compared with an alternative. Medical interventions that have relatively low ICERs (i.e., less cost but more effective) are considered good value and, thus, would have a higher priority for the available resources. Conversely, medical interventions with higher ICERs (i.e., more cost but less effective) would have lower priority when competing for resources. CEA can also inform decisions in situations where, when compared to existing alternatives, a new technology is either more costly but more effective, or less costly but less effective.

Obtaining the cost component ($\Delta Cost$) for use in an ICER is considered to be relatively straightforward, and is not of specific relevance for this thesis. Determining the effectiveness component ($\Delta Effectiveness$), however, is more complex. Two methods that can be used to assess effectiveness are clinical outcomes or quality-adjusted life years (QALYs).

As the name suggests, clinical outcomes are units associated with the health condition under investigation. Examples include number of symptom-free days and the reduction of blood pressure (mm Hg) in the treatments of asthma and hypertension, respectively (Sculpher and Buxton, 1993; Mark et al., 1995); an even simpler measure is the length of life gained. The use of these outcomes allows the detection of the patient's improvement (or deterioration) if a dominant (or dominated) relationship exists between the before and after health states. However, these measures are not easily comparable between disease states.

The other option for use in the denominator of the ICER (Equation 2.1) is QALYs. Using QALYs in an ICER is sometimes referred to as 'cost-utility analysis' (Drummond et al., 1997). The QALY contains information pertaining to both length and quality of life (QOL) of a health state. As different health states can exist for a particular condition, the QALYs for the individual states are summed together to yield a single index:

$$QALY_{Total} = \sum^{n} t_i V_i \quad , \tag{2.2}$$

where t_i is the time spent in a health state i, V_i is the QOL value individuals place on living in that health state, and n is the number of health states for the investigated condition. NICE specifically recommends the use of QALYs in an economic evaluation because, unlike clinical outcomes, QALYs permit comparisons across different diseases (National Institute for Health and Clinical Excellence, 2008).

Values describing QOL (V_i) represent the desirability individuals place on living in a particular health state. They are anchored by a value of 'one' for full health and 'zero' for dead, where a higher value indicates a greater preference for a given health state. The lower boundary can be extended into the negative region as some health states – for example, time spent in a coma (Patrick et al., 1994) – can attract negative values. Values in the negative range indicate, from a societal perspective, health states worse than dead. Negative values cannot be interpreted to mean that society wants such patients to be allowed to die. They merely represent the fact that members of the general population consider that existing in such a health state is worse than being dead. However, if a patient was to provide a negative value for their health then it is likely that the individual probably would prefer to be dead rather than living in their current state.

2.1.1 Elicitation of Health State Values

While measuring length of life (t_i) is straightforward, the values (V_i) for use in QALYs are more challenging to obtain. These values tend to be based on respondents' appraisals of health states described in written format. Therefore, for QALYs to be of use in economic evaluation, the health states need to be well described and the valuations of these states needs to be done appropriately. This section describes how an individual's values for health states are elicited.

First, health states are developed. These states need to describe the condition under investigation as accurately as possible. If the health states are not well described, it is possible that individuals will appraise the health states differently from each another; the implication of using such insensitive health state descriptions is discussed further in Chapter Three.

There are many methods which researchers can use in the development of health states. One approach would be to develop customized health state descriptions for the condition under investigation. This can be achieved by conducting an extensive literature review or by consulting a panel of experts. A review of the literature can identify key features of the health condition that are commonly experienced by individuals currently living in the impaired state. Another approach would be to consult specialist physicians and patients directly. This allows in-depth knowledge about life in different health states to be gained. This may be done through the use of focus groups, face-to-face interviews, or responses on instruments assessing QOL. QOL instruments may include condition-specific measures, which are capable of obtaining specific information about the condition under investigation. However, the descriptive system of such instruments may exclude co-morbidities and other complications which are important to an individual's overall QOL. As a result, responses to 'generic' multi-dimensional health status classification systems (described below) may be used to provide a wider scope of information. The methods mentioned above are not mutually exclusive and therefore a combination of approaches may be undertaken in the development of health states.

Once the health states are developed, respondents are asked to appraise them and provide a value for these states using a number of valuation techniques. The most widely used methods to measure the value of a health state are the standard gamble, the time trade-off, and the rating scale; these are described below.

The standard gamble (SG) is a classic method of measuring cardinal preferences. Grounded in von Neumann-Morgenstern expected utility theory, the SG method requires the respondent to make a choice between outcomes, in which one outcome involves an element of uncertainty (Drummond et al., 2005). This 'gamble' involves a probability of a better or worse outcome than the certain outcome (Tengs and Wallace, 2000). The goal of this approach is to determine the probability in the gamble at which the respondent is indifferent between the certain and uncertain alternatives.

Designed as another option to the SG, the time trade-off (TTO) approach aims to overcome the problems encountered when explaining probabilities to the respondents (Torrance et al., 1972; Drummond et al., 2005). The respondents are asked to choose between a shorter life span in full health *versus* a longer life span in an impaired health state. The time in full health is varied until the respondent is indifferent between the two alternatives. Unlike SG, the TTO choice is not made under uncertainty. The TTO approach is one of the valuation method used for this thesis; as a result, it is discussed in greater detail in Chapter Six.

The rating scale (RS) is the simplest approach for measuring values for health states. The fundamental purpose of this method is to create an interval scale of preferences (Drummond et al., 2005). There are many variations on the RS approach. They typically have a numerical scale: others include category scales, which consist of a small number of categories that are assumed to be equally spaced, and visual analogue scales (VASs), which consist of a line on a page, often 10 cm in length. with clearly defined endpoints with or without other marks along the line. The RS technique is often used as a prelude to administering other valuation exercises to familiarize the respondents with the descriptions of the health states. The VAS is the other valuation method used in this thesis; this approach is also described in Chapter Six.

While the previous paragraphs described the processes to develop, and to value, health states, there are also instances when individuals are asked to value their own current health (e.g., patients participating in clinical trials). Respondents are asked to think about what they are currently experiencing and to provide a value for their own health using SG, TTO, and RS approaches, as outlined above. As such, health states do not need to be developed. This approach is typically used to obtain patient-level health state values.

Another approach would be to have individuals respond to pre-scored multi-attribute health status classification systems. These classification systems have been developed in an attempt to alleviate the complex and time-consuming nature of collecting health state values using, for example, the SG and TTO approaches (Drummond et al., 2003). They consist of a defined health state descriptive system, as well as a matching value set, and therefore can dually serve to describe and to attach a value to health states.

The most widely used multi-attribute health status classification systems are the EuroQol-5D (Brooks, 1996), the Health Utilities Index (Torrance et al., 1996; Feeny et al., 2002), and the Medical Outcomes Short Form 6D (Brazier et al., 2002). These classification systems differ in the dimensions of health they cover, in the description and the number of levels defined for each dimension, and in the severity of the most severe level. As the use of the EuroQol-5D (EQ-5D) in economic evaluation is recommended for healthcare decision-making in the NICE guidance (National Institute for Health and Clinical Excellence, 2008), this multi-attribute health status classification system is used in the empirical studies, discussed in Chapters Seven and Eight of this thesis.

The EQ-5D is a self-administered survey consisting of a descriptive health state classification system with five dimensions (mobility, self care, usual activity, pain/discomfort, and anxiety/depression) and a VAS 'health thermometer' (Essink-Bot et al., 1993; Brooks, 1996) (refer to Appendix A.2, page 241, for a copy of the

EQ-5D). The 'health thermometer' represents a subjective, global evaluation of the respondent's current health status on a vertical scale between zero and 100, where zero represents the worst imaginable health state and 100 represents the best imaginable health. These ratings can be standardized to ensure that they are anchored at zero, representing dead, and one, representing full health; this is described in greater detail in Chapter Six.

Each of the five dimensions in the EQ-5D has three levels – no problem, some problems, and major problems – yielding a total of 243 (3^5) possible health states. These health states, along with 'unconscious' and 'dead' states, were originally valued by a random sample of approximately 3000 members of the adult population in the UK (Dolan et al., 1996; Dolan and Gudex, 2000) using the TTO technique. The health state values were then subjected to econometric modelling to obtain regression coefficients for TTO tariffs (i.e., weights for each health state defined by the classification system). These tariffs are combined with the individual's responses when completing the EQ-5D descriptive system to afford an overall index. The EQ-5D indices can fall on the zero (dead) to one (full health) value scale, with a lower bound of -0.59 to account for states worse than dead.

2.2 Whose Values Should Be Used?

While the previous section highlighted the importance of health state elicitation for use in CEA and the variety of ways of doing this, researchers have also questioned whose health states values – patients or members of the general population – should be given the most consideration when making decisions regarding the implementation of healthcare technologies. The following sections illustrate the advantages and disadvantages for solely using patient values or general population values within an economic evaluation framework.

2.2.1 Patient Values

The main rationale for using the patient's perspective in healthcare decision-making is to place greater emphasis on the preferences of those most directly affected by an intervention or policy (Froberg and Kane, 1989). Patients, due to their direct experience, are better suited to value their own health (Nord et al., 1999) although self-reports of their QOL may be distorted when they feel the need to modify their values to please their doctor or family members, or to increase their chances of receiving a certain treatment (Ubel et al., 2003). Patient values may also be subjected to recall bias as patients might not accurately remember what full health was like. In addition to having a better understanding of the implications of living in an impaired health state, patients may incorporate disease adaptation into their valuations.

Disease Adaptation

Disease adaptation refers to the process of adjusting to a new or altered health state. This can be achieved physically, through the use of corrective devices (e.g., prosthetics or walking cane), or emotionally, through an increased self-awareness of the impact of the health state on everyday activities. Adaptation may also be achieved through a patient's alteration of activities, desires, goals, and values. The nature of the adaptation depends on whether the individual's self-reported health state remains the same or changes. This thesis will focus specifically on those situations where individuals' self-reports of their health remain the same but their values for their states alter due to adaptation.

The concept of disease adaptation – or, more specifically, psychosocial adaptation – has been explored in the health psychology literature. The majority of these studies have investigated adaptation within a specific health condition (e.g., multiple sclerosis and cancers) and have not considered the broader question of how this adaptation occurs or how generalizable this process is to other conditions. Only a few studies have attempted to establish conceptual frameworks by which a patient may adapt to their chronic illness or disability (e.g., Devins, 1994; Bishop, 2005); however, there are disagreements about the identity of the individual components of the adaptation process (Livneh, 2001). The proposed frameworks appreciate that the uniqueness of each individual makes it difficult to adopt a generalized sequence to describe the process of adaptation.

In general, the proposed frameworks identify that the onset of a chronic illness tends to result in a reduction in the individual's overall QOL. This level of QOL may be disproportionately affected by the individual's degree of satisfaction within those life domains that are considered to be more important, or central, to the individual (Bishop, 2005). Devins (1994) postulates that chronic illness can affect QOL in two ways: reduction of opportunities to experience satisfaction in centrally important areas of life, and reduction of feelings of personal control. An individual may actively seek to achieve (e.g., by changing the focus of the disease impact on certain aspects of life) and maintain (e.g., by achieving perceived personal control) a maximum level of overall QOL. The perceived level of control may serve as a means for the patient to influence his/her ability to effectively deal with a difficult situation. As a result, the individual's behaviour towards the impaired health state is affected; this, in turn, may influence his/her psychological and physical outcomes. An individual's response to a chronic illness may also be affected by complex interactions related to the condition (e.g., symptom severity, functional limitation), the person (e.g., personality attributes, sociodemographic characteristics), and the environment (e.g., medical services availability, living conditions) (Livneh, 2001). Bishop (2005) further proposes that three responses may be observed when a reduction in QOL is experienced at the onset of a chronic illness: (i) an alteration of the importance of domains so that previously perceived as central – but highly affected – domains become less important while less affected domains become more central; (ii) an increase in perceived control (e.g., self-management) so that the impact of centrally-perceived domains is reduced; or (iii) a lack of change in the situation occurs, such that the individual continues to experience a decrease in his/her overall QOL.

Beyond the general frameworks discussed above, a number of other studies have investigated the specific details of adaptation. A fundamental component of adaptation is an increase in the ability of the patient to cope with the challenges posed by the chronic illness. This may be achieved through efforts to manage the psychological stress associated to illness-related losses (e.g., the impact of the loss of independence on psychological well-being) (Sinclair and Blackburn, 2008). Specifically, patients may adopt problem-focused and/or emotion-focused coping strategies to help them adapt to life in the impaired health state. Problem-focused strategies include efforts to alter a difficult situation (e.g., playing a musical instrument to replace previous participation in sports), whereas emotion-focused strategies involve efforts to change the relative meaning of what is, in fact, actually happening (e.g., distancing oneself from the situation) (Lazarus, 1993). These processes lessen the threat of being in an unfavourable, or different, situation by allowing the patient to build confidence about facing the challenges.

Another model that describes how a patient may adapt to a chronic health condition is response shift. This phenomenon describes occasions where a long-term change in health leads to changes in individuals' subjective standards – and, hence, in their self-reported QOL – despite there being no underlying change in their objective state of health (Spranger and Schwartz, 1999); a further discussion of response shift, in relation to the difference between patient and general population values, is presented in Section 3.1.2.

As there are wider and narrower definitions of adaptation, response shift can be regarded as a wider concept. In general, adaptation involves an individual's internal process to accept an impaired health state, possibly including changes to their lifestyle to maximize their QOL. Response shift, on the other hand, is simply due to social comparisons being made by the patient (Ubel et al., 2003). Specifically, patients may, when assessing their own QOL, compare themselves to other patients rather than to healthy individuals. In this thesis, the definition of adaptation will not include response shift. This decision was due to the complexity of informing the general population about the social comparisons involved with response shift.

While there may be no clearly good or bad approaches to enable a patient to adapt to a chronic illness, some are considered to be better, or worse, than others (Lazarus, 1993). Recently, Menzel et al. (2002) described eight constitutive elements of adaptation. These elements portray admirable attributes (*skill enhancement*, *activity adjustment*, *substantive goal adjustment*, and *altered conception of health*); non-admirable attributes (*cognitive denial of functional health state*, *suppressed recognition of full health*, and *lowered expectations*); and a combination of both attribute types (*heightened stoicism*). While the negative components highlight the psychological problems of living with a chronic health condition, the positive elements illustrate the strengthening of an individual's psychosocial capacity. The positive elements are achieved by patients developing greater ability from those skills unaffected by the health condition. This will eventually lead to the patient acknowledging their new health state, through the adjustment of personal activities and goals. These positive elements will then lead to a shift in the individual's perception of their health. In contrast to the positive elements of adaptation, lowered expectations involve individuals changing their perceived levels of achievement toward accomplishing a goal; this attribute is regarded as the least admirable of the three negative elements (Menzel et al., 2002). The final element, heightened stoicism, emphasizes the dynamic process of disease adaptation, in which happiness is controlled by what the individual interprets as achievable.

Although Menzel et al. (2002) categorized cognitive denial as a non-admirable element of adaptation, Lazarus (1993) deemed denial to combine both positive and negative adaptation attributes. Denial may be counterproductive – and dangerous – when the patient is interpreting his/her symptoms; this may result in delays in receiving help. Conversely, denial has been suggested to produce favourable responses for several adaptation-related outcomes of surgery (e.g., healing rate, hospitalization duration) (Lazarus, 1993).

The preceding paragraphs demonstrate that adaptation is a complex, multi-faceted, and individualized process that patients may undergo over the course of their illness. Therefore, the decision was made to align the central concept of adaptation in this thesis to that proposed by Menzel et al. (2002). The reason for this decision was the fact that the primary aim of this thesis was to explore the impact of disease adaptation information on general population values for different health states. To this end, it was more important to assess the specific aspects of adaptation that may encourage the general population to change their initial health state values rather than the overall adaptation process. The work by Menzel et al. (2002) was selected as it most closely represents the specific mechanisms relating to how a patient may adapt. The other identified models, such as Livneh (2001) and Bishop (2005), tend to focus more on the process rather than the underlying reasons for it, and hence are less applicable to this work.

2.2.2 General Population Values

The use of general population values is preferred over patient values from a policy perspective. When making decisions regarding the alternative allocation of healthcare resources, responses from the general population should be most advantageous for society at large. The Washington Panel on Cost-effectiveness in Health and Medicine advises that using a societal perspective in economic evaluations should yield fair weighting amongst all individuals and all activities (Gold et al., 1996). Rational citizens, operating behind a 'veil of ignorance' (i.e., an unawareness of their own future health states and needs), would give rise to decisions that lead to maximum aggregate benefit within that society. The average values of individuals without specific interest in particular health states would seem most appropriate from this perspective because this will maximize total health benefits.

The problem with values provided by members of the general population is that, due to insensitive health state descriptions, they may not understand the true implications of living in the impaired state. More importantly, the general population may neglect the fact that it is possible to adapt to the health state over time. While adaptation to a disease is a very natural process for patients living with an impaired health state, members of the general population, who have not experienced this adaptation, may have a difficult time anticipating it (Kahneman and Snell, 2000).

2.2.3 Policy Implications of Using General Population Values

The use of general population values can significantly affect the decision of whether or not a healthcare technology should be considered for funding. General population values can either benefit or hinder patients, depending on whether a cure is available for the health condition under investigation.

In situations where complete removal of all adverse symptoms can be achieved (i.e., full health), the use of general population values, rather than patient values, ultimately benefits patients (Gold et al., 1996). This is because members of the general population tend not to consider the possibility of adaptation; as a result, they give lower health state values when compared to patients' valuations. For example, if general population respondents value an impaired state at 0.5 but patients value it at 0.7 because they incorporate adaptation into their appraisals, a difference of 0.5 and 0.3, respectively, is observed relative to a full health state (i.e., a value of unity). As a result, there is a larger gain in QALYs for the general population values in the case of a 'cure' being applied (i.e., state returned to full health) (Brazier et al., 2005). This, in turn, yields a smaller ICER (Equation 2.1), when compared to a case where patient values are elicited, and, therefore, results in a higher priority for healthcare technologies aimed at either completely curing or preventing a condition.

However, for health conditions which currently have no known cures, such as rheumatoid arthritis (RA), the opposite trend is observed. For example, general population respondents may value an impaired health state that lasts for five years at 0.5 while patients value the same state at 0.7 over the same time period. These valuations lead to a lower QALY from the general population responses (i.e., 5*0.5 = 2.5 QALYs) compared to the patient responses (i.e., 5*0.7 = 3.5 QALYs). The lower general population values give lesser weight to saving the lives of the unhealthy than the healthy compared to the case if patient values were used. As a result, life-saving healthcare technologies would appear to be less attractive for decision-makers using general population values rather than patient values. Furthermore, members of the general population tend to undervalue transitions between different severe health states, as is described in further detail in Chapter Three. These effects could result in general population values resulting in inequitable health care resource allocation decisions.

As discussed above, the use of either patient values or general population values within an economic evaluation framework can afford different outcomes. This can impact the rank order of the ICERs – incremental cost per QALY gained – when compared to all other healthcare technologies under investigation. These rankings, also known as league tables, ensure that healthcare technologies that appear to represent the best value for money – greatest gain per pound Sterling spent – are funded first. This aims to maximize the net QALYs gained, subject to the resources available. However, whether one uses patient or general population health state values can significantly influence this rank order. To get around the limitations of solely using patient or general population values to guide healthcare decisions, researchers have proposed a third approach – that values provided by *informed* members of the general population should be used (Gold et al., 1996; Fryback, 2003; Ubel et al., 2003; Brazier et al., 2005).

2.3 Informed General Population Values

The use of 'informed' general population health state values provides an attractive solution to the normative debate of whether to use values either provided by patients or 'uninformed' members of the general population in a healthcare decision-making context. The aim of informing the general population is to provide sufficient information to the respondents such that they fully understand the health state they are being asked to value. The objective of informing the respondents is not to yield identical patient and general population values but rather to allow the respondents to incorporate all available information regarding the health condition into their own assessment, if they wish to do so. This is also regarded as an improvement to using general population values, which may be uninformed, or patient values, which may incorporate non-admirable adaptation attributes (e.g., cognitive denial, suppressed recognition of full health, and lowered expectations). The range of health state values provided by the patients is another significant concern. A patient entering into an impaired health state is unlikely to provide the same value as a patient who has lived in the state for some length of time. In fact, a *new* patient may provide similar values as an uninformed member of the general population (Dolan, 1999). As such, the use of informed general population values ensures that the limitations of solely using values provided either by patients or members of the general population are alleviated.

2.3.1 The Construction of Informed General Population Values

Currently, a challenge for researchers is to find ways to refine and enhance the elicitation of health state valuations such that respondents are informed and are able to effectively formulate their values for the health states. Although the descriptive system can never be absolutely complete, there are ways to ensure that the effect of illness on common or important life functions is not overlooked. For example, a leaflet discussing patients' experiences with the condition under investigation can provide such information. There is, however, a trade-off between achieving sufficient realism and overburdening the respondents (Brazier et al., 2005).

The act of providing respondents with more information – especially about adapting to life in the investigated health state – has not been empirically examined in sufficient detail. Consequently, the optimum method of informing members of the general population has not yet been determined. Possible techniques include: (1) providing information sessions to offer respondents more descriptive health states or information on the size and the nature of adaptation experienced by patients over time; (2) encouraging respondents to reflect and deliberate on the health states; and (3) presenting respondents with their personal values, as well as patient values, for the investigated health states.

Technique One: Information Session

There has been minimal effort devoted to producing more descriptive health states. Only a small number of studies have attempted to inform respondents about what it is like to live in the investigated health states. These studies used an array of different information techniques, including portraying symptoms of the investigated condition through multimedia presentations (Clarke et al., 1997), videos (Lee et al., 2002), and audio-recordings (Happich et al., 2005). The results of these studies are described in greater detail in Chapter Three; this current section presents more novel approaches aimed at informing respondents.

Recently, two studies simulated different states of visual impairment in a sample from the general population. While the study was small in size, Aballéa and Tsuchiya (2007) concluded that respondents' experiences of visual impairment *via* the use of spectacles were well received; respondents with pre-existing disabilities reported that they had found it difficult to envision the visual impairment alone. In another study, contact lenses were used to simulate the central vision loss related to age-related macular degeneration (Czoski-Murray et al., 2009). Both studies found that the simulated health states were more realistic than health states presented in written form. It provided the respondents with the opportunity to imagine themselves in the health state with minimal risk and discomfort, and reduced the cognitive burden placed on them. Such methods, however, are not practicable for conditions which would be difficult to simulate for unafflicted respondents; this includes the general restrictions on mobility induced by RA. Furthermore, the process of simulation cannot address the issue of long-term adaptation.

Adaptation Exercises

In the same vein as offering more descriptive health states, a few studies have incorporated adaptation exercises into their design (Ubel et al., 2005; Damschroder et al., 2005 and 2008). These studies adopted introspective approaches to encourage respondents to consider the possibility of adaptation. These studies are described in detail below.

Ubel et al. (2005) used a convenient sample from the general population (i.e., prospective jurors) to determine whether ratings of paraplegia (on a 0-100 scale) were influenced by one of three different adaptation exercises. The first of these, the 'broad adaptation exercise' encouraged participants to think about a previous life event that was emotionally difficult and to assess whether their emotion towards this particular event altered over time. These participants were also asked about specific adaptation processes associated with paraplegia (i.e., activities to aid physical and emotional adaptation) and whether they thought the experience with paraplegia would change over time. The second exercise, the 'narrow adaptation exercise' encouraged participants to think about a negative life event and assess whether their emotional response towards this event got stronger or weaker over time. The final exercise, the 'multiple time-points adaptation exercise', had the participants rate the QOL if they had paraplegia at two time-points: one month and five years. All participants were randomized to receive one of the three different adaptation exercises; they were then further sub-divided into two groups. For both the broad and narrow adaptation exercises, one group completed a set of ratings before and after administration of the intervention, while the other group completed the ratings only once, after the adaptation exercise. The purpose of having these two sub-groups within each exercise was to alleviate the potential that participants might 'anchor' on their initial estimate. For the multiple time-points adaptation exercise, one group rated the QOL of paraplegia at one month and then at five years, whereas the other group rated the scenarios in reverse order (i.e., five years then one month). The authors hypothesized that each of the adaptation exercises would result in an increase the individuals' QOL estimates.

In general, the results from the Ubel et al. (2005) study revealed that individuals accepted that a patient's view of life with paraplegia will improve over time. By encouraging them to consider the possibility of adaptation, the QOL ratings for paraplegia increased. For participants who provided ratings both before and after the broad adaptation exercise (n = 123), an increase in estimates was observed (i.e., from a mean (standard deviation, SD) of 47.0 (27.3) to 51.6 (27.2) on a 0-100 scale).

Those who provided estimates only after this adaptation exercise (n = 56) rated the QOL of paraplegia at 62.2 (19.7); this estimate was significantly higher than the pre-adaptation rating of the first participant group. For the narrow adaptation exercise, those who provided ratings before and after the intervention (n = 151) increased their estimates from a mean (SD) of 50.6 (26.8) to 58.5 (23.8). Those who only provided a rating after the intervention (n = 145) provided an estimate of 52.7 (23.7); this, however, was not significantly different to the aggregate rating of participants who had not yet undergone the narrow adaptation exercise. These results demonstrated that the broad and narrow adaptation exercises encouraged individuals to consider adaptation when valuing paraplegia. For the multiple time-points adaptation exercise, participants rated the QOL of being paraplegic at one month followed by at five years (n = 72) gave mean (SD) estimates of 46.4 (25.3) and 59.0 (29.0), respectively. Those who rated the QOL of being paraplegic in reverse time points (i.e., five years followed by one month) (n = 76) provided estimates of 47.7 (30.2) (at one month) and 49.8 (26.7) (at five years). Although a larger difference between ratings is observed for those who valued the QOL of being paraplegic at one month then five years, the aggregate QOL rating at five years was not statistically significantly higher than at one month for either valuation order. It is possible that the lack of difference between the values at the different time-points may be due to respondents anchoring on their initial rating.

Ubel et al. (2005) were the first to try to assess the impacts of disease adaptation on general population health state valuations. Since then, the results from two other studies that have used adaptation exercises have been reported in the literature. Damschroder et al. (2005 and 2008) informed the general population about disease adaptation using Ubel et al.'s (2005) narrow adaptation exercise.

In the first study, Damschroder et al. (2005) used person trade-off, another elicitation technique to those described in Section 2.1.1, to evaluate general population's preferences towards people with paraplegia. This approach requires individuals to make decisions using a social perspective. The study participants were advised that they were a regional health system director making a decision regarding the funding of two treatment programs. One group appraised the baseline life-saving treatment program of curing blood infections that would enable patients to return to full health and the alternate program of saving the lives of people with pre-existing paraplegia suffering from blood clots. The other group appraised the same baseline program (i.e., curing blood infections) and another alternate program of saving the lives of people with new onset paraplegia. For each group, the indifference point – the number of lives that needed to be saved in the alternate program to be equally good as saving 100 lives of previously healthy patients – was determined. Half the participants in each group received an adaptation exercise which encouraged them to consider their own ability to emotionally adapt to negative life events and to life with paraplegia. From a total of 359 participants, 178 received the adaptation exercise (i.e., 84 receiving the pre-existing paraplegia scenario and 94 receiving the onset scenario). The remaining 181 participants did not undergo the adaptation exercise prior to the valuation (i.e., 93 received the pre-existing scenario and 88 receiving the onset scenario).

The findings indicated that participants who received the adaptation exercise reduced the differences between the valuations of pre-existing and new onset paraplegia. Using the Mann-Whitney U-test, the median (interquartile range) indifference point for the pre-existing paraplegic treatment option was 102 (100-5,000) and 100 (100-300) for those not receiving the adaptation exercise and those receiving it, respectively; although the medians appear similar, the difference between the two distributions were statistically significant (p = 0.03). For individuals appraising the treatment program for people with newly onset paraplegia, the median (interquartile range) indifference point was 1,000 (100-1,000,000) and 102 (100-47,500) for those not receiving the adaptation exercise and those receiving it, respectively (p = 0.05). These results imply that, without the adaptation exercise, respondents place less value on saving the lives of patients who experience new onset paraplegia compared to saving the lives of patients with pre-existing paraplegia. After undergoing the adaptation exercise, respondents increased the value placed on pre-existing paraplegia (p = 0.03) and on new onset paraplegia, relative to saving healthy lives.

In their second study (a follow-up to their 2005 work discussed above), Damschroder et al. (2008) evaluated the impact of adaptation on health state values using the TTO and SG approaches; the adaptation exercise encouraged the participants (n = 1,284) to consider their own ability to emotionally adapt to negative life events. The health states that were investigated were paraplegia, below-the-knee amputation, colostomy, and severe back pain. Participants were randomized to value these states using TTO or SG; they were further randomized so half of them completed the valuation with an adaptation exercise, while the other half completed it without one. The adaptation exercises encouraged individuals to think back to an emotionally challenging event and assess how their emotions towards this event changed over time. The authors hypothesized that the adaptation exercise would encourage the individuals to give higher values for paraplegia, below-the-knee amputation, and colostomy states. They speculated that the adaptation exercise would not impact the individual's valuation of the severe back pain state because they felt that it would be difficult for people without severe pain experience to fully appreciate the extent to which pain may affect QOL. The results, however, did not meet their hypotheses. Specifically, the impact of informing the participants with the adaptation exercise did not result in values that were statistically significantly different from the values from the participants that were not informed about adaptation for any of the four health states. This result was independent of the valuation technique used. For example, the paraplegic state was appraised at mean (SD) TTO values of 0.79 (0.31) and 0.80 (0.29) for those who received (n = 325) and did not receive the adaptation exercise (n = 305), respectively; and at mean SG values of 0.63 (0.34) and 0.67 (0.31) for those who received (n = 241) and did not receive the adaptation exercise (n = 241), respectively. The authors speculated that a more personalized approach is adopted when responding to either a TTO or SG compared to the person trade-off used in their previous work (Damschroder et al., 2005). Although the person trade-off method involves multiple potential deaths from a societal perspective, the TTO and SG approaches require individuals to consider their own potential death. As such, individuals may be more realistic in assessing their own ability to adapt to the health condition when making trade-offs that involve their own death.

The aforementioned studies revealed that the use of adaptation exercises, in general, increased members of the general population's values for hypothetical health states; the only exception was the results obtained from the study of Damschroder et al. (2008), which showed no differences. This provided evidence that individuals do not consider the possibility of adaptation when valuing hypothetical health states with no further details given on adaptation. However, there are concerns regarding to how the studies were conducted. First, only Ubel et al. (2005) specifically mentioned that the study sample was powered at 80% to detect between-group differences, while the other two studies used the p-values of either parametric or non-parametric between-group tests to determine whether a difference actually existed. This raises the question as to whether the studies by Damschroder et al. (2005 and 2008) were sufficiently powered to detect any differences between the groups. This is a particularly important concern for Damschroder et al.'s (2008) study which reported no significant difference between the valuations of individuals that underwent the adaptation exercise and those that did not. For these studies, a post-hoc power calculation would have clarified the ability of the collected sample to detect differences between the groups. Second, although they asked individuals to consider a previous difficult life event as the adaptation exercise, the researchers were not clear as to what these life events were. Individuals may have thought of a non-health event (e.g., divorce, set back at work); this, however, may not be appropriate when the general population are valuing health states pertaining to a specific condition. There may be a discrepancy between the adaptation processes required to deal with, for example, a divorce and with a chronic health illness; this potentially may lead to an artificial change – either an increase or a decrease – in the value for the health state. An adaptation exercise of this type may not have engaged the participants fully about adaptation; this may explain the lack of differences observed in the health state values in the study by Damschroder et al. (2008).

Finally, the conclusions drawn from the valuation methods used in the three studies also raise some concern. The adaptation exercise was shown to affect individual's values using a rating scale (Ubel et al., 2005) and person trade-off (Damschroder et al., 2005) but not for the TTO and SG approaches (Damschroder et al., 2008). It is possible that the lack of influence of the adaptation exercise on the TTO and SG values may be a result of participants having to utilize a personalized perspective with these methods when compared to the person trade-off technique. Furthermore, unlike a rating scale, the TTO and SG methods require individuals to make a choice between life and death. As a result, individuals may be more conservative in their assessment of their ability to adapt to life in the hypothetical health states because they had to consider it relative to their own potential death.

Technique Two: Deliberation and Reflection

Another way of achieving informed general population values is to allow respondents more time to reflect on the different health states, and perhaps to deliberate with others; this may better reflect the way people make such decisions in real life situations. Deliberation is a type of discussion focusing on careful weighting of reasons for and against a specific issue (Johri et al., 2009). Shiell et al. (2000), in particular, stated that if values are at first based on uninformed judgements (i.e., individuals perceive the health state to be much worse than it actually is) but these judgements develop during the process of making the assessment, then the initial values may be an underestimation of the individual's preference for living in that health state.

The use of deliberation and reflection has been examined in several studies but the results have been inconclusive. Dolan et al. (1999) examined the effect of two group discussions (separated by two weeks) when setting healthcare priorities for different patient groups. The results from the study revealed that responses from a sample of the general population systematically differed after being provided with an opportunity to discuss the issues. At first, half the respondents wanted to give lower priority to smokers, heavy drinkers, and illegal drug users; however, after reflection and group discussion, they were indiscriminative towards them.

The impact that deliberation and reflection may have on health state values was also evaluated by Stein et al. (2006). This study explored the effect of group discussion on health state valuations. Although the discussion resulted in at least one change in the respondents' values over the course of the study, the overall impact of changes at the group level was negligible. The respondents reported in their semi-structured interviews that the group discussion was very important because it provided reassurance about their initial values, confirmed their assumptions regarding the health states, increased group cohesion, and satisfied their curiosity. This provided evidence that individuals' values became more informed by discussing the health states with other people. Although these studies demonstrated the advantage of including deliberation and reflection periods alongside the valuation process, further empirical studies are needed to examine whether this provided a valid means for members of the general population to become better informed about the health states they are valuing.

Technique Three: Presentation of Health State Values

The final method that can be used to inform members of the general population is to provide them with both their own values for hypothetical states and patient values pertaining to the same health states. Baker and Robinson (2004) suggested that, due to respondents having reservations about their valuations being used to guide treatment decisions, the respondents should be invited to reflect upon the responses they provided during the valuation exercise. Furthermore, patient values, collected at different time points, should be presented to general population respondents (Brazier et al., 2005). The patient values should reflect a series of events, such as the onset of disease, during the adaptation process, and after a period of adaptation.

Shiell et al. (2003) assessed the impact of providing respondents with their own health state values, as well as a reflection period. Through the use of control and intervention groups, the authors assessed the change in health state values over three time points. Individuals assigned to the intervention group had their values transcribed so that they could review them before the follow-up sessions. While this exercise elicited a change in most of the individuals' responses, there was no statistically significant impact at the aggregate level.

The provision of patient health state values has not been empirically evaluated and, therefore, its influence on general population values is currently unknown. However, the concern with this type of information is that respondents from the general population may focus solely on the numerical health state value rather than incorporating the underlying information into their assessment.

2.4 Summary and Conclusions

Current NICE guidance recommends that CEA using QALYs as a measure of effectiveness is the preferred method to appraise the relative merits of healthcare technologies. The QALY combines information pertaining to quantity and quality of life in a single index. While obtaining information about the quantity of life for use in the QALYs is explicit, there is a range of ways of eliciting values describing QOL. These include SG, TTO, and RS approaches, and multi-attribute health status classification systems.

While the use of the QALY in CEA is advantageous because it enables comparisons across different diseases, a debate exists amongst researchers regarding whose values for health states should be used when making decisions in the allocation of healthcare resources. Both patient and general population values are subject to biases and can lead to different results when incorporated into an economic evaluation framework. As such, the use of informed general population values may overcome the limitations of solely using patient or general population values.

The purpose of informed general population values is to ensure that respondents are aware of the pertinent information regarding the condition under investigation. They are allowed, if they wish, to incorporate this information into their health state values. However, the process of how to inform a respondent remains inconclusive. Possible methods include provision of a realistic description of the condition, a deliberation and reflection period, and actual comparisons with patient values.

In conclusion, this chapter demonstrates that there is a need for empirical evidence of how to effectively inform respondents about the health states they are appraising. However, before this can be achieved, the factors that result in the differences between patient and general population values need to be explored. Results from the following literature review, presented in Chapter Three, will shed light on the areas that need to be highlighted for the respondents during the information process.

Chapter Three

Exploring the Differences Between Patient and General Population Values: A Review of the Literature

While the choice of whose values should be used in an economic evaluation framework is a normative one (Williams, 1991), previous comparative valuation studies have shown that different health state values are obtained by patients and by members of the general population (e.g., Sackett and Torrance, 1978). The difference in health state values can lead to different outcomes when addressing the allocation of scarce healthcare resources, as demonstrated in the previous chapter. One possible reason for the differences observed between patient and general population values is the incorporation – and, similarly, the lack of incorporation – of adaptation. However, given the significance of whose values should be used when making allocation decisions in healthcare, researchers have attempted to extract other potential factors which may result in these differences (Ubel et al., 2003; Brazier et al., 2005; Stiggelbout and de Voel-Voogt, 2008). Despite these theories, there has been a lack of empirical evidence evaluating the etiologies of the differences observed between patient and general population health state values.

This chapter aims to review the current state-of-knowledge regarding why patient and population values differ by meeting three objectives. First, to present proposed theories speculating why patient and general population values differ. Second, to conduct a systematic review to identify studies that have reported health state valuations by *both* patients and members of the general population; the results of these studies are summarized. In view of the fact that rheumatoid arthritis (RA) is the representative disease chosen for this thesis, particular attention has been paid to studies that have examined *both* patient and general population values in this chronic health condition. Finally, to explore potential factors that may explain the differences observed between patient and general population health state values using quantitative methodologies.

3.1 Potential Sources of Discrepancies Between Patient and General Population Values

Ubel et al. (2003), Brazier et al. (2005), and Stiggelbout and de Voel-Voogt (2008) have proposed frameworks exploring the differences arising between health state valuations obtained from patients and general population respondents. This section is a synthesis of the information presented in the three aforementioned studies.

The main explanations for the differences observed between the values provided by patients and by members of the general population are that:

- the same health state is being compared differently;
- the measurement scale is being used differently; and
- disease adaptation is being incorporated into the valuation differently.

Each of these reasons is discussed in the following sections.

3.1.1 Different Comparisons of the Same Health States

The conditions under investigation are displayed in the form of health states for respondents to value. While the health states can be presented as either domains and levels or as vignettes, it is crucial that the health state descriptions be accurate. If the descriptive system ignores subtle differences within a domain, or even neglects significant domains, the potential for values provided by patients and by the general population to diverge is heightened. For example, the insensitivity of the description to address the health condition as a whole may result in respondents filling in information based on their own personal experiences or stereotypes which may be different for patients and members of the general population.

In general, the health state descriptions highlight symptoms commonly experienced by the patients. In an attempt to reduce the cognitive burden placed on the respondents when completing valuation surveys, life domains (as opposed to health domains) that are unaltered by the condition under investigation tend to be omitted from the descriptive system. As a result, respondents from the general population may disproportionately focus on specific activities that are negatively affected by the impaired health state and ignore other aspects of life, such as personal and spiritual relationships, which may be unaffected – or even enhanced – by a changed state of health (Dolan and Kahneman, 2008). This inattention to broad life domains and overestimation of the emotional impact of an event is known as *focusing illusion* (Schkade and Kahneman, 1998). This can be problematic when patients consider all aspects of their illness and when the general population only focuses on the negative aspects of a health state and ignores the remaining positive features (Ubel et al., 2003). For example, when members of the general population appraise paraplegic states, they tend to disregard the ability to communicate and spend quality time with family and friends, dwelling instead on the inability to use their lower limbs (Damschroder et al., 2005). This focusing illusion is speculated to affect health state values differently depending on the type of condition being valued; however, it is expected to greatly impact conditions that have major effects on only a few health domains (Ubel et al., 2003; Brazier et al., 2005).

A commonly cited non-health example of focusing illusion is the work conducted by Schkade and Kahneman (1998). In this study, students attending colleges in California and Michigan in the United States were asked to indicate not only their current happiness level but also their predicted happiness if they were to live in the alternative state. Groups of students in the two geographical areas reported similar levels of current happiness and predicted that they would be happier living in California than in Michigan. The latter outcome was due to the students narrowly focusing on the sunny weather commonly associated with California.

3.1.2 Different Usage of the Same Measurement Scale

Over a period of time, it is possible that there may be a change in the way patients report their quality of life (QOL). Known as response shift, it is a phenomenon that arises when changes in health lead to changes in individuals' internal standards, despite there being no underlying change in their QOL (Spranger and Schwartz, 1999); this can only be detected by asking the individual. The problem with response shift is intensified where respondents are asked to provide an overall value to describe their QOL.

A difference between patient and general population values may arise if patients make comparisons with other patients rather than with a healthy individual of a similar age. Patients might inflate their personal values as they are comparing themselves to other patients in poorer health rather than unaffected individuals of similar ages. Response shift is expected to increase for patients who have lived with the impaired health state for a long period of time compared to newly diagnosed patients.

The current state of health of an individual can also lead to a variation in the way they value health states. Due to their frame of reference, respondents from the general population tend to undervalue transitions between severe health states; this is known as prospect theory (Kahneman and Tversky, 1979). Figure 3.1 (page 66) demonstrates how values (represented on the y-axis) for different health states (represented on the x-axis) can differ depending on whether the individual is holding the viewpoint of the patient or the viewpoint of the general population. An individual's preference for different health state values describing gains and losses is represented by an *s*-shaped curve. The shape of such a curve highlights the importance of the perspective – or reference point, represented by the inflection point – the individual holds (Feeny and Eng, 2005). The curve is concave above the reference point, indicating health states that are preferred to the individual's current state, and convex below it, indicating health states that are considered undesirable relative to the current state.

Figure 3.1 illustrates the transitions between different health states based on two medical interventions that enable a patient, currently bed-ridden, to either use a wheelchair or reach a walking state (Lenert et al., 1999; Treadwell and Lenert, 1999). Due to their higher reference point (i.e., their current walking state), members of the general population (represented by the dashed curve on the right) regard being in a bed-ridden state (i.e., point 4) and being in a wheelchair state (i.e., point 5) as undesirable states to live in and hence give them lower health state values than being in the walking state (i.e., point 6). In addition, they see the outcomes from interventions enabling a bed-ridden individual to be upgraded to a wheelchair-mobile state as achieving only a small and insignificant change in value. Bed-ridden patients, on the other hand, with their lower reference points (represented by the solid curve on the left) regard the gain from the same transition between the bed-ridden state to the wheelchair state as a greater benefit (i.e., the difference between points 1 and 2). Conversely, the health gain from the patient's perspective in going from wheelchair to walking states (i.e., the difference between points 2 and 3) is not as great as observed from the perspective of the general population (i.e., the difference between points 5 and 6). This example shows that, when compared to patient values, the use of general population values underestimates benefits – and, hence, gains in the QOL for an impaired health states – for treatments which do not restore an individual to full health (i.e., only enabling a bed-ridden patient to gain a wheelchair-mobile state). This potentially contributes to the differences observed between patient and general population values.

Contrast effect occurs when someone with experience of chronic and/or major illness is less affected by acute and/or minor illness when compared to someone with no such experience (Ubel et al., 2003). In this case, patients may be unaffected by other negative events whereas members of the general population may overestimate the impact the illness may have on their well-being.

Shifting *inter-* and *intra-personal comparisons* may also play a role in the differences between patient and general population values. These comparisons refer to the individual's self-evaluation of his/her own current health which is partly determined by what health state one's peers are in and by one's own past experiences. A patient's initial experience with new illness will be very distressing if they compare it to their previous health; however, over time, their experience will not be so influenced by such comparisons. As patients come into contact with other patients, some of them with more severe conditions, their own level of distress may be reduced. The general population, on the other hand, is unaware of these effects, leading them to underestimate the QOL of the impaired health states.

3.1.3 Different Incorporation of Disease Adaptation in the Health State Valuations

The discrepancies between patient and general population values can also be attributed to the fact that patients incorporate disease adaptation when valuing health states, as discussed earlier in Section 2.2.1. Adaptation refers to the process of adjusting to a new or changed situation. While adaptation to a disease is a very natural process for patients living with an impaired health state, members of the general population have a difficult time anticipating adaptation (Kahneman and Snell, 2000). Even patients themselves may be unaware that they have adapted to their health condition. Although disease adaptation and response shift are considered as distinct concepts, they share a commonality in that they both affect the psychological perception of an individual's health. In contrast to response shift, the inability to consider disease adaptation is, to a certain extent, a failure to appreciate the fact that one's emotional response to the given change in circumstances will diminish over time.

Questions in a valuation survey often ask respondents to imagine what it would be like to live in an impaired health state. Patients may consider what it is like to live chronically with the condition whereas members of the general population may consider the immediate onset of the medical condition. The latter responses would result in lower health state values because they cannot imagine adapting to living in an impaired state over a longer term. Answers from the patients will depend, in part, on how long they have experienced the state; for example, patients who have only just started to experience the health problem are believed to report similar values to those of the general population (Dolan, 1999).

Healthy respondents tend to use 'transition heuristics' when appraising hypothetical health descriptions, which means that they focus on the time of entering into a poor health state and do not consider the possibility of adapting to that state (Stiggelbout and de Voel-Voogt, 2008). For example, when individuals are asked to consider a state of blindness, the general population will focus on the immediate impact of vision loss: the inability to see family members, watch television, read books, or to move around freely. They do not recognize that, over time, they could learn how to read Braille and to manoeuvre using a walking cane or with a seeing-eye dog. Furthermore, when asked about adaptation, respondents from the general population tend to underpredict their ability to adapt to a new situation (Kahneman and Snell, 2000), resulting in lower values from the general population when compared to patient values.

3.2 Methods

For the remainder of this chapter, a two-step process was undertaken to evaluate empirically the factors that may potentially influence the differences observed between patient and general population values. First, a review of the literature was systematically conducted and the retrieved studies were described. Second, using the studies extracted from the previous step, the health state values obtained from patients and members of the general population were explored in greater depth using statistical tests of association.

3.2.1 Literature Search

A review of the scientific literature identified those published studies that have empirically compared health states valued by *both* patients and the general population. Consistent and replicable searching strategies, such as those commonly employed in systematic reviews, were implemented. While there is potentially a virtually unlimited number of studies investigating patient and general population values – which, in addition to QOL research, may include other areas such as medical decision-making, health psychology, and sociology – the decision was made not to conduct an exhaustive literature review but rather to utilize the information drawn from pre-specified databases in a quantitative appraisal. This aims to empirically evaluate the factors that may induce the observed differences between patient and general population values.

The first stage of the review involved conducting an electronic search of all comparative valuation studies available in English published to date in journals referenced through Medline (PubMed). The search strategy used the following key words: 'health state*' and 'patient' and 'utilit*' and '(population or public or community)'. Studies had to have health state values elicited from *both* patients and members of the general population; studies were still eligible for inclusion if values were obtained from additional populations, such as healthcare professionals or family members. Studies were excluded from the review if health state values were obtained for *only* patients or *only* the general population.

A secondary search of the reference lists of the retrieved articles provided a further list of publications that were not identified in the primary search. A final search was performed using the cited reference search function based on the seminal paper in this area (Sackett and Torrance, 1978). This was done to identify any other articles that referenced this work through the Web of Knowledge. Any comparative valuation studies that attempted to compare patient and general population values in RA but did not fully meet the initial inclusion criteria were also summarized.

3.2.2 Descriptive Analyses

All studies that met the inclusion criteria were then categorized based on whether the results identified:

- no significant difference observed between patient and general population values;
- a significant difference observed between patient and general population values; and
- patient and general population values in RA.

Descriptive information extracted from the studies included details of the study populations (e.g., sample size, recruitment of general population respondents, information level of general population respondents); health states (e.g., derivation, presentation, states being valued by the patients); valuation techniques used (e.g., standard gamble, time trade-off, visual analogue scale); and disease type being valued (e.g., acute conditions, chronic conditions, cancers). While information about the size of the two respondent groups was included to examine the statistical reliability of the study results, the other variables were selected to test whether the differences between patient and general population values were due to health states being compared differently or disease adaptation being incorporated differently into the valuations; these hypotheses are described in greater detail in the following section. All study results were tabulated and compared as published: potential explanations for the variations between studies are presented and discussed.

3.2.3 Statistical Analyses

The mean patient and mean general population values reported by all studies were evaluated. In studies where multiple health states were appraised, the most severe health state value was examined in greater detail. The decision to use the values of the severe states in the analyses aimed to illustrate the most pertinent reasons that might explain the discrepancies observed between patient and general population values. In cases where the valuation method afforded a value not measured along a scale anchored from zero to one (e.g., visual analogue scale), the value was converted; for example, when the valuation scale was measured from zero to 100 but used the same anchors (i.e., zero for dead and 100 for full health), the resulting value was divided by 100.

The differences between mean patient and mean general population values were calculated for each study's valuation method (e.g., if a study used the standard gamble and time trade-off to obtain health states values, then this study would have two entries to correspond to the two valuation techniques). The differences were then ordered along a continuous gradient, where a positive number indicated that patient values were greater than general population values and a negative number indicated that patient values were less than those of the general population's. Paired t-tests were used to evaluate whether a difference existed between the mean patient and mean general population values. If a difference between patient and general population values was identified, independent t-tests and one-way analysis of variances (ANOVAs) were used to examine whether this difference was due to: (i) health states being compared differently or (ii) disease adaptation being incorporated differently into their appraisals. The presence of response shift (i.e., different measurement scale) was not explored because, as described in Section 2.2.1, it is primarily a mechanism by which patients compare themselves with other patients that are perceived to be worse off. However, it is not anticipated that members of the general population, when valuing hypothetical health states, are aware of this social comparison. As such, it is speculated that general population values, to all intents and purposes, are not affected by response shift. The impact of response shift on the differences observed between patient and general population values will not be quantitatively explored in this chapter because the general population are very unlikely to incorporate the impact of response shift on their own.

Whether the two respondent groups assessed the health states differently was evaluated as a function of:

- the derivation of the health states;
- the presentation of the health states; and
- the type of valuation method used to appraise the health states.

It was hypothesized that health states constructed by literature reviews and/or expert panels and presented as vignettes would yield similar patient and general population values because the health states would be richer in description when compared to health states constructed from multi-attribute health status classification systems and presented as domains and levels, respectively. As previous reports indicated that different valuation methods are prone to yield different results (Brazier et al., 2007), no hypothesis was generated as to which valuation method would contribute to the differences between patient and general population values.

In addition, the respondents' different incorporation of disease adaptation was evaluated as a function of:

- the type of disease being valued;
- the recruitment of the general population respondents;
- the information level of the general population respondents; and
- the health states being valued by the patients.

It was hypothesized that, when valuing acute health conditions, there would not be a difference between patients and general population values because the possibility for patients to adapt to these conditions is limited when compared to chronic diseases and, perhaps, cancers. Respondents from the general population who were conveniently recruited to participate in the study were expected to have values that differed from the patients'; responses from a specialized general population sample (e.g., students, employees) were expected to accentuate any stereotypes or biases related to the health state under investigation. Individuals who were informed about the condition under investigation, either by health state labels or by the specifics of the condition through an information session within the study design, were *not* expected to contribute to the differences observed between patient and general population values. Patients who valued their own health state rather than hypothetical states were expected to have different values from those of the general population because patients can incorporate adaptation into their assessments of their own health.

Due to the exploratory nature of this phase of the research, sample size weighting of the reviewed studies was not conducted. Significance was defined at $p \leq 0.10$. All statistical analyses were conducted using SPSS version 14.0 for Windows (SPSS, Chicago, USA).

3.3 Results

One hundred and fifty studies met the initial search criteria; only ten of these studies actually met the detailed review criteria, outlined previously in Section 3.2.1. The remaining 140 were excluded for various reasons: some elicited health state values for only one population group (n = 72); some were written in a language other than English (n = 3); some conducted decision-analytic modelling or were review articles (n = 52); some transformed patient values into general population values using mathematical transformations (n = 10); and others were conceptual/opinion articles (n = 3). An additional five studies were retrieved by scanning the reference lists of the included articles in the initial database search. Finally, a further 11 relevant studies were identified by conducting a cited reference search based on the article by Sackett and Torrance (1978). These methods generated a total of 27 studies to be critically appraised in this review. However, a further three studies retrieved in the initial search – excluded for transforming patients' responses into general population values – pertained directly to RA. These studies are also included in a separate section of this chapter, where they are compared with each other and with the one other RA study that actually met the specified inclusion criteria.

The majority of the studies included in this review implemented one of three commonly used valuation techniques – standard gamble (SG), time trade-off (TTO), and visual analogue scale (VAS) – with TTO being most often administered. The descriptions for the health states to be valued were commonly derived by reviewing the literature, consulting experts, or using the profiles from the multi-attribute health status classification system, EuroQol-5D (EQ-5D).

3.3.1 No Differences Observed Between Patient and General Population Values

Five of the 27 reviewed studies reported finding no significant differences between the valuations of patients and general population (Table 3.1, page 58). Clarke et al. (1996) obtained values for three Gaucher disease[3] states using descriptions developed from medical textbooks, which were then verified by physicians and

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[3] Gaucher disease is an inherited enzyme deficiency disorder. Common symptoms may include anaemia, fatigue, easy bruising and a tendency to bleed; an enlarged spleen and liver may also occur.

patients. In addition to patients and healthy volunteers, values from chronically ill respondents (e.g., individuals with rheumatic and respiratory illnesses) were also elicited; all respondents were informed that the health states pertained to Gaucher disease. Through the use of multimedia equipment, each of the three health states was introduced by patients currently living in the state. Respondents valued all states by SG, TTO, and risk-risk trade-off, a method that assesses the relative value of two uncertain events; all respondents also valued their own health by TTO. The results indicated no statistically significant difference in the values obtained from the healthy volunteers, from the patients with Gaucher disease, or from patients suffering from a chronic condition.

Korfage et al. (2007) also found no significant differences between patient and the general population values. In this work, an interactive internet questionnaire was used to elicit values for five prostate cancer states – without mentioning the cancer association – using both TTO and VAS techniques. The health state descriptions were developed using four of the five EQ-5D dimensions; 'pain or discomfort' was replaced with statements pertaining to urinary, bowel, and erectile function.

Three other studies also found no significant differences between patient and general population values. In a study of dentofacial deformities, Cunningham and Hunt (2000) used verbal descriptions and photographs of dental patients, and discovered that there were no statistically significant differences between patients and members of the general population when valuing such deformities by SG, TTO, and VAS. In addition, SG results from a study of diabetic retinopathy, vision loss due to diabetes, indicated that there were no significant differences corresponding to the different severities of the disease (Lloyd et al., 2008). In this work, besides obtaining values from individuals suffering from diabetic retinopathy and the general population, researchers also obtained values from patients at risk for diabetic retinopathy. The results revealed that the values did not differ depending on the type of respondent appraising the health state. Finally, Schmitt et al. (2007) examined values provided for health states pertaining to controlled and uncontrolled psoriasis and atopic eczema. Overall, members of the general population, who were recruited from among visitors to the surgical ward of the study centre, valued the health states similarly to the patients living with the conditions.

3.3.2 Differences Observed Between Patient and General Population Values

QOL research has, in most cases, reported that health state values provided by patients differ significantly from the values elicited from the general population; this result was observed in the remaining 22 studies identified in this review. The difference in values suggested either that people with disabilities incorporate their experience with the condition into their valuation (Ubel et al., 2003), or that members of the general population neglect the ability to adapt to an impaired health state over time, or perhaps a combination of both.

Patients Valuing Health States Lower than the General Population

Of the 22 studies demonstrating a difference in valuations between patients and general population members, six revealed that the impaired health state was valued lower by the patients. The characteristics of these studies are presented in Table 3.2 (page 59) and described below. Four of the six studies informed the respondents as to what the health state was being valued (Stein et al., 2003; Stolk and Busschbach, 2003; Souchek et al., 2005; Pyne et al., 2009); it was not clear whether labels were provided in the descriptions of the study methods of the remaining two studies (van der Donk et al., 1995; Lieu et al., 2008).

Health states describing treatment for laryngeal cancer were valued by patients with a previous history of laryngeal and floor-of-the-mouth cancers, members of the general population, and clinicians (van der Donk et al., 1995). Using SG, TTO, and VAS approaches, the authors discovered that both the general population and clinicians valued these health states higher than the patients.

Stolk and Busschbach (2003) elicited valuations regarding the QOL impacts of erectile dysfunction. The TTO results, based on valuations of 28 distinct health states, indicated that patients, on average, valued these health states lower than did the general population. Complicating these results was the fact that, although only men can experience erectile dysfunction, the general population sample included both male and female respondents. Even though the women were asked to imagine themselves as men when valuing these states, the sex of the respondent did not have a significant effect in explaining the differences between patient and general population values ($p = 0.38$) when included in a multivariate repeated-measure analysis. The authors suggest that, while differences were observed, patients and the general population respondents were “like-minded” towards living with erectile dysfunction. This was due to the fact that similar health state values were provided by patients and the general population, although the overall magnitude of appreciation differed.

Another health condition where values elicited from patients were lower than those from the general population was age-related macular degeneration, a type of visual impairment (Stein et al., 2003). Although the authors administered an unconventional TTO – asking respondents to predict how many additional years they expected to live and then asking them to make corresponding trade-offs in accordance to their hypothesized life span rather than imposing a fixed time frame – general population respondents, along with physicians, rated the mild, moderate, and severe age-related macular degeneration states higher than the patients. For example, for the severe state of age-related macular degeneration, values of 0.86, 0.82, and 0.57 were obtained for the general population, physicians, and patients, respectively. Statistically, there was no significant difference between the responses of the general population and physicians ($p = 0.39$) but the values provided by both these groups differed significantly from those provided by the patient group ($p < 0.01$).

Souchek et al. (2005) conducted a study where patients and representatives of the general population valued two scenarios pertaining to mild and severe osteoarthritis using both SG and VAS techniques; the health states were defined by EQ-5D profiles. While the primary purpose of this study was to investigate the valuations of these health states across different ethnic groups, the results indicated that members of the general population had higher values for the two health states than did patients, even after adjustment for respondent characteristics (e.g., age, sex, and ethnicity).

Lieu et al. (2008) investigated the perceptions that patients and the general population have towards preventing herpes zoster (or shingles). The general population, when compared to the responses of the patients, were willing to give up fewer days to avoid living with this health condition in all but the mildest case.

Recently, Pyne et al. (2009) compared patient and general population values pertaining to depression states. For this study, three types of patients were used: those with less severe current depression, those with more severe current depression and those with a previous history of depression but not currently depressed. Using health states derived from the Patient Health Questionnaire-9 and Medical Outcomes Short Form-12, depressed patients reported lower values for depression states when compared to the general population.

Patients Valuing Health States Higher than the General Population

In the 16 remaining studies that fit the inclusion criteria, the patients valued the impaired health states higher than did the general population; the key characteristics of these studies are shown in Tables 3.3a (page 60) and 3.3b (page 61). Due to the large number of studies demonstrating this trend, these results have been subdivided into studies evaluating: (i) acute conditions (n = 2); (ii) chronic conditions (n = 8); and (iii) cancers (n = 6). The rationale for this categorization was to evaluate the role adaptation plays in individual's valuations as a function of disease type; this is discussed previously in Section 3.2.3.

Acute Conditions

Two studies investigated the health state valuations for acute conditions (Murphy et al., 2001; Happich and von Lengerke, 2005). Of these, Murphy et al. (2001) examined the experience of severe stroke on health state valuations (Table 3.3a, page 60). Patients who had recently suffered a severely disabling stroke were matched by sex and age to a sample of individuals conveniently recruited from another hospital clinic; the latter group of respondents had to not have suffered a stroke. SG values were obtained from these two groups as well as from health professionals. The results revealed that the reported values varied widely across the three stroke states investigated; in general, the general population values were significantly lower than those of either the patients or the health professionals.

The other study investigating acute conditions focused on tinnitus (Happich and von Lengerke, 2005); this condition is defined by the perception of sound in the ear in the absence of corresponding external sounds. Members of the general population who were uninformed regarding the effects of tinnitus listened to samples of audio-recordings, which were developed on the basis of patient reports. The results revealed that patient values differed from those of the informed general population: as in the Murphy et al. (2001) study, general population values were lower than those of the patients. This result was consistent using any of the three employed valuation methods (SG, TTO, and VAS).

Chronic Conditions

Eight studies valued chronic health conditions in a wide range of clinical areas, including renal failure, osteoporosis, schizophrenia, haemophilia, and multiple sclerosis (Table 3.3a, page 60). Of particular relevance for this thesis is the work of Suarez-Alamazor and Conor-Spady (2001), which examined valuations in RA; however, these results are discussed in Section 3.3.3 alongside the other RA studies that were identified.

The seminal comparative valuation study, by Sackett and Torrance (1978), evaluated individuals' attitudes towards dialysis treatments (i.e., renal failure) and other health conditions. Patient and general population respondents valued scenarios that were developed in collaboration with clinicians. These scenarios – which included hospital and home dialysis, kidney transplant, tuberculosis, unnamed contagious disease, and breast cancer – described the physical, social, and emotional characteristics and the limitations of each health state. The study results revealed that living in the investigated health state played a role in the valuation; for example, patients currently undergoing home dialysis assigned a higher value to living in the impaired state than did members of the general population. Specifically, patients undergoing home dialysis valued this state at 0.56, whereas the general population valued it at 0.39. The labelling of conditions also influenced the valuations, as tuberculosis was valued more highly when compared to an unnamed contagious disease. Results pertaining to health states other than dialysis were presented aggregately and did not segregate between patient and general population values.

Since the publication of Sackett and Torrance's (1978) study, there have been additional studies valuing health states associated with dialysis. De Wit et al. (2000) compared values of patients on dialysis and a group of volunteer students using both TTO and SG methods. These individuals valued three hypothetical health states, which were described using the EQ-5D dimensions. The authors adjusted for the age differences between the two groups; however, this did not have a significant effect on the results. Patients consistently reported higher values for the states describing mild, moderate, and severe disease requiring dialysis. More recently, patients and members of the general population – matched by age, sex, race, and education level – were presented with a scenario describing the experience of a hemodialysis patient (Riis et al., 2005). As the authors hypothesized that under- or over-estimation of mood may be a key component of the discrepancies observed between patient and general population values, mood level for the specific health state was explored. For the Riis et al. (2005) study, general population respondents were asked to imagine that they had been patients for either one year or for as long as their matched patient and to provide a mood rating. Using a five-point response scale, all individuals rated the mood for the specified state, which ranged from 2 (very pleasant) to 0 (neutral) to -2 (very unpleasant). On average, the patients rated their own mood level at 0.70, whereas the general population rated it at -0.38.

Gabriel et al. (1999) examined the valuation of osteoporosis in women who had suffered hip fractures, women who had sustained one or more vertebral fractures, and women in the general population without known osteoporosis. They used the TTO method to value three hypothetical health states – established osteoporosis, multiple vertebral fractures, and disabling hip fracture – developed in consultation with clinical experts and women currently experiencing these states. The results revealed that the values for hypothetical osteoporosis health states of the subjects who had not experienced the health states were approximately 50% lower than the values provided by the women who had.

Studies investigating valuations of chronic health conditions have not been restricted to physical health states. Lee et al. (200) investigated the valuations of different schizophrenic health states amongst various stakeholders, including patients, family members, representatives of the general population, and health professionals. In an attempt to inform all the respondents, videos using trained actors depicting the types of mental health impairment were presented alongside the written health state descriptions. The results revealed that individuals with schizophrenia valued the health states higher than did the general population (p = 0.02). While the values for these states were similar for patients and family members, the values by health professionals and general population were lower.

Naraine et al. (2002) used the SG method to assess the value of seven haemophilia treatment scenarios amongst parents of haemophilic children, adults with haemophilia, and healthy individuals with some experience with haemophilia. Although haemophilia is an X-chromosome-linked inherited bleeding disorder (i.e., only males can have this condition), females from the general population and mothers of haemophilic children participated in the valuations. While all respondent groups favoured prophylactic treatment, which reduces the frequency of bleeding, parents of haemophilic children and adults with haemophilia rated the scenarios similarly to each other (p = 0.47) but provided substantially higher ratings when compared to the general population (p = 0.01 and 0.02, respectively).

The final identified study investigating chronic conditions, by Prosser et al. (2003), investigated values for multiple sclerosis. For this study, six health states describing multiple sclerosis were developed by searching the literature and consulting neurologists. These states were valued by a convenient sample of patients and members of the general population. The results indicated that patients assigned higher mean values for these states by SG, with the difference between the patient and the general population values becoming greater as the health states worsened.

Cancers

Six studies, which appraised different cancer states, demonstrated that patient values were higher than those of the general population (Table 3.3b, page 61). Boyd et al. (1990) compared the valuations in three groups – patients with rectal cancer, healthy general population members, and healthcare professionals – using SG and VAS. In general, patients with colostomies and physicians assigned significantly higher values to their health states than did either patients who had not experienced a colostomy or members of the general population. Jalukar et al. (1998) investigated the valuations provided by patients, healthcare professionals, and students (enrolled in an introductory-level psychology course), regarding head and neck cancer health states. Although the study results were not controlled for age, the TTO revealed that patients and healthcare professionals had similar values regarding life in health states pertaining to head and neck cancers. Students, however, valued these states lower, particularly for health states indicating more severe impairments or disfigurements. In another study, mild and severe hypothetical situations reflecting chemotherapeutic-induced toxicities were assessed using the TTO approach (Calhoun et al., 2004). The valuations were compared in four groups: women with ovarian cancer; women participating in an ovarian cancer detection program (i.e., at a higher risk for ovarian cancer); women from the general population; and physicians (both males and females). Overall, women either suffering from, or at a greater risk for, ovarian cancer assigned similar values to the various health states, while the general population assigned less favourable values when compared to the aforementioned groups. For example, for two severe toxicity states (ototoxicity[4] and nephrotoxicity[5]), patient values were 0.42 and 0.40, respectively, whereas women from the general population valued these states at 0.38 and 0.27, respectively.

The remaining three studies evaluated health state values in breast cancer. Using six scenarios to reflect the physical, emotional, and social health states of patients with breast cancer, Ashby et al. (1994) investigated the difference in values in patients, nurses, hospital doctors, general practitioners, and, as a proxy for the general population, staff members at a university (academic, library, secretarial, clerical, and ground staff). Although the results from this study were reported to be preliminary, the authors noted that the personal experiences of breast cancer patients appear to result in higher values. For example, women without experience of breast cancer valued the mastectomy scenarios significantly lower than women who had

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[4] Ototoxicity is the tendency of certain therapeutic agents to cause functional impairment and cellular degeneration of the inner ear and of the eighth cranial nerve. It may be reversible or irreversible.

[5] Nephrotoxicity is a poisonous effect of some therapeutic agents on the kidneys.

experienced breast cancer, most of whom had undergone a mastectomy. In another study, women – both patients and members of the general population – evaluating preferences for health states describing outcomes from breast cancer treatments favoured treatment and prevention options that involved minimal physical invasiveness (Cappelli et al., 2001). The SG results showed that women with breast cancer rated health states describing the outcomes from lumpectomy and radiation treatment more highly than women from the general population. Grann et al. (1999) investigated the valuations assigned to cancer states and prevention measures by women with breast cancer, women who were at high risk for breast cancer, and women from the general population; healthy women were further separated into two age groups: 20 to 32 years and 33 to 50 years. All groups valued having breast cancer higher than having ovarian and metastatic cancer by TTO. While patients gave the highest values for the health states, healthy women in the highest age range provided the lowest health state values.

3.3.3 Values for Rheumatoid Arthritis States

Using the pre-specified inclusion criteria, the literature survey only revealed one study that focused on the area of RA. Suarez-Alamzor and Conner-Spady (2001) obtained valuations for two arthritic states from patients, members of the general population, and physicians. Using health profiles obtained from the EQ-5D, respondents valued mild and severe states by VAS, TTO, and SG. Although similar values were obtained from all the respondents for the mild scenario, statistically significant differences, with the patient values substantially above those of the general population, were observed between the respondent groups for the severe scenario.

During the review process, three additional studies pertaining to RA were identified that did not meet the inclusion criteria (Table 3.4, page 62). These studies obtained valuations only from patients, but then used the results to predict the general population values. Balaban et al. (1986) used the Quality of Well-being (QWB) instrument as a method for obtaining respondents' valuations for RA states. While this instrument claims to be valid and reliable for assessing health outcomes in both the elderly and patients with specific chronic or disabling conditions (Kaplan et al., 1984), the QWB weights were derived from a sample of the general population. In the original study (Patrick et al., 1973), which reported the development of the QWB instrument, individuals were asked to rank 132 cards describing health states; these states included information containing: an age level; a function level (composed of mobility, physical activity, and social activity); and a symptom-problem complex.

Patients in the Balaban et al. (1986) study rank ordered the same 132 states as in the original study (Patrick et al., 1973) and a close agreement was found when compared to the general population weights. However, the study was not capable of evaluating the magnitudes of the values, and hence the patient values may still be higher than those of the general population.

Two additional studies used indirect methods to assess the values of patients and of the general population. Hurst et al. (1994) used both the health status classification system and the VAS of the EQ-5D for their comparisons. Using only patients in their study, the authors converted their responses on the classification system using the societal tariffs to form the EQ-5D indices based on TTO; these scores were inferred as 'general population' values. These indices were then compared to the VAS scores (e.g., patient values); the results indicated that general population respondents rate RA states more severely than patients who actually experience those states. Rashidi et al. (2006) used an algorithm to convert patient-reported VAS values into SG values, which were subsequently compared to patients' responses on the Health Utility Index 3; these latter values were inferred to be equivalent to 'general population' values. Although the results from the latter two studies indicated higher patient values, this result may be a function of obtaining 'general population' values using econometric modelling rather than being truly representative of the values that general population respondents would place on RA health states.

3.3.4 Exploring Why Patient and General Population Values Differ

As some of the 27 reviewed studies employed multiple valuation techniques in their design, 47 pairs of values were used to extricate the factors which might have led to the differences observed between the health state values of patients and the general population. Table 3.5 (page 63) shows the gradient of the differences in values. The differences between patient and general population values ranged from 0.41 to -0.38. The majority of these (n = 27 pairs) indicated that patient values were higher than the general population, with 16 pairs of values revealing a difference of 0.10 or greater.

The unadjusted paired t-test revealed that patient values differed statistically from those obtained from the general population (p = 0.03) (Table 3.6, page 65). This result provides further support to the apparent differences identified in the descriptive analysis of the literature review. Because of this result, independent t-tests and ANOVAs were conducted, as mentioned in Section 3.2.3, to examine whether this difference was due to: (i) health states being compared differently or (ii) disease adaptation being incorporated differently into their appraisals.

Is the Same Health State Being Compared Differently?

How the health states were constructed played a role in the differences between patient and general population values. Health states can be derived using a variety of methods; of the reviewed studies, the most common methods were consulting the literature and/or an expert panel (e.g., physicians and patients) (n = 15) and using EQ-5D profiles (n = 4). The use of multi-attribute health status classification systems, such as the EQ-5D, tends to select a small number of dimensions pertinent to the investigated condition. The use of a literature review and/or expert panel was expected to provide a richer health state description. The result from the independent t-test demonstrated that a difference between patient and general population values may be due to the use of health states constructed through the use of literature reviews and/or expert panels *versus* health states comprised of EQ-5D dimensions (p = 0.06).

While health states can be presented in different formats, the use of domains and levels to describe health states is believed to lack sensitivity in describing the investigated health states when compared to the use of vignettes. This was hypothesized to be a factor that may contribute to the discrepancies observed between patient and general population values. Although not controlled for the use of disease labels, the independent t-test indicated that there was no significant difference between patient and general population values based on how the health state was presented (p = 0.65).

Results from the one-way ANOVA indicated that the valuation method – SG, TTO, and VAS – did not influence the difference between patient and general population values (p = 0.85). This result was not unexpected as past reports have found that valuation methods yield significantly different responses within the same respondent group (Brazier et al., 2007). As a result, the valuation method used was not expected to significantly contribute to the difference between patient and general population values.

Is Disease Adaptation Being Incorporated Differently into the Appraisals?

A range of different health conditions was valued by the respondents in the reviewed studies. These were broadly classified into three categories (i.e., acute conditions, chronic conditions, and cancers). The one-way ANOVA revealed that, whether the respondents were valuing acute conditions, chronic conditions, or cancers, the difference was observed between patient and general population values is independent of disease type ($p = 0.31$). It was expected that patients with chronic conditions would incorporate aspects of adaptation in their valuation, thereby contributing to a larger difference between their values and the general population's; the statistical results, however, do not support this hypothesis.

While none of the studies specifically informed the respondents about disease adaptation, this section presents findings from studies that increased the respondents' awareness of the health states they were valuing. Six studies attempted to inform their respondents about the health states themselves. One would expect that providing additional information would alleviate the difference between both sets of health state values; the result from the independent t-test confirmed this hypothesis ($p = 0.31$).

It was anticipated that responses from conveniently recruited general population respondents would magnify any stereotypes or biases related to the health state under investigation. The statistical results indicated that the use of a conveniently recruited sample *versus* a representative sample did not have any effect on the difference between patient and general population values ($p = 0.53$).

The majority of the studies ($n = 17$) in this review opted to identify the health states so that the respondents were aware of the medical condition that they were valuing. This labelling resulted in a difference in patient and general population values ($p \leq 0.03$). This difference may be a result of the labels enabling patients to apply more information to their valuations, or members of the general population to introduce emotion and stereotype into theirs.

3.4 Discussion

The main, although not entirely unequivocal, finding of the published studies presented in this chapter is that patients value impaired health states differently from members of the general population; this was observed in 22 of the 27 reviewed studies. Of those 22 studies, 16 revealed that patients valued the impaired health states higher than the general population. The studies included in this review were considered to be heterogeneous, in terms of valuation methods, disease types, and study participants. Therefore, the results of the reviewed studies are synthesized and discussed in the aforementioned categories (i.e., valuation, disease types, and participants) in this section. The statistical results are presented alongside the descriptive results in order to provide greater insight into the differences observed between the patient and the general population values.

3.4.1 Valuation

Techniques Used to Value the Health States

The reviewed studies utilized a number of different techniques for respondents to value the health states. While most studies used the SG, TTO or VAS approaches, either alone or in combination, one study used only a response scale (Riis et al., 2005) and another used a risk-risk trade-off protocol alongside the SG and TTO (Clarke et al., 1997). However, the statistical analysis indicated that the type of valuation method used to elicit health state values did not influence the difference observed between patient and general population values ($p = 0.85$).

Amongst the studies reviewed, the TTO method was the most commonly used ($n = 18$). One study in particular administered an unconventional TTO in the valuation of visually impaired states (Stein et al., 2003). Rather than imposing a fixed time horizon for the valuation method for all respondents, the authors used the respondent's self-estimated life years as the time horizon. While this type of TTO provides a customized instrument for each respondent, the varying time horizons could impair the interpretation of the aggregate valuation obtained. Furthermore, the question can be raised whether the study was sufficiently powered to address the individually-varying time horizons of the respondents.

Methods Used to Construct the Health States

The method of constructing health states can have a substantial effect on the respondents' valuations. While the majority of authors conducted an extensive literature review, consulted specialist physicians or patients in the field, or used a combination of both procedures, four studies utilized the profiles from the EQ-5D. The brevity of this multi-attribute health status classification system can result in only a small number of the dimensions being relevant for the specific health state under investigation (Marra et al., 2005a). The brief description of the health states might result in the differences between patient and general population health state values when the health condition is named as the former most likely would inject their personal experience of living in the various health states into their valuation, while the latter's focus would be limited to the five dimensions of the EQ-5D. The statistical test of association revealed that the difference between patient and general population values may be influenced by how the health states were derived ($p = 0.06$) (i.e., states developed using a literature review and/or expert panel *versus* states developed from EQ-5D dimensions).

The presentation of the health states did not have an effect on the valuations. Ten studies described the health states using domains and levels; as mentioned above. four of them used the specific dimensions of the EQ-5D. Fourteen studies portrayed the health states in the form of vignettes. Three studies did not indicate how the health states were presented to the respondents. While there has not been an empirical study to assess the differences in responses between those who value health states using domains and levels *versus* those exposed to vignettes, it is assumed that the use of domains and levels would not create as descriptive a picture of the health states under investigation as would vignettes. When this hypothesis was tested in this review, there was no indication of a dependence of the differences between patient and general population values when the health states were described as domains and levels or when they were described as vignettes ($p = 0.65$) (Table 3.6, page 65).

Ideally, the differences between patient and general population values should be compared on the basis of patients valuing their *own* health while the general population values the same health states that these patients are living in. However, in this review whether the patients valued their own health state or the same hypothetical health state as the general population varied amongst the studies. The majority of studies requested the patients to value the same hypothetical health states as provided to the general population respondents; only one study strictly used patients valuing their own health states (Stein et al., 2003). The authors, looking at age-related macular degeneration, asked the patients to value their own health state. The patients' visual acuity, examined by a trained ophthalmologist, classified the individuals as to whether they had mild, moderate, or severe versions of the disease. Although this comparison may be more accurate in the normative debate of whose values should be used, the conclusions drawn from comparing the values of the general population and the patients may be inaccurate. For example, the description of the 'mild visual impairment' health state provided to the general population respondents may not be sufficiently similar to the 'mild visual impairment' defined by the ophthalmologist. To complicate the issue further, the authors do not state how the hypothetical health states provided to the general population were derived. Four other studies requested that the patients value both the hypothetical states and their own health (Clarke et al., 1997; De Wit et al., 2000; Murphy et al., 2001; Lieu et al., 2008). However, the severity of the patient's disease was not classified, making it difficult to compare the values from patients when valuing their own health and those from the general population when valuing the hypothetical health state.

3.4.2 Disease Types

It is difficult to make generalizations regarding the valuation trends across the various disease states because of the diversity of the conditions under which they were determined. Health conditions differ according to chronicity, prevalence, and affected population. Although this review consisted of studies appraising many types of diseases, there were multiple studies appraising patient and the general population values in the same disease area. Sackett and Torrance (1978) and De Wit et al. (2000) used TTO to obtain valuations for dialysis; however, the latter study also used SG. These studies found similar results; namely, patients valued their impaired states higher than did the general population. Riis et al. (2005) also conducted valuations in dialysis and found similar results as the above studies; however, the respondents valued mood on a response scale instead of directly valuing QOL. In the area of breast cancer, three independent studies reported that patient values were higher than those of the sample recruited from the general population (Ashby et al., 1994; Grann et al., 1999; Cappelli et al., 2001). From Table 3.3b (page 61), it appears that the valuation of cancer states consistently produced higher patient values; however, valuations for prostate cancer elicited similar responses from patients and the general population. Overall, the one-way ANOVA indicated that what disease the respondents valued did not affect the differences observed between patient and general population values ($p = 0.31$).

In addition to the disease itself, it is possible that pre-conceptions regarding the disease under investigation contributed to the discrepancies observed between patient and general population values. Although the majority of the studies in this review ($n = 17$) informed the respondents what health conditions they were evaluating, six studies did not provide labels for the health states. Connotations that arise from a disease label could be more detrimental than informative; this is shown when respondents valued an unnamed contagious disease higher than tuberculosis which is itself, of course, a contagious disease (Sackett and Torrance, 1978). This was further confirmed as the statistical tests of association showed that the use of labels contributed to the difference between patient and general population values ($p \leq 0.03$).

Rheumatoid Arthritis

Within the area of RA, the results obtained by Suarez-Almazor and Conner-Spady (2001) were consistent with the trends generally observed in this review; namely, that patients valued impaired health states more highly than did the general population. However, although osteoarthritis is similar to RA (both are degenerative joint diseases with similar impacts in terms of pain and limited mobility), it is surprising to note that the valuations for osteoarthritis opposed the reported RA results: osteoarthritis patient values were lower than those elicited from the general population (Souchek et al., 2005).

Three studies which examined valuations in RA but did not meet the inclusion criteria for the present review generated further inconsistent results (Table 3.4, page 62). Balaban et al. (1986) found no significant difference between the rank orders of health states provided by patients and the general population; however, as mentioned earlier, this did not directly assess quantitative valuations of the health states. Two other studies reported the same trends as Suarez-Almazor and Conner-Spady (2001) (Hurst et al., 1994; Rashidi et al., 2005), with patient values substantially higher than the general population's. However, it is important to point out that these results must be interpreted with caution as patient values were converted using previously derived tariffs to obtain 'general population' values.

3.4.3 Study Respondents

The use of a convenience sample for either the patient group or the general population group, or for both, was reported for 11 studies. Often recruiting patients in a convenient fashion is a combined result of the constraints of the condition under investigation (e.g., severe stroke) and the fact that the valuation techniques require mentally capable individuals to complete such activities in a reliable manner. The same arguments cannot be made for respondents from the general population, where, for example, the use of students (De Wit et al., 2000), university staff members (Ashby et al., 1994), or residents of an apartment complex (Prosser et al., 2005) may well provide valuations that do not simulate the responses that would be elicited from a truly random sample from the general population. Although students, staff members, and apartment residents constitute components of the general population, it is highly unlikely that these subgroups are generally representative, in terms of such characteristics as age or socioeconomic status. De Wit et al. (2000) attempted to address this issue by factoring the age differences between the two groups into their analyses; however, they found that this adjustment had no significant effect on their results. Similar to the previous study (De Wit et al., 2000), the independent t-test, which was conducted as a part of this review, found that the use of a convenience sample did not contribute to the differences observed between patient and general population values ($p = 0.53$).

A further issue pertaining to the study samples was size. Many of the studies reported small sample sizes for both patient and general population respondent groups. As most of the studies did not include sample size calculations, it was not possible to ascertain whether the study had the statistical power to detect differences, if present, between groups. This is particularly important for the studies which reported no significant difference between valuations (Table 3.1, page 58).

A concern with using conveniently recruited respondents and small sample sizes is the potential for selection bias. The validity of the results may be jeopardized by the difference in characteristics of those individuals participating in research studies and those choosing not to participate; the likelihood of detecting a true difference between patient and general population values may be affected. This concern could potentially be alleviated by matching the demographic characteristics of the samples (Murphy et al., 2001; Calhoun et al., 2004; Riis et al., 2005). The composition of the general population respondent groups was of particular importance for the studies that examined valuations in diseases pertaining to a single sex. While the general population valuations for the male conditions (e.g., haemophilia and erectile dysfunction) were elicited from all members, the general population valuations for the female conditions (e.g., osteoporosis – not exclusively a female health issue but commonly regarded as one – and ovarian and breast cancers) were restricted to only women respondents. The policy implications of segregating these general population valuations were not discussed in further detail. Stratified valuations by sex could potentially affect the observed differences, even if only one sex has the potential to be afflicted by the condition.

Informed Respondents

The majority of the studies presented in this review used general population respondents who were presumed to be ignorant of the disease state under investigation; six studies, however, made use of informed members of the general population. Two studies selected general population members who had experience with the disease under investigation: haemophilia (Naraine et al., 2002) and dental deformity (Cunningham and Hunt, 2000); although the authors do not clarify what this experience entailed nor whether all had the same level of experience. The four other studies informed the population participants *via* multimedia presentations (Clarke et al., 1997), information sheets (Cappelli et al., 2001), videos (Lee et al., 2002), or audio-recordings (Happich et al., 2005) pertaining to the health states under investigation. While Clarke et al. (1997) demonstrated that there were no differences between patient and general population values, the other studies found that patients still valued the impaired state higher than did the general population. Only Happich et al. (2005), however, attempted to evaluate the impact of informing the general population respondents on the resulting health state valuations. That study did not find that this process generated a measurable effect. When this factor was tested using statistical tests, informing the respondents did not contribute to the difference observed between the patient and general population values ($p = 0.35$).

In addition to the majority of the reviewed studies not informing the general population about the specifics of the health state under investigation, none provided information regarding the prognosis of the condition. Due to their direct experiences, patients can bring specific knowledge regarding certain stages of a disease. As a result, not controlling for prognosis was also expected to contribute to the differences observed between patient and general population values.

3.4.4 Disease Adaptation

None of the reviewed studies informed the general population respondents about disease adaptation, either within the health state description or as an additional source of information during the valuation process. As such, it is difficult to determine the influence of adaptation on the health state valuations; however, the results from the independent t-tests provided some indications that disease adaptation could have played a role in the differences observed between patient and general population values.

One of the initial hypotheses of the present work was that health states developed from literature reviews or expert panels, rather than EQ-5D dimensions, would minimize the differences between patient and general population health state values. The result from the independent t-test confirmed this hypothesis. While this may be due to more descriptive health states being developed from reviewing the literature or consulting experts, it may also be due to disease adaptation impacting patient values but not those of the general population when appraising states derived from the EQ-5D. Both of these factors may have increased the difference observed between the two sets of health state values. Also, the use of labels to define the health states was expected to contribute to the differences between patient and general population values; this hypothesis was confirmed. The presence of labels would allow patients to incorporate their experiences – and, hence, their ability to adapt – into their valuations. These results demonstrate the need for empirical work addressing the impact disease adaptation may have on an individual's valuations.

3.5 Study Limitations

This chapter was intended to highlight research which has compared patient and general population health state values and to shed light on the factors which may influence the difference between those values as observed in comparative valuation studies. There are, however, limitations within the present study design that must be borne in mind, as discussed below.

Firstly, the conducted review consists of research found in only two databases and does not claim to be exhaustive. The results presented in this chapter were intended to provide an empirical illustration of the possible factors which may influence health state valuations rather than to provide a definitive cause for the difference observed between patient and general population values. Secondly, values from only the severe health states, in cases where multiple health states were used, were use in the statistical analyses. The decision to use these values was to maximize the potential for identifying a difference between patient and general population values, if such a difference existed. Even using only the severe health state values in the analysis did not yield large numbers of statistically significant variables that could help to explain in the observed differences in valuations. Finally, since many of the reviewed studies used multiple valuation methods to appraise the health states, more sophisticated statistical approaches are needed in the analysis. These would account for multiple effects when the same study evaluated the effect of, for example, the derivation and description of health states using more than one valuation technique.

3.6 Summary and Conclusions

This chapter first identifies theories that have been proposed to explain the reasons for observed differences between general population and patient values. Then, a systematic, but not necessarily comprehensive, overview of studies comparing the health state values elicited from patients and the general population relating to specific disease states is provided.

Overall, the material presented supports the generally accepted understanding that health state values elicited from patients differs from those from the general population. While there is currently a lack of empirical evidence as to why this discrepancy exists, it has been proposed that members of the general population may be either valuing different health states or are not anticipating disease adaptation (Ubel et al., 2003; Brazier et al., 2005; Stiggelbout and de Voogt, 2008). The former effect may be an artefact of the descriptions of the health states themselves, while the latter may be a process that a population respondent may not consider when focusing on the initial onset of the disease. Because of these two possibilities, it is of particular interest to inform members of the general population of adaptation to ensure that they are both valuing the 'same' health state as the patients and are also incorporating adaptation into their valuation.

The debate pertaining to the use of patient versus general population values is hampered by the fact that some of the most commonly cited literature – for example, Hurst et al. (1994 and 1997) – may not be accurately comparing patient and general population data. Specifically, these studies transformed patients' self-reported values to generate 'general population' values. This was done, for example, using the social tariffs of multi-attribute health classification systems, such as the EQ-5D. Results from studies using such mathematical transformations raise questions regarding the validity of the comparisons made and the conclusions drawn relating to the differences in valuations between patient and general population respondents.

The collection of studies identified in this chapter demonstrates that the comparative valuation studies in the literature are very heterogeneous, in terms of the study respondents, valuation techniques, health state descriptions, and the amount of information provided to the respondents. This diversity may have led to the inconclusiveness of the synthesized results; this lack of clarity also indicates that there is a pressing need for further investigation to overcome the various limitations discussed. More importantly, there is a need to determine whether informing members of the general population about disease adaptation impacts their values for hypothetical health states.

To help meet this need, a new study needs to be designed to include a representative sample of the general population with sufficient statistical power to detect any differences that may arise between their initial values and values after being informed about disease adaptation. Such a study would have to be based upon health state descriptions that accurately reflect the conditions under investigation. Six of the reviewed studies used informed respondents, but the inconsistent results are an indication that the approach should be improved: none of the studies informed the respondents about disease adaptation. Therefore an intervention, effective at informing respondents about how patients adapt to living with RA, needs to be designed. Thus, the new directions and foci for the work discussed in the rest of this thesis follow directly from this review of the work to date.

3.7 Tables and Figures

Table 3.1: Studies showing equivalent patient and general population values[a]

Authors	Method	Health State			Patient	Sample	Participants		Disease	
		Descrip	Deriv	Label	Valuation	Size	Convenience	Inform	Specific	Type
Clarke et al. (1997)	SG, TTO, risk-risk tradeoff	Vignettes	Medical textbook	Yes	Hypo & own	Pat = 32 Pop = 39	Yes	Yes	Gaucher disease	Chronic
Cunningham & Hunt (2000)	SG, TTO, VAS	Verbal descrip. & photos	Expert opinion	Yes	Hypo	Pat = 57 Pop = 57	Yes	Yes	Dental deformity	Chronic
Korfage et al. (2007)	TTO, VAS	Domains & levels	EQ-5D	No	Hypo	Pat = 54 Pop = 53	No	No	Prostate cancer	Cancer
Schmitt et al. (2007)	TTO	Vignettes & photos	Expert opinion	N/A	Hypo	Pat = 58, 62^b Pop = 139	Yes	No	Psoriasis & atopic eczema	Chronic
Lloyd et al. (2008)	SG	Domains & levels	Lit review	No	Hypo	Pat = 122 Pop = 150	No	No	Diabetic retinopathy	Chronic

[a] Deriv: derivation; Descrip: description; EQ-5D: EuroQol-5D; Hypo: hypothetical states; N/A: not applicable, information not provided; Own: own state; Pat: patient; Pop: general population; SG: standard gamble; TTO: time trade-off; VAS: visual analogue scale; WTP = willingess-to-pay

[b] Two patient samples refer to 58 with psoriasis and 62 with atopic eczema

Table 3.2: Studies showing patient values lower than general population values[a]

Authors	Method	Health State			Patient	Sample	Participants		Disease	
		Descrip	Deriv	Label	Valuation	Size	Convenience	Inform	Specific	Type
van der Donk et al. (1995)	SG, TTO, VAS	Vignettes	Lit review & expert opinion	NA	Hypo	Pat = 10 Pop = 10	Yes	No	Larngeal cancer	Cancer
Stein et al. (2003)	TTO	N/A	N/A	Yes	Own	Pat = 115 Pop = 142	No	No	Age-related macular degeneration	Chronic
Stolk & Busschbach (2003)	TTO	Domains & levels	Disease-specific survey	Yes	Hypo	Pat = 106 Pop = 169	No	No	Erectile dysfunction	Acute
Souchek et al. (2005)	SG, TTO, VAS	Domains & levels	EQ-5D	Yes	Hypo	Pat = 198 Pop = 194	No	No	Osteoarthritis	Chronic
Lieu et al. (2008)	TTO, WTP	Vignettes	N/A	N/A	Hypo & own	Pat = 519 Pop = 527	No	No	Herpes zoster	Chronic
Pyne et al. (2009)	SG, VAS	Vignettes	Disease-specific survey	Yes	Hypo & own	Pat = 246 Pop = 95	No	No	Depression	Chronic

[a] Deriv: derivation; Descrip: description; EQ-5D: EuroQol-5D; Hypo: hypothetical states; N/A: not applicable, information not provided; Own: own state; Pat: patient; Pop: general population; SG: standard gamble; TTO: time trade-off; VAS: visual analogue scale

Table 3.3a: Studies showing patient values higher than general population values: acute and chronic conditions[a]

Authors	Method	Health State			Patient Valuation	Sample Size	Participants		Disease	
		Descrip	Deriv	Label			Convenience	Inform	Specific	Type
Sackett & Torrance (1978)	TTO	Vignettes	Expert opinion	Yes	Hypo	Pat = 129 Pop = 246	No	No	Renal failure	Chronic
Gabriel et al. (1999)	TTO	Vignettes	Expert opinion	Yes	Hypo	Pat = 183 Pop = 199	No	No	Osteoporosis	Chronic
De Wit et al. (2000)	SG, TTO	Domains & levels	EQ-5D	No	Hypo & own	Pat = 165 Pop = 103	Yes	No	Renal failure	Chronic
Lee et al. (2000)	SG, VAS	Vignettes	Meta analysis	Yes	Hypo	Pat = 20 Pop = 20	No	Yes	Schizophrenia	Chronic
Murphy et al. (2001)	SG	Domains & levels	N/A	Yes	Hypo & own	Pat = 11 Pop = 22	Yes	No	Severe stroke	Acute
Suarez-Almazor & Conner-Spady (2001)	SG, TTO, VAS	Domains & levels	EQ-5D	Yes	Hypo	Pat = 51 Pop = 104	Yes	No	Rheumatoid arthritis	Chronic
Naraine et al. (2002)	SG	Vignettes	Lit review & expert opinion	Yes	Hypo	Pat = 28 Pop = 30	No	Yes	Severe haemophilia	Chronic
Prosser et al. (2003)	SG	Domains & levels	Lit review	No	Hypo	Pat = 62 Pop = 67	Yes	No	Multiple sclerosis	Chronic
Happich et al. (2005)	SG, TTO, VAS	N/A	Lit review & expert opinion	Yes	N/A	Pat = 210 Pop = 210	No	Yes	Tinnitus	Acute
Riis et al. (2005)	5-point scale	Vignettes	N/A	Yes	Hypo	Pat = 49 Pop = 49	No	No	Renal failure	Chronic

[a] Deriv: derivation; Descrip: description; EQ-5D: EuroQol-5D; Hypo: hypothetical states; N/A: not applicable, information not provided; Own: own state; Pat: patient; Pop: general population; SG: standard gamble; TTO: time trade-off; VAS: visual analogue scale

Table 3.3b: Studies showing patient values higher than general population values: cancers[a]

Authors	Method	Health State			Patient	Sample	Participants		Disease	
		Descrip	Deriv	Label	Valuation	Size	Convenience	Inform	Specific	Type
Boyd et al. (1990)	SG, VAS	Vignettes	Lit review	Yes	Hypo	Pat = 51 Pop = 59	No	No	Colon cancer	Cancer
Ashby et al. (1994)	TTO	Vignettes	Lit review	Yes	Hypo	Pat = 17 Pop = 28	Yes	No	Breast cancer	Cancer
Jalukar et al. (1998)	TTO	Domains & levels	Lit review	No	Hypo	Pat = 49 Pop = 86	Yes	No	Head & neck cancers	Cancer
Grann et al. (1999)	TTO, VAS	Vignettes	Lit review & expert opinion	Yes	Hypo	Pat = 21 Pop = 135	No	No	Breast cancer	Cancer
Cappelli et al. (2001)	SG, VAS	Vignettes	Expert opinion	N/A	Hypo	Pat = 60 Pop = 51	No	Yes	Breast cancer	Cancer
Calhoun et al. (2004)	TTO	Domains & levels	Expert opinion	No	Hypo	Pat = 39 Pop = 39	Yes	No	Ovarian cancer	Cancer

[a] Deriv: derivation; Descrip: description; EQ-5D: EuroQol-5D; Hypo: hypothetical states; N/A: not applicable, information not provided; Own: own state; Pat: patient; Pop: general population; SG: standard gamble; TTO: time trade-off; VAS: visual analogue scale

Table 3.4: Studies showing discrepant patient and general population values: rheumatoid arthritis

Authors	Sample Size	Patient Valuation[a]	Derivation of General Population Valuation[*]	Result
Balaban et al. (1986)	288	Ranking health scenarios	Used previously derived population weight on the Quality of Well-being	Close agreement found between general population and patient (R = 0.94)
Hurst et al. (1994)	55	Patients' responses on EQ-5D VAS	Patients' responses on EQ-5D classification system	EQ-5D VAS (pat) = 0.60, EQ-5D classification system (pop) = 0.29
Rashidi et al. (2006)	320	Patients' responses on VAS & transformed to SG values	Patients' responses on HUI-2 & HUI-3	At the individual level, patient & population values show moderate to strong agreement but at the group level, SG (pat) values were greater than HUI (pop) scores

[a] EQ-5D: EuroQol-5D; HUI-2: Health Utility Index Mark 2; HUI-3: Health Utility Index Mark 3; Pat: patient, Pop: general population; SG: standard gamble; TTO: time trade-off; VAS: visual analogue scale

Table 3.5: Patient and general population values of the reviewed studies

Authors	Valuation Method[a]	Mean Values		Difference in Mean Values[b]
		Patient	General Population	
Gabriel et al. (1999)	TTO	0.84	0.43	0.41
Murphy et al. (2001)	SG	0.40[c]	0[c]	0.40
De Wit et al. (2002)	TTO	0.50[c]	0.20[c]	0.30
Jalukar et al. (1998)	TTO	0.40[c]	0.10[c]	0.30
Boyd et al. (1990)	VAS	0.81	0.53	0.28
Sackett & Torrance (1978)	TTO	0.56	0.32	0.24
Clarke et al. (1997)	RRTO	0.45[c]	0.24[c]	0.21
Grann et al. (1999)	TTO	0.89[c]	0.68[c]	0.21
Prosser et al. (2003)	SG	0.70[c]	0.49[c]	0.21
Ashby et al. 1994)	TTO	0.40[c]	0.20[c]	0.20
Happich et al. (2005)	VAS	0.54[c]	0.35[c]	0.19
Lee et al. (2000)	SG	0.62[c]	0.48[c]	0.14
Boyd et al. (1990)	SG	0.92[c]	0.80[c]	0.11
De Wit et al. (2002)	SG	0.42[c]	0.31[c]	0.11
Lee et al. (2000)	VAS	0.91[c]	0.80[c]	0.11
Suarez-Almazor & Connor-Spady (2001)	SG	0.66[c]	0.56[c]	0.10
Happich et al. (2005)	SG	0.88[c]	0.80[c]	0.08
Cappelli et al. (2002)	SG	0.24[c]	0.17[c]	0.07
Grann et al. (1999)	VAS	0.64[c]	0.57[c]	0.07
Naraine et al. (2002)	SG	0.90[c]	0.83[c]	0.07
Happich et al. (2005)	TTO	0.83[c]	0.77[c]	0.06
Riis et al. (2005)	Response scale	0.66	0.60	0.06
Calhoun et al. (2004)	TTO	0.42[c]	0.38[c]	0.04
Suarez-Almazor & Connor-Spady (2001)	TTO	0.58[c]	0.54[c]	0.04
Clarke et al. (1997)	TTO	0.82[c]	0.80[c]	0.02

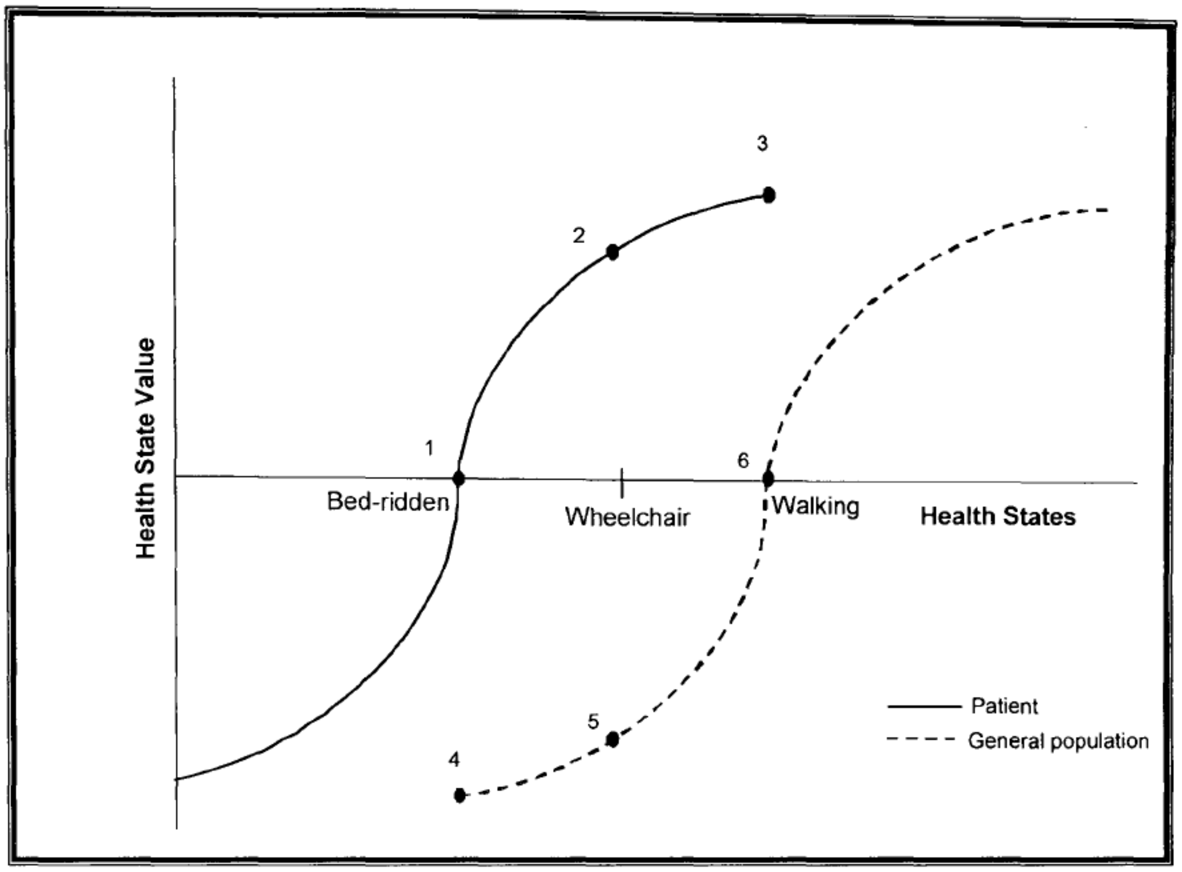
Korfage et al. (2007)	TTO	0.47[c]	0.46[c]	0.01
Cunningham & Hunt (2000)	SG	0.85	0.85	0
Cunningham & Hunt (2000)	TTO	0.75	0.75	0
Cunningham & Hunt (2000)	VAS	0.57	0.57	0
Lloyd et al. (2008)	SG	0.58[c]	0.58[c]	0
Cappelli et al. (2002)	VAS	0.06[c]	0.07[c]	-0.01
Souchek et al. (2005)	VAS	0.45[c]	0.46[c]	-0.01
Clarke et al. (1997)	SG	0.80[c]	0.82[c]	-0.02
Souchek et al. (2005)	TTO	0.77[c]	0.78[c]	-0.01
van der Donk et al. (1995)	VAS	0.60	0.64	-0.04
Korfage et al. (2007)	TTO	0.61[c]	0.66[c]	-0.05
Schmitt et al. (2007)	TTO	0.56	0.61	-0.05
Stolk & Busschbach (2003)	TTO	0.68[c]	0.74[c]	-0.06
Suarez-Almazor & Connor-Spady (2001)	VAS	0.44[c]	0.51[c]	-0.07
Souchek et al. (2005)	SG	0.66[c]	0.77[c]	-0.11
Schmitt et al. (2007)	TTO	0.45	0.56	-0.11
van der Donk et al. (1995)	TTO	0.66	0.80	-0.14
van der Donk et al. (1995)	SG	0.65	0.83	-0.18
Pyne et al. (2009)	SG	0.60[c]	0.83[c]	-0.23
Stein et al. (2003)	TTO	0.57[c]	0.86[c]	-0.29
Pyne et al. (2009)	VAS	0.49[c]	0.85[c]	-0.36
Lieu et al. (2008)	TTO	0.18	0.56	-0.38

[a] RRTO: risk-risk trade-off; SG: standard gamble; TTO: time trade-off; VAS: visual analogue scale.
[b] Determined by (patient value – general population value).
[c] Values for severe health states reported.

Table 3.6: Testing the hypotheses to identify the factors that contribute the difference between patient and general population values

Hypothesis	Number of studies	Pairs of health state values used	Values, mean (standard deviation) Patient	Values, mean (standard deviation) General Population	P-value
Do patient and general population values differ?					
Unadjusted model	27	47	0.60 (0.20)	0.55 (0.23)	0.02
Are patients and general population comparing the same health states differently?					
Derivation of the health states					0.06
EuroQol-5D dimensions	4	10	0.55 (0.11)	0.53 (0.05)	
Literature review &/or expert panel	15	24	0.64 (0.23)	0.54 (0.25)	
Description of the health states					0.65
Domains & levels	10	16	0.55 (0.12)	0.47 (0.23)	
Vignettes	14	23	0.61 (0.22)	0.57 (0.23)	
Valuation of the health states					0.85
Standard gamble	15	15	0.67 (0.21)	0.60 (0.28)	
Time trade-off	18	18	0.61 (0.19)	0.56 (0.23)	
Visual analogue scale	11	11	0.53 (0.23)	0.49 (0.18)	
Are patients and general population incorporating disease adaptation differently?					
Disease type being valued					0.31
Acute conditions	3	5	0.69 (0.17)	0.60 (0.29)	
Cancers	8	14	0.56 (0.24)	0.49 (0.27)	
Chronic conditions	16	27	0.60 (0.19)	0.57 (0.20)	
Presence of an information session					0.35
Yes	6	14	0.66 (0.27)	0.60 (0.29)	
No	21	33	0.58 (0.17)	0.53 (0.22)	
Use of conveniently recruited general population respondents					0.53
Yes	11	20	0.58 (0.14)	0.51 (0.25)	
No	16	27	0.62 (0.24)	0.58 (0.21)	
Use of labels to identify the health states					0.03
Yes	17	30	0.68 (0.17)	0.59 (0.24)	
No	6	8	0.53 (0.11)	0.44 (0.16)	

Figure 3.1: Illustration of the transitions between different health states[6]



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[6] This figure is reproduced from Lenert, L.A., Treadwell, J.R., and Schwartz, E.C. (1999). Associations between health status and utilities: implications for policy. *Medical Care*, **31**(5), 470-489.

Chapter Four

A General Overview of the Methodology

The earlier chapters of this thesis have examined the ongoing debate amongst researchers as to whose health state values should be the basis for decisions regarding health and healthcare issues (Brazier et al., 2007). Although general population values may seem more appropriate as a guide for decisions in the allocation of scarce resources, they, unlike patients living with the condition, may not recognize the ability to adapt to an impaired health state. For this reason, the judgement of *informed* members of the general population may be preferable (Gold et al., 1996; Fryback, 2003; Ubel et al., 2003; Brazier et al., 2005). The challenge is how to inform the respondents; that is, how to provide them with an understanding of the potential to adapt to an impaired health state over time.

As discussed in Chapter Two, the optimal method of informing members of the general population about disease adaptation, so that they are able to effectively make informed decisions regarding various health states, has not been established. For this reason, there is a need to develop an intervention that is capable of informing general population respondents about how patients adapt to their condition; rheumatoid arthritis (RA) is used as the case in point for this thesis. Alongside this main objective, there is also a need to explore the influence disease adaptation information may have on general population respondents' perceptions of RA, to quantify the impact that disease adaptation information has on their health state values, and to explore the factors that influence them to change their initial values after being informed about disease adaptation. In order to meet the above objectives, it is vital to construct accurate and comprehensible RA health states.

Thus, the objectives of this short chapter are twofold. First, to present the overall methodology used in the thesis and second, to address the research aims of the individual study components.

Overview of Methodology

4.1 A Mixed-Methods Study

A mixed-methods study design was employed for the project described in this thesis (Figure 4.1, page 71). Also known as multi-method or multiple methods research, a mixed-methods study involves the combined use of both quantitative and qualitative techniques to aid in – amongst other research activities – defining the question, designing the study, and performing the analysis (O'Cathain et al., 2007). For this project, a mixed-methods design was utilized because of the range of objectives that need to be addressed.

Specifically, a sequential mixed-methods study was used. This mixed-method approach aimed to meet the thesis objectives, as outlined previously in Section 1.3, by utilizing three distinct components in a sequential fashion. First, quantitative methodologies were used to construct health states that describe the symptoms experienced by a patient with RA. These health states were then incorporated into a qualitative component. Here, the health states served as an aid to gain an understanding of the effect disease adaptation information may have had on individuals' perceptions of RA. Furthermore, this component served to pilot-test the intervention aimed at informing the participants about disease adaptation. The qualitative results were used to design a questionnaire for use in the third, and final, component. In this final phase, a quantitative approach was undertaken to determine whether or not information about disease adaptation was effective at altering the respondents' initial health state values, as well as investigating the factors that contributed to these changes.

The three components described above are presented in this thesis in sequential order. A further discussion of the study design used in the qualitative and quantitative components of this thesis (Chapters Seven and Eight, respectively) is provided in Chapter Six, once the development of the health states has been described (Chapter Five). In the final chapter of this thesis, Chapter Nine, results from each of the components are synthesized together to highlight and discuss the key findings from this thesis. By 'mixing' the results, a greater understanding of the empirical findings can be gained.

As this thesis predominately uses quantitative approaches to meet the objectives, a third person voice is adopted to highlight the methodologies and its findings. However, in the chapter that describes the qualitative component (i.e., Chapter Seven), the first person voice is used. This change in voice acknowledges the potential influence the Ph.D. candidate may have had on the qualitative aspects of the research process (O'Cathain, 2009).

4.2 Overview of the Study Components

The three components of the sequential mixed-methods study are summarized here. A more thorough account of the methods employed in these components, as well as the results obtained, is presented in their relevant chapters.

4.2.1 Development of Rheumatoid Arthritis States for Use in Valuation Studies

In order to examine how members of the general population value life with RA, health states describing the common symptoms of the disease needed to be constructed. This was done on the basis of data collected by the National Data Bank for Rheumatic Diseases (NDB)[7] in Wichita, Kansas, United States (National Data Bank for Rheumatic Diseases, n.d.). This data contains longitudinal outcomes from rheumatology patients reporting on all aspects of their illnesses in detailed semi-annual questionnaires. These include, amongst others, the complete Stanford Health Assessment Questionnaire (HAQ) (Fries et al., 1980); visual analogue scales assessing overall severity, pain, fatigue, sleep problems, and gastrointestinal symptoms; and the EuroQol-5D (EQ-5D) (Brooks, 1996).

For this work, three RA descriptions were created using selected items of the HAQ as well as the pain and discomfort dimension of the EQ-5D. The HAQ, a mandated outcome measure for clinical trials in RA (Bruce and Fries, 2003), is a measure of physical disability. The instrument specifically assesses the ability of an individual to complete everyday tasks in dressing and grooming, arising, eating, walking, personal hygiene, reach, grip, and other activities. While the HAQ is a condition-specific instrument, the EQ-5D is a multi-attribute health status classification system that describes and values an individual's overall QOL (Brooks, 1996). (See Section 2.1.1 for a description of the EQ-5D.)

A detailed account of the development and description of the RA states, along with information about the HAQ, is presented in Chapter Five.

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[7] The NDB is a non-profit organization that performs research in RA, osteoarthritis, fibromyalgia, lupus, and other rheumatic diseases. Their research is designed to improve the treatment and health outcomes of patients with rheumatic conditions.

4.2.2 A Qualitative Assessment of the Influence of Disease Adaptation Information

A qualitative approach was undertaken to determine the impact disease adaptation information may have on general population respondents' perceptions of RA. The use of individual face-to-face interviews provided an opportunity not only to explore the research question in greater depth, but also to pilot the key features of the study design (to be described in Chapter Six). The results from the interviews were used to develop a questionnaire to assess an individual's reasons for changing or not changing his/her initial health state values. The constructed questionnaire was administered in the final quantitative component of this sequential mixed-methods study.

Chapter Seven addresses the methodology used in, and the research findings from, this qualitative component. The developed questionnaire based on analyses of the interviews is also presented in that chapter.

4.2.3 A Quantitative Assessment of the Influence of Disease Adaptation Information

Building on the results gained from the previous components, the final component of this thesis is a large-scale, quantitative assessment of whether or not information about disease adaptation altered general population respondents' values for the three RA health states. In addition, the factors which influenced individuals to alter their values were identified. Through the use of statistical tests of association and regression techniques, it was possible to determine the impact on their health state valuations of informing the respondents about disease adaptation.

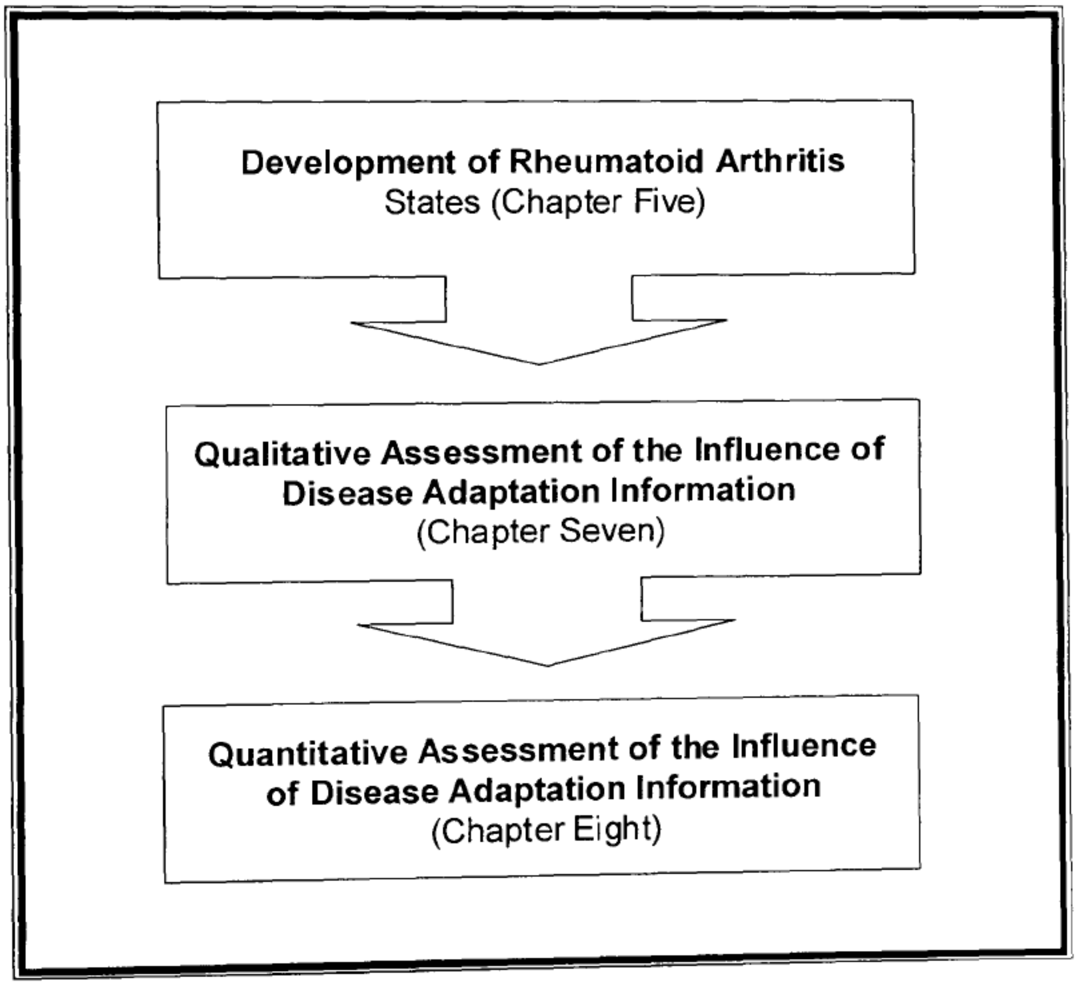
A detailed description of all statistical methods used, along with their results, for the final component of the sequential mixed-methods study is presented in Chapter Eight.

4.3 Summary

This chapter describes the sequential mixed-methods study design, consisting of three components, to be used in this research project. A mixed-methods design addresses the main objective of the thesis: whether or not the values for impaired health states are altered once respondents are informed about the ability to adapt to the disease under investigation. A secondary objective of this work was to explore the motivation for a respondent to make the decision to alter or not to alter their values for specific health states. The first component involved the development of RA states using novel statistical techniques while the remaining two components explored the factors which impact changes in general population values using a combination of qualitative and quantitative methodologies. A thorough discussion of the individual empirical components, the results of the statistical processing of the data, and the resulting interpretation, are presented in the subsequent chapters of this thesis.

4.4 Figure

Figure 4.1: Components of the sequential mixed-methods study



Chapter Five

Component One: The Development of Rheumatoid Arthritis States Using Rasch and Cluster Analyses*[†]

5.1 Introduction

Defining health states that describe the impact of rheumatoid arthritis (RA) on a patient's quality of life (QOL) are central to the work reported in this thesis. These health states form the basis of the valuations that members of the general population will make in the subsequent empirical studies (described in Chapters Seven and Eight). As such, the states need to credibly describe the symptoms patients may experience when living with varying severities of RA.

Health states can be developed using various methods, as described previously in Section 2.1.1. Expert judgements, such as those from physicians, permit a range of patient experiences to be elicited; however, these opinions are subject to biases. Physicians' viewpoints of health states may be distorted if their patients exaggerate their QOL in an attempt to please their doctor or family members. Patient responses, on the other hand, provide direct information about how the investigated health state impacts their lives. The use of patient interviews and focus groups allows researchers to gain in-depth knowledge about life in different health states but the results may be subject to volunteer bias, as the small number of participants might not be representative of most patients. For this reason, using responses on condition-specific instruments from a large sample of patients may be a better alternative in the development of health states.

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[*] A version of this chapter is published as McTaggart-Cowan, H., Brazier, J., and Tsuchiya, A. (2008). Combining Rasch and cluster analyses: A novel method for developing rheumatoid arthritis states for use in valuation studies. *Discussion Paper, 08/15*. Sheffield: Health Economics and Decision Science.

[†] A version of this chapter is currently under review for publication in *Value in Health*.

Typically, condition-specific instruments are comprised of numerous items addressing different aspects of the disease under investigation. However, for patients' responses on condition-specific instruments to be of use in the construction of health states to be used in valuation studies, the number of items included needs to be minimized to reduce the burden placed on respondents when completing the valuation exercise. In addition, responses on condition-specific instruments need to be combined in such a way that patients with similar QOL are grouped together so that distinct health states are formed.

One method that identifies the most representative items of unidimensional instruments is Rasch analysis. This technique converts categorical responses into a continuous latent scale (Rasch, 1960; Tesio, 2003). It has been employed in the development of QOL instruments (Tennant et al., 2004) and, more recently, in the construction of health state classification systems (Young et al., 2007 and 2009). While Rasch analysis is not the only method for selecting maximally discriminatory items, this technique was chosen because it offers a systematic approach to identify items while making the best use of the richness and sensitivity of the original instrument. As Rasch analysis identifies the instrument's most meaningful items, the cognitive and time constraints placed on respondents are minimized when they are valuing the health states. Furthermore, each item can be ranked in terms of severity; for example, from 'least severe' (i.e., an easy to complete item) to 'most severe' (i.e., a difficult to complete item). This classification ensures that health state descriptions can capture the widest possible range of severity. Rasch analysis aims to create a system that is unidimensional, ordered, additive, and of specific objectivity.

While Rasch analysis selects the representative items from the condition-specific instrument, there is still a need to identify different combinations of levels of these selected items to form distinct health states. A technique that meets this objective is cluster analysis. The main purpose of this statistical approach is to combine similar item responses; for example, individuals within the same cluster are more similar to each other than to individuals from different clusters. One of the first studies that employed cluster analysis to develop health states did so by identifying patterns in the physical and mental health domains of the Medical Outcomes Short Form-12 questionnaire (Sugar et al., 1998); other studies have since followed (e.g., Sugar et al., 2004; James et al., 2006). It is important to note that the aim of this process is not to provide clinical definitions of mild, moderate, and severe RA. Rather, the desired end result will be descriptions of three distinct RA states of varying severity levels, which describe potential specific health conditions for a patient with a stated level of RA.

This chapter aims to meet two objectives. The first objective is to describe the Rasch process and the rationale used for the final selection of the items to produce the health state descriptive system. The second objective is to present the use of cluster analysis to generate discrete health states to be used in the subsequent empirical studies of this thesis.

5.2 Methods

The health states were derived using a two-step process. Using previously collected patient-level data, the Rasch analysis selects those items of the Stanford Health Assessment Questionnaire (HAQ) that best describe the disability encountered by individuals living with RA; this condition-specific instrument is described below. The Rasch analysis approach used here is a modification of that proposed by Young et al. (2009). Cluster analysis, used to determine the item levels for each defined group, is adapted from the technique described by Sugar et al. (1998). To ensure that the final health states are plausible for those valuing them, the pain and discomfort dimension of the EuroQol (EQ-5D) (Brooks et al., 1996) was also incorporated in the health state descriptions.

5.2.1 The HAQ

For this thesis, the health state descriptions were primarily derived from items in the HAQ (the abridged version of the instrument is presented in Appendix A.3, page 243). The complete HAQ[8] is a widely used functional status questionnaire in rheumatic diseases (Fries et al., 1980). The instrument is well validated and sensitive across different outcome measures. Previous work has shown that the HAQ score predicts mortality (Wolfe et al., 2003), work disability (Wolfe and Hawley, 1998), joint replacement (Wolfe and Zwillich, 1998), and medical costs (Michaud et al., 2003). The instrument is commonly used to evaluate RA outcomes in clinical trials (Bruce and Fries, 2003).

The HAQ contains 20 items and assesses a respondent's ability to complete daily tasks in eight domains: dressing and grooming, arising, eating, walking, personal hygiene, reach, grip, and other activities. Two or three items comprise each domain.

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[8] Stanford University School of Medicine. *The Health Assessment Questionnaire* [online]. (2001). Available from URL: http://www.chcr.brown.edu/pcoc/EHAQDESCRSCORINGHAQ372.PDF [Accessed: 5 October 2009].

A description of the items is presented in Table 5.1 (page 100). Each item has four levels, representing no difficulty, some difficulty, or much difficulty in performing the task, as well as complete inability to perform the task. Respondents can select a score between zero and three, with higher scores implying a greater disability. The score on an individual item can also be increased by one point when the respondent requires assistive devices or additional help. For each domain, the item of greater value (i.e., most difficulty) yields the overall score for that domain. The domain scores are then averaged to afford an overall HAQ score.

While there are other condition-specific instruments that can assess disability in RA – for example, the Rheumatoid Arthritis Quality of Life questionnaire (de Jong et al., 1997) – the HAQ was chosen because it has been administered in various rheumatic populations for nearly three decades. The HAQ has been demonstrated to be valid (Marra et al., 2005a) and responsive (Marra et al., 2005b). It is also capable of obtaining information regarding both upper- and lower-limb disability in patients. Despite its popularity, the full HAQ is lengthy, posing a cognitive burden for respondents, and the scoring is not straightforward, especially when incorporating the respondent's use of additional devices and aids (Wolfe et al., 2004). The HAQ also has a floor effect: the insensitivity of the instrument at its lower end (i.e., mild limitations) of measurement is demonstrated by the fact that physically disabled individuals can be represented by decent (i.e., lower) HAQ scores (Wolfe et al., 2004).

5.2.2 The Dataset

An anonymized dataset containing information from RA patients living in the United States (US) (Wolfe, pers. comm., 18 September 2007), obtained from the National Data Bank (NDB) for Rheumatic Diseases in Wichita, Kansas, was used for this study. The NDB contains longitudinal outcomes research data from rheumatology patients reporting on all aspects of their illness (National Data Bank for Rheumatic Diseases, n.d.). This information is collected from enrolled patients every six months using a detailed postal survey. The survey includes the complete HAQ; visual analogue scales assessing overall severity, pain, and fatigue; and the EQ-5D. Participants are asked to return the completed survey in a provided pre-paid envelope. While no compensation is given to the respondents for completing the survey, $1,000 prize lotteries are offered for those individuals who return the completed survey within two weeks.

The sample size used in the development of RA health states is based on the fact that Rasch analysis is sensitive to large sample sizes. Using too large a sample generates a greater frequency of statistically significant items (Rasch, 1960), making item reduction difficult. A review of the literature indicates that a range of sample sizes has been used (Uprichard et al., 2009; Lundström and Pesudovs, 2009) with studies typically using between 400-700 responses. Based on these findings, a dataset size of 600 patients has been selected for the work reported in this chapter.

Within the dataset, an equal number of individuals in each severity group (n = 200) was included to ensure that each level of disability was well represented[9]. The classification of severity was determined by the patients' total HAQ score, such that individuals with scores of less than one, between one and two, and greater than two were broadly classified as to having 'mild', 'moderate', and 'severe' RA, respectively. If a true RA population had been used, then there would be a greater number of individuals classified with having 'mild' and 'moderate' RA. However, in the development of the health states for this thesis, it was more important to have a good distribution of responses for each level, rather than obtaining a sample that would be proportionate to the prevalence of each severity.

The dataset provided by the NDB was characterized using descriptive statistics, in terms of age, sex, RA duration, HAQ score, EQ-5D index, and unstandardized EQ-5D visual analogue scale (VAS) value. Continuous variables are presented as means and standard deviations (SD), while categorical variables are presented as the proportion of the sample within each group. Each severity subgroup, in terms of HAQ range, was characterized by the mean RA duration, mean HAQ score, mean EQ-5D index, and mean EQ-5D VAS value. SPSS Version 14.0 for Windows (SPSS, Chicago, USA) was used to evaluate the descriptive statistics.

5.2.3 Initial Criteria for Reducing the HAQ

One of the goals of this study was to reduce the 20-item HAQ to obtain items that best addressed disability in the given dataset of RA patients. In addition to the statistical methodology used to reduce the HAQ items, *a priori* criteria were set to ensure that the final health states would contain items that would best describe the situation to an individual who might not be informed about RA. To this end, it was vital that the final set include a combination of upper- and lower-limb disabilities; this is discussed further in Section 5.3.2, below. Furthermore, each of the items

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[9] Dr. Wolfe, founder of the NDB, randomly selected the individuals from a dataset of 23,261 RA patients.

selected had to belong in a separate domain of the HAQ; this was done to avoid any potential for collinearity. Finally, the items had to capture the widest range of severity possible. This final criterion was verified by the distribution of the items along the difficulty (logit) scale (to be described in Section 5.2.5) during Rasch modelling.

In addition to the above criteria, it was necessary to complement Rasch analysis with psychometric techniques to provide greater strength in the results. Therefore, the frequency of responses and the internal consistency – the correlations between item and domain scores – were also evaluated for the HAQ responses (Young et al., 2009). If some items elicited poor responses (e.g., low frequency) or poor internal consistency (e.g., weak correlation), those items were considered to be less representative of disability for the given dataset.

The internal consistency of the HAQ responses was tested using Spearman's correlation coefficients. As in previous work (e.g., Young et al., 2009), the cut-off value for this criterion was rho ≤ 0.7.

Questions pertaining to the use of assistive devices in the HAQ were excluded from the analysis, as done in previous studies (Tennant et al., 1996; Wolfe et al., 2004). Although the cited papers do not explicitly state the rationale for excluding this information from their Rasch models, it was felt the relationship between the uses of aids and the HAQ items was ambiguous. For example, the use of a walking cane may directly relate to the walking domain (i.e., walking on flat surfaces and climbing up steps) but it is possible that this device may also aid in the arising domain (e.g., standing up from a straight and armless chair). This potential correlation made it impossible to accurately determine which aid corresponded to which item, making it difficult to incorporate such aspects in the modelling procedures.

The psychometric analyses – frequency of responses and internal consistency – were performed using SPSS versions 14.0.

5.2.4 Initial Observations on the Context of the HAQ Items

Although the 20 items in the HAQ are distinct activities describing disability (Table 5.1, page 100), there were concerns regarding the applicability of the items across different individual groups (e.g., the sex of the respondents may affect how they respond to different items). The following discussion assesses which HAQ items are likely contenders for inclusion in the final health states prior to conducting Rasch analysis[10].

The *dressing and grooming* domain comprises the ability to dress oneself (*dresself*) and the ability to shampoo one's hair (*shampoo*). While these items address disability in the upper limbs, the ability to shampoo one's own hair may result in discrepancies between sexes; for example, older males may be less likely to perform this task due to an absence of hair (Wolfe, pers. comm., 18 September 2007).

The *arising* domain is composed of the ability to stand up from a straight chair (*standup*) and the ability to get into and out of bed (*inbed*). A combination of both upper and lower limbs is used in completing these tasks. *Inbed* has consistently resulted in missing responses, as well as response discrepancies between males and females for unknown reasons (Wolfe, pers. comm., 18 September 2007). It is possible that *standup* may result in confusion as some straight chairs may have arms on them (Wolfe, pers. comm., 19 December 2007). To prevent any potential misunderstandings that may arise, the word 'armless' would be included in the description if this item was selected to be included in the final health state.

The *eating* domain encompasses one's ability to cut one's own meat (*cutmeat*), the ability to lift a full cup or glass to one's mouth (*liftcup*), and the ability to open a new milk carton (*openmilk*). These items specifically address disability in the upper limbs. The ability to open a new milk carton may be interpreted differently amongst respondents, as there are many different designs of milk cartons in common use (Wolfe, pers. comm., 18 September 2007). The ability to cut meat may also be considered passé, as people's dietary habits have evolved over time, due to health and cultural beliefs.

The *walking* domain includes both the ability to walk outdoors on flat ground (*walkflat*) and the ability to climb up five steps (*climstep*). Both items are believed to be classic symptoms of RA that address lower-limb disability.

The *hygiene* domain includes the ability to independently take a bath in a bathtub (*tubbath*), get on and off the toilet (*ontoilet*), and wash and dry one's body (*washbody*). These items, like those comprising the *arising* domain, may require a combination of using both upper and lower limbs to be successfully completed.

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[10] These views are those of the Ph.D. candidate, based on discussions with Dr. Wolfe and the Ph.D. supervisory panel, and also in consideration of the conceptual underpinnings of the items.

While these items are considered to be daily, mundane activities, the *tubbath* item may be problematic, as the prevalence of individuals taking tub baths, since the development of the HAQ in 1980, may have decreased due to the increased number of homes with only showers installed. Furthermore, there is a potential correlation between the items *tubbath* and *washbody* for those people who would normally take a bath as a way of washing themselves.

The *reach* domain is comprised of the ability to reach above one's head and get down a five-pound object (such as a bag of sugar) (*overhead*) and the ability to bend down to pick up clothing from the floor (*benddown*). Both items address a combined upper- and lower-limb disability.

The *grip* domain involves the ability to open a car door (*opencar*), the ability to open previously opened jars (*openjars*), and the ability to turn faucets – or taps, as they are more commonly known in the United Kingdom (UK) – (*fauceton*). These items focus on upper-limb disability. The task of opening car doors may potentially not be applicable to everyone, as some individuals may rely solely on public transportation or not drive themselves. An increasing prevalence of lever taps may also alter the 'grip' concept in operating taps.

The final domain, *activities*, includes the ability to run errands and shop (*runerand*), the ability to get in and out of a car (*inoutcar*), and the ability to do chores, such as vacuuming or yard work (*vacuum*). These items demonstrate a combined upper- and lower-limb disability. It is believed that the vacuum and yard work item may potentially elicit social class response differentiation. For example, individuals of higher socioeconomic classes may employ someone to clean the house and/or work in the garden. Furthermore, individuals may rely on their travel companions to help them in and out of the car.

While the preceding discussion of the domain descriptions was conceptual in nature, it will also play a role in the final selection of HAQ items. Carefully considering the context and interpretation of the original items ensures that the resulting health states create a descriptive picture of distinct RA states which respondents from the general population will be able to understand and value.

5.2.5 Selection of HAQ Items That Best Describe Disability

A maximum of five items was proposed for the development of the health state classification system. Five items were chosen as there have been a number of instruments constructed with five domains, such as the EQ-5D (Brooks, 1996) and the Asthma Quality of Life Utility Index (Yang et al., 2007). There are other systems with even greater numbers of domains – for example, the Health Utility Index has eight (Torrance et al., 1996; Feeny et al., 2002) – but developing a classification system with more than five domains for use in the latter components of this mixed-methods study was believed to introduce unnecessary complexity for the respondents.

While the main purpose of using Rasch analysis in this study is to reduce the number of items in the health state descriptions, and hence the cognitive burden placed on study participants when appraising the health states, it is also essential that the constructed states be descriptive and plausible for members of the general population to comprehend. In addition to the limitations encountered when performing daily tasks, such as those described by the items in the HAQ, pain and discomfort is commonly experienced amongst most patients with RA (National Rheumatoid Arthritis Society, 2008). It is therefore vital that this domain be included to create a realistic picture of the chronic condition being valued.

In the full HAQ, the level of pain experienced is measured continuously on a VAS, ranging from no pain to severe pain. While the results from a VAS are meaningful, within a Rasch framework, discrete rather than continuous variables need to be utilized in the modelling process, as discussed later. Consequently, for this work, the aspect of pain and discomfort for the health status classification system was captured by the dimension of the same name in the EQ-5D (Appendix A.2, page 241), which uses a discrete set of levels. As it was decided *a priori* that one of the five items describing the health states had to represent pain and discomfort, only four items were selected from the HAQ.

Rasch Analysis

Rasch analysis was selected as the main technique to develop the health states because it offers an opportunity to make the best use of the richness and sensitivity of the original instrument (Young et al., 2009). In lieu of Rasch analysis, factor analysis and qualitative methodology could have been employed in the development of health classification systems. However, Rasch analysis was chosen because it can combine information about the severity of the instrument's items with information about the ability of the patients. This will prove to be beneficial as the end result will be health states representative of individuals living with RA yet capturing the widest possible range of severity. When directly compared to factor analysis, which examines multi-dimensionality within an instrument, Rasch analysis is more appropriate because the HAQ addresses disability as a single dimension.

The concept of Rasch analysis is illustrated by the following example. Patients with RA were given a hypothetical instrument consisting of the following items: difficulty lifting a cup to the mouth; difficulty getting in and out of bed; and difficulty walking. The respondents were evaluated based on whether or not they have experienced the aforementioned symptoms. In terms of severity, difficulty lifting a cup to the mouth is considered to be more severe than difficulty getting in and out of bed, which, in turn, is more severe than difficulty walking. For this example, three patients responded to this instrument and their responses are:

- Patient 1 experiences difficulty lifting a cup to the mouth, difficulty getting in and out of bed, and difficulty walking;
- Patient 2 experiences difficulty getting in and out of bed and difficulty walking; and
- Patient 3 experiences difficulty walking.

Under the Rasch model it is assumed that the underlying QOL of Patient 3 is better than Patient 2, who, in turn, has a better QOL than Patient 1, who has the worst QOL. Furthermore, it is assumed that patients experiencing difficulties lifting a cup to the mouth have a greater detriment to their QOL than patients with difficulties getting in and out of bed and difficulties walking, respectively.

Rasch analysis verifies that the scale of the instrument is unidimensional. Unidimensionality ensures that the overall score of the instrument is, in fact, describing what is actually happening, such that the overall effect is not diluted by those items that are insensitive to the construct of the instrument (Streiner and Norman, 1989). Fitting data to the Rasch model allows inferences to be made regarding desirable characteristics of the instrument (Tennant et al., 1996).

The two main claims of Rasch analysis are that: (i) the easier the item is, the more likely it will be passed (or affirmed) by a given individual; and (ii) the more able the respondent, the more likely they will pass (or affirm) an item (i.e., do a task) compared to a less able respondent. Rasch analysis deconstructs each item of the instrument into its component steps – for example in the HAQ, from zero to one, from one to two, and from two to three – and examines how successful people are in taking those steps. This gives an estimate of item difficulty, which is then used to assess the ability of the person. As unidimensionality is a pre-requisite for the summation of any set of items (Streiner and Norman, 1989), the Rasch model assumes that the probability of a given patient passing (affirming) an item or task is a logistic function of the relative distance between the item location parameter (i.e., the difficulty of the task) and the respondent location parameter (i.e., the ability of the patient) as shown in the following equation:

$$p_i(\theta)=\frac{e^{(\theta-b_i)}}{1+e^{(\theta-b_i)}}, \qquad (5.1)$$

where $p_i(\theta)$ is the probability that patients with ability θ will be able to complete the item (task) i, and b is the item (task) difficulty parameter.

The Rasch analysis then seeks to combine person ability and item difficulty by taking the difference between these two values ($\theta - b_i$). This difference governs the probability of what is supposed to happen when a person of given ability uses that ability against a given task (Tennant et al., 1996). The Rasch transformation, which is reported in logits, converts the discrete items onto a continuous scale based on the natural logarithm ($e = 2.718$). The relationship between person ability and item difficulty can be best understood by the fact that, for example, a person with a logit score of 2.0 (θ) will have an equal probability ($p_i(\theta) = 0.5$) of passing (affirming) or not passing an item, or a step on an item, with a difficulty level of 2.0 logits (b).

The overall goodness-of-fit test statistic determines how well the Rasch model fits the underlying data. This was examined in terms of item-trait interaction, the person separation index, and the person and item fit residuals.

Item-trait interaction measures whether the data fit the Rasch model for the given respondent group. These groups are selected by dividing the respondents into a series of sub-groups based on the position of the respondents on the latent scale of the model; thus, respondents who have similar QOL are grouped together. Observed and expected responses are compared across items and traits, with the difference between these responses summarized using the χ^2-test statistic. A good-fitting Rasch model should support the null hypothesis, such that there is no deviation between the observed and expected responses. Kubinger (2005) suggested that the p-value for the overall model should be greater than the critical value of 0.01.

Person separation index (PSI) measures the level of agreement between respondents. The PSI lies on a scale between zero and one, such that a higher value indicates a better agreement between respondents. A PSI $\geq$ 0.7 indicates a well-fitting Rasch model (Nunnally, 1978).

Fit residuals estimate the degree of divergence between the expected and observed responses for each respondent or item response. Fit residuals are summed over all items (*item fit residuals*) or over all persons (*person fit residuals*). The residuals are standardized to approximate the Z-score, such that the mean residual should be approximately zero with a SD approximately equal to one. For this study, the individual item fit values were used to provide further evidence as to whether or not items should be eliminated.

The computer program RUMM2010 (RUMM Laboratory, Duncraig, Australia) was used to fit the Rasch model.

Conducting Rasch Analysis

The use of Rasch analysis in the development of health state descriptions is relatively new; hence, there is a paucity of literature stating an exact formula to perform this type of analysis. The framework used to implement Rasch analysis, derived from Young et al. (2009), is described below.

Step I: Execute Rasch Analysis on the Full HAQ

The 20 items of the complete HAQ were subjected to Rasch analysis. The summary statistics for this analysis were examined.

Step II: Establish Whether the HAQ Domains Fit the Rasch Model

Prior to using Rasch analysis to determine which of the 20 HAQ items best represented disability, each domain was checked to determine whether it fitted the Rasch model. To do this, the items pertaining to each of the eight HAQ domains were individually fitted to a Rasch model and the resulting goodness-of-fit test statistics for each model were examined. This preliminary step is considered to be more crucial for instruments with domains consisting of numerous items. For this reason, less emphasis was placed on these results as there were only, at most, three items per HAQ domain. Due to the small number of items per domain for this instrument, reducing HAQ items based on this criterion would prove to be premature;

nevertheless, this step was still included in the current study to provide a complete picture of the analysis procedure.

Step III: Shortlist the HAQ Items

The threshold probability curves for each item of the HAQ were examined. These curves show the distribution of the item levels across latent space; examples are shown in Figure 5.1 (pages 113-114). Although the HAQ has four response levels (i.e., no difficulty, some difficulty, much difficulty in performing the task, and inability to perform the task), three curves are used to demonstrate the thresholds between item levels (e.g., from item level zero to item level one, from item level one to item level two, and from item level two and item level three). The horizontal and vertical axes of the threshold probability curve represent the underlying latent (logit) scale and the probability of being in a particular item level, respectively. Ideally, the item levels in the threshold probability curves should be appropriately ordered and spaced, and have an opportunity of occurring (i.e., not lying on the horizontal axis) as shown in, for example, Figure 5.1a (page 113).

However, if any of the item levels are disordered, such that a difficult level is more likely to be attained than an easier level, the disordered levels are merged together; an example of disordered levels is shown in Figure 5.1b (page 113). In this example, 'much difficulty' (i.e., a HAQ score of two) is less probable than 'unable to do' (i.e., a HAQ score of three); as a result, these two levels will be merged. As done previously (Young et al., 2009), formal guidelines were not used to merge item levels; instead, adjacent item levels were collapsed using an item-by-item approach to achieve order.

With each merged disordered item, a new base model resulted and the threshold probability curves for each item were re-examined with the new base model. This step was repeated until all the item levels were appropriately ordered as shown in Figure 5.1a, the ideal graph. Although the merged disordered items were included with each subsequent Rasch modelling run, these items will be excluded from consideration in the final health state classification systems because respondents could not discriminate between levels of these items.

If, after merging of the disordered item levels, any of the levels for all remaining items were poorly spread (i.e., item level curves were not of approximately equal distance spacing when inspected visually) or had a low chance of occurring (i.e., an item level curve lies close to the bottom horizontal axis), these item levels were further merged together; an example of poorly spread levels is shown on Figure 5.1c (page 114) and an example of a level lying close to 0% probability is shown on Figure 5.1d (page 114). This step required that the suspect level be merged with the adjacent level, *one item at a time*. As this step was conducted independently for each item, both the overall item-trait fit and the individual item fit test statistic needed to be re-examined to determine the best possible model that arose. If any of the individual items did not fit the model (i.e., had a significance level of $p < 0.01$ because there is a deviation between the observed and expected responses, as mentioned in Section 5.2.5) it was excluded from any subsequent modelling, as this item did not contribute to the underlying latent scale. The model with the smallest overall item-fit test statistic (i.e., largest p-value) was chosen to be the best model that resulted from the merging of levels. The model was re-fitted and the overall goodness-of-fit statistic was examined for the new model. The process was repeated until only well-fitting items remained (such as in Figure 5.1a) and the overall item-trait goodness-of-fit of the model was greater than $p = 0.01$.

Once the model fit criteria were satisfied, the items that were excluded from the construction of the health states were: (i) items that needed merging at the initial Rasch model-fitting stage (i.e., respondents were unable to distinguish between levels); and (ii) items that did not measure the underlying QOL trait of the HAQ domains (i.e., the items did not fit the Rasch model). This agrees with previously published techniques (e.g., Young et al., 2009).

Step IV: Differential Item Functioning

Differential item functioning (DIF) analyses were conducted as a further attempt to shortlist the HAQ items for inclusion in the final health state descriptions. DIF analysis is a procedure that examines whether any items in the instrument result in discrepant responses amongst different respondent characteristics. The item characteristic curves were examined to assess whether sex, age (e.g., less than 50 years of age, between 50 and 64, and greater than 65), and duration of RA (e.g.. less than 10 years, between 10 and 20 years, and greater than 20 years) influenced item responses between subgroups. The chosen variables were believed to potentially influence individual's responses on the HAQ items. The levels were selected to ensure approximately equal numbers of individuals in each group.

The potential for DIF was detected by item characteristic curves and item-by-item characteristic analysis of variance (ANOVA) statistics. If inspection of the item characteristic curves suggested that the likelihood of responses differed significantly between subgroups or between one of the subgroups and the mean response, it was deemed possible that this item did not fit the model well and should not be considered for inclusion in the final health states. Figure 5.2 (page 115) shows an example, where responses to the *openjars* by the male patients (represented by the circles) deviated from the mean response (represented by the solid line) to this HAQ item. The discrepancy observed between the levels of the item characteristic curves was verified by interpreting the F-test statistic, with the null hypothesis being no difference between subgroups. In general, those items for which it was necessary to adjust for systematic DIF across groups of respondents are considered to be of limited value for making cross-population comparison (Young et al., 2009). Any items which demonstrated DIF were not excluded at this point, but were considered as a suspect for removal in Step V.

Step V: Final Selection of HAQ Items

With the best-fitting model obtained from Step III, the remaining items – after shortlisting – were removed one at a time to assess which item's elimination resulted in the best-fitting model. This was done based on the position of the item on the location scale. The position on the location scale represents the severity of the item: a negative value indicates less severity (i.e., most individuals with of RA would not be able to complete the task) and a positive value indicates greater severity (i.e., only individuals with a more severe form of RA would not be able to complete the task). In addition to a model resulting in the strongest goodness-of-fit, the items that remained needed to describe both limitations in the upper and lower limbs, be from different domains, and capture a wide range of severity. This stage was repeated until the desired number of items remained; as mentioned above, for this study, the final number was pre-defined as four items.

5.2.6 Pain and Discomfort Domain

After the four most representative items of the HAQ were selected, an item pertaining to pain and discomfort needed to be included in the classification system. Within the EQ-5D, the levels pertaining to this dimension are defined as (i) no pain or discomfort; (ii) moderate pain or discomfort; and (iii) extreme pain or discomfort. (Refer to Appendix A.2, page 241, for a copy of the EQ-5D.) This information was included after the Rasch process was completed but before cluster analysis was conducted.

5.2.7 Forming Health States

Cluster Analysis

The next step was to conduct cluster analysis in order to form distinct RA states. *A priori* decision was made to define three states. This was to ensure that participants in the latter components of the sequential mixed-methods study – previously described in Section 4.2 – would have a range of states to value yet not overburden them when having to fulfill the other study requirements. These individuals not only need to value the health states but also undergo an information session about disease adaptation; as such, having more than three states to value would increase the time required, and the burden placed on the respondent, to complete the entire interview process. Specific details about the study design are described in Chapter Six.

As mentioned earlier, the RA health state descriptions are comprised of four HAQ items (each with four levels) and the pain and discomfort dimension of the EQ-5D (with three levels). This affords 768 possible health states ($4^4 * 3$). The use of cluster analysis aims to reduce these health states to a set of well-defined states that summarize RA patients' disability. In addition to being distinct, the final health states need to describe three states that commonly arise amongst RA patients.

In the past, the personal judgements of experts have been used to define and describe health states. However, the use of cluster analysis overcomes any bias associated with personal opinion by assessing the natural groupings of the data observed in a specific population (Sugar et al., 1998). Cluster analysis requires that patients in the same health state be as similar as possible and also that the health states accurately represent the clinical status of a patient population. In place of cluster analysis, factorial design can also be employed, as it simultaneously examines the effects of multiple independent variables and their degree of interaction. Cluster analysis was selected for use here because of its ability to let the data drive the description of the health states.

As the health state classification system was to be described by five items – four from the HAQ and one from the EQ-5D – the clusters were derived from the location of the five-dimensional data points. Typically, clusters are formed by dividing the range of data into a uniform grid, in such a way that each item will be evenly classified into several levels. However, this technique is considered inefficient and restrictive, as many of the health states defined in this manner may be empty because the particular combinations of item levels may not exist in the patient population. While there are several types of cluster analysis – for example, hierarchical, two-step, and expected maximization – the *k*-means algorithm was employed for this work; this is similar to previous successful applications of this technique (Sugar et al., 1998 and 2004; James et al., 2006). The main purpose of using *k*-means cluster analysis over other cluster analysis techniques is to exploit its ability to produce a unique, possibly asymmetrically spaced, set of discrete clusters that best describes the given dataset (Sugar et al., 1998). Furthermore, this approach provides the researcher with liberty in pre-selecting the number of clusters to suit the needs of the study requirements.

The *k*-means algorithm aims to group *n* observations into *k* partitions or clusters. This is achieved by finding the centres of natural clusters in the given dataset. The algorithm starts by randomly partitioning the data points into *k* initial sets. Then the mean point, or centre, is calculated in each set. The algorithm then constructs a new partition by associating each point with the closest centre. The centres are re-calculated for the new clusters and the algorithm is repeated until convergence is achieved; that is, the data points no longer switch clusters. This approach seeks to identify a set of groups, which both minimizes within-cluster variation and maximizes between-cluster variation in a fashion similar to that of ANOVA.

While three health states (clusters) are needed for the latter two components of the mixed-methods study, the stability of the clusters was examined by running the *k*-means algorithm using three, four, and five clusters. By running cluster analyses on a range of cluster numbers, the optimal spread of the data can be determined by assessing whether the combination of item levels changes with the increasing number of clusters.

K-means cluster analysis was conducted using SPSS version 14.0.

5.2.8 Additional Analysis

With the final three health states, one-way ANOVAs were conducted. This statistical test evaluated whether the differences existed amongst the respondents' age, RA duration, and instrument scores when stratified by cluster membership.

5.3 Results

5.3.1 Characteristics of the Study Population

Table 5.2 (page 101) displays information regarding the demographic variables and the QOL of the NDB participants. The mean (SD) number of years these individuals had been living with diagnosed RA was 16.7 (11.7). In terms of EQ-5D, the mean (SD) score, derived from converting the individual responses on the classification system using the US societal tariff (Shaw et al., 2005), is 0.67 (0.22). The unstandardized mean (SD) score from the EQ-5D VAS is 63.16 (21.04). As expected, a gradient is observed across severity levels; people with severe RA reported poorer QOL scores than individuals with mild and moderate RA (Table 5.3, page 101), independent of which measure was used.

5.3.2 Frequency of the HAQ Responses

The results, shown in Table 5.4 (page 102), demonstrate a gradient between the frequency of responses and the difficulty of the items: patients were more likely to report 'no difficulty' than 'some difficulty' and so forth for the items. The most difficult level, 'unable to do', was relatively infrequent amongst most items; however, the items *overhead, vacuum*, and *tubbath* produced a greater frequency of these responses when compared to the adjacent level of 'much difficulty'. Interestingly, in the case of *tubbath* the 'unable to do' level was the most frequently selected at 40%.

5.3.3 Internal Consistency of the HAQ Responses

The correlation between the item and domain scores was also examined (Table 5.5, page 103). The majority of items were internally consistent, such that rho ≥ 0.7; however *fauceton* (rho = 0.68), *washbody* (rho = 0.59), and *ontoilet* (rho = 0.59) did not fit this criterion. This provides evidence that these items may not be representative of the domain – especially since two of the three items of the hygiene domain (e.g., *washbody* and *ontoilet*) demonstrated a weak correlation – and thus may prove to be inappropriate for inclusion in the final health state descriptions.

5.3.4 Rasch Analysis

Step I: Execute Rasch Analysis on Full HAQ

All items of the HAQ were modelled using Rasch analysis. For this model (M1), the overall goodness-of-fit test statistic was determined to be $\chi^2 = 310.5$ ($p \leq 0.001$) for 180 degrees of freedom. The information obtained from this step was used to provide a reference for the subsequent steps of the Rasch modelling process.

Step II: Establish Whether the HAQ Domains Fit the Rasch Model

The next step was to assess whether each of the eight HAQ domains achieved overall Rasch model goodness-of-fit (i.e., individual items should be $p > 0.01$ to satisfy the null hypothesis, as described previously in Section 5.2.5). Table 5.6 (page 104) summarizes the overall Rasch model statistics – item-trait, PSI, item fit, and person fit – for each of the eight HAQ domains. Only the hygiene satisfied the goodness-of-fit criteria since it was the only domain that had an item-trait p-value greater than 0.01 ($\chi^2 = 25.70$, $p = 0.32$). The PSIs for the domains were above the range for a well-fitting Rasch model (i.e., PSI ≥ 0.7). The values for the item fit and person fit residuals showed that the only item fit values approximately had a Z-distribution. However, since Rasch analysis requires at least two items per domain, and each of the HAQ domains had at most three items, it was decided to keep all items in the model at this point in the analysis.

Step III: Shortlist HAQ Items

The threshold probability curves for the full HAQ were examined. The results indicated that all the item levels were ordered appropriately except for the most severe levels of *tubbath* and *shampoo*; the misfitting nature of these items (e.g., Figure 5.1b, page 113) has been reported elsewhere (Wolfe et al., 2004). As a result, the two most severe levels of these items (i.e., much difficulty and unable to perform the task) were merged to form a new base model. This new model (M2) is identical to the original HAQ, except that *tubbath* and *shampoo* now contain three levels – no difficulty, some difficulty, and much difficulty with the task – instead of four.

For the new base model (M2), the overall goodness-of-fit and individual item-fit test statistics were assessed. The individual item-fit test indicated that the *runerand* item did not fit the Rasch model ($p \leq 0.009$) because it was less than $p \leq 0.01$. Thus, this item was removed from subsequent Rasch modelling. The new base model (M3) is similar to the original HAQ, except that both *tubbath* and *shampoo* contain a total of three levels and that *runerand* is not included.

Once again a Rasch model was conducted on the current base model (M3) and, although 19 of the 20 HAQ items were included in the modelling (i.e., *runerand* was removed), the threshold probability curves for all items containing four levels were re-evaluated. The merged levels of *tubbath* and *shampoo* indicated that respondents were unable to distinguish between the levels. From these curves, the two most severe levels of *overhead* and *vacuum* were closely distributed together (e.g., Figure

5.1c, page 114) and the 'unable to do' level of *fauceton* and *inbed* lay close to the 0% probability line as illustrated by Figure 5.1d (page 114). For each of the aforementioned items, the two most severe levels were merged one item at a time and the overall goodness-of-fit and individual item fit were examined for each merged item. The merging of the two most severe levels of *vacuum* resulted in the model with the lowest χ^2 value and the highest p-value, and hence a better-fitting model (χ^2 = 197.2, p = 0.08), compared to merging of the other items. As a result, *vacuum* was modified by merging the two most severe levels, as described above, and a new base model (M4) was developed.

With the base model (M4) – identical to that of the original HAQ except that *tubbath*, *shampoo*, and *vacuum* contain a total of three levels and *runerand* is excluded – the two most severe levels (e.g., much difficulty and unable to do) of the poorly fitting items mentioned above (e.g., *overhead*, *fauceton*, and *inbed*) were merged once again. The merging of the two most severe levels of *fauceton* resulted in the better model (i.e., a lowest χ^2-test statistic and a highest p-value) (M5) (χ^2 = 197.3, p = 0.08) when compared to models with merged *overhead* and merged *inbed*. Once again, the Rasch models were re-analyzed with the inclusion of the merged *fauceton* levels as the new base model (M5), along with merged *tubbath*, *shampoo*, and *vacuum* levels, and excluding *runerand*. The merging of the two most severe levels of *inbed* resulted in the best model (M6) (χ^2 = 202.0, p = 0.05). Finally, with the new base model (M6), which includes the merged *tubbath*, *shampoo*, *vacuum*, *fauceton*, and *inbed*, and with the removal of *runerand*, a better model (M7) arose when *overhead* was merged (χ^2 = 206.7, p = 0.03).

The new base model (M7) differed from the original HAQ in that *tubbath*, *shampoo*, *vacuum*, *fauceton*, *inbed*, and *overhead* now contained three levels each, and *runerand* was now removed. The threshold probability curves for the new base model indicated that the two most severe levels were not well distributed (i.e., the levels that lay close together; Figure 5.1c, page 114) for the following items: *washbody*, *dresself*, *openmilk*, and *ontoilet*. The two most severe levels of these items were merged one item at a time, as previously done, and the summary and individual item test statistics were examined to determine which of the merged items produced the best overall model. The subsequent Rasch models revealed that four levels remained for nine items: *benddown*, *climstep*, *liftcup*, *standup*, *walkflat*, *openjars*, *opencar*, *inoutcar*, and *cutmeat* after the merging of *openmilk* (χ^2 =198.9, p = 0.07), *ontoilet* (χ^2 = 196.2, p = 0.09), *washbody* (χ^2 = 202.9, p = 0.05), and *dresself* ($\chi^2 = 208.7$, p = 0.03). With this new base model (M8) (Table 5.7, page 105), none of the items showed disordered levels (Figure 5.1b, page 112), poorly spread levels (Figure 5.1c, page 114), or levels lying close to the 0% probability axis (Figure 5.1d, page 114). The remaining HAQ items with four levels were appropriately ordered (Figure 5.1a, page 113).

Step IV: Differential Item Functioning

Three types of DIF analyses were conducted to determine whether or not the HAQ items resulted in differential responses between patient subgroups. For items still under consideration for the final health states (i.e., items still containing four levels), both the item characteristic curves and the F-test statistics were inspected to identify which items did not fit the Rasch model well. The item characteristic curve indicated that the item *openjars* could potentially be removed from the final health states because the individual curves deviated from the mean item characteristic curve (see Figure 5.2, page 115, for an example of this based on sex differentiation). The difference in male and female responses for *openjars* was also verified by a significant F-test statistic (p = 0.03). Thus, *openjars* was determined to be eligible for removal but this was verified in Step V. None of the other items appeared to indicate DIF.

Step V: Final Selection of HAQ Items

As there were still nine items to be considered for the final health states (M8) (Table 5.7, page 105), each item was removed one at a time, based on the position of the item on the location scale, until the best model with four items was produced. The location scale indicated that, for example, most individuals with RA would have problems climbing up steps (i.e., represented by a negative location value); however, only the most severe cases in this group would be unable to lift a cup to their mouth (i.e., represented by a positive location value). From the difficulty (logit) scale in Table 5.7 (i.e., items are placed from greatest to least difficulty), of the items still under consideration (indicated in bolded font) only *climstep* had a negative location value, representing the mildest form of severity. Thus, *climstep* was retained as it ensured that the widest range of severity would be achieved for the final model.

Using this sequential removal-reassessment process, where one item was removed at a time and individual item test statistics of each model were examined to determine the best model that arose, the following items were removed from the current base model (M8): *opencar* ($\chi^2 = 196.7$, p = 0.03), *openjars* ($\chi^2 = 182.0$, p = 0.06), and *cutmeat* (χ^2 = 180.8, p = 0.02) (results not tabulated). As a result, the new base model (M9) items contained *benddown*, *climstep*, *liftcup*, *standup*, *walkflat*, and *inoutcar* (Table 5.8, page 106). As one of the item inclusion criteria is to have items from separate domains to avoid collinearity, *walkflat* was removed from the final model (M10) (χ^2 = 179.3, p = 0.006) (Table 5.9, page 107). The rationale for excluding *walkflat* was that retaining *climstep* in the model would keep an item that represents the mildest form of severity; as discussed earlier, the inability to climb stairs is most often experienced by RA patients.

The five items that were still under consideration for the final health states were *benddown*, *climstep*, *liftcup*, *standup*, and *inoutcar*. Their individual item fit statistics are shown in Table 5.9 (page 107). It was desirable to retain *climstep* as it represented the mildest form of severity; therefore, one of either *benddown*, *liftcup*, *standup* or *inoutcar* had to be eliminated from the set of five items. As previously described in Section 5.2.4, it was thought that the *inoutcar* item might potentially result in discrepancies between patients' responses, because of the assistance provided by travelling companions and variations in car size between study populations (particularly between the UK and the US, where the latter tend to have larger, higher cars which are easier to enter and exit). Accordingly, the item *inoutcar* was removed from the final health state description. The final reduced HAQ model is, therefore, composed of: *benddown*, *climstep*, *liftcup*, and *standup*. The final four items were modelled (M11) using Rasch analysis without the items not considered for the final health state classification system (χ^2 = 44.4, p = 0.03) (Table 5.10, page 107). A summary of items eliminated by Rasch analysis is presented in Table 5.11 (page 108).

5.3.5 Cluster Analysis

Using the raw scores from the four selected HAQ items and the pain and discomfort dimension of the EQ-5D, the *k*-means algorithm was conducted using three, four, and five clusters (Table 5.12a, page 109). A five-digit code number relating to the relevant level of each item represented was established to code each composite health state. Thus, cluster one for the three-, four-, and five-cluster models was denoted as '00001', which represents no difficulty standing up from a straight and armless chair, no difficulty lifting a cup to one's mouth, no difficulty climbing up five steps, no difficulty bending down to pick up clothes from the floor, and mild pain and discomfort. As the number of clusters increased, the combinations of item levels either remained the same or only moved one unit of disability (e.g., from one to two).

As mentioned earlier, three clusters to represent three different RA states were needed for valuations by members of the general population in the subsequent empirical studies. However, the identified three-cluster model did not suit the purpose of this thesis. As four items out of the five represented no disability, the first cluster of the three-cluster model described very limited disability. As the purpose of constructing these health states was to evaluate the impact of disease adaptation information on general population values, there was need for these descriptions to be distinct, comprehensible, and demonstrate a level of disability. If the first cluster of the three-cluster model was generally considered to be synonymous to full health, this would result in only two impaired health states for respondents to value. As a result, cluster centres two, three, and four of the four-cluster model were used to describe three states that all included a level of disability. When further analyzing the item levels comprising the three clusters of the four-cluster model, there was a concern that respondents might have a difficult time differentiating the levels between the cluster centre two and three. Thus, it was decided *post hoc* to repeat the *k*-means algorithm with only the HAQ items and to incorporate the pain and discomfort levels from the EQ-5D based on the frequencies of responses to that dimension for the chosen cluster.

As happened in the *k*-means clustering using both HAQ items and the EQ-5D domain, the first cluster of the HAQ-only three-, four-, and five-cluster models generally showed no disability (Table 5.12b, page 110). Therefore, the three-cluster model was insufficient to describe the three health states required and the four-cluster model was used instead. Table 5.13 (page 110) shows the frequency of responses to the EQ-5D pain and discomfort dimension (no, moderate, extreme) for the cluster centres one, two, three, and four of the four-cluster model (e.g., very mild RA, mild RA, moderate RA, severe RA). Amongst all severity groups, the most frequent response was moderate pain and discomfort; this implies that the pain and discomfort domain for the three health states should be labelled as 'moderate'. However, to ensure that the general population respondents could differentiate between the states when asked to value them, pain and discomfort was labelled as mild, moderate, and extreme to describe the three health states (Table 5.14, page 111).

5.3.6 Respondent Characteristics of the Final Health States

Cluster analysis automatically assigns the study sample into one of the four identified clusters based on their responses on the HAQ. Table 5.15 (page 112) displays information regarding demographic variables and QOL of the study participants in the three of the four defined clusters. Significant numbers of individuals in the mild (n = 163), moderate (n = 108), and severe (n = 50) cluster groups existed. Although not characterized in the table, the dataset consisted of 248 patients in the first cluster of the four-cluster model (i.e., very mild state); as mentioned above, this cluster will not be used in the health state descriptions. (These numbers do not add up to 600 because 31 respondents did not provide a response for at least one of the HAQ items and therefore were excluded from the cluster analysis.) While this may seem like a limitation to exclude these patients for having a very mild form of RA, the primary purpose of conducting cluster analysis on the Rasch-reduced HAQ is not to represent comprehensively all cases of the given dataset but rather to find three health states that represent RA patients to be used in future valuation studies.

The ANOVA results revealed that there were no differences in the patients' age and RA duration across cluster groups (p = 0.54 and 0.07, respectively). However, in terms of HAQ scores, EQ-5D indices, and EQ-VAS values, the QOL measures differentiated well across the severity groups ($p \leq 0.001$). A monotonic gradient was observed, such that higher HAQ scores, lower EQ-5D indices, and lower EQ-VAS values are associated with more severe forms of RA. These results provided evidence that the health states constructed by cluster analysis had the ability to discriminate between the different levels of RA severity.

The mild, moderate, and severe health states were labelled as States X, Y, and Z, respectively, when used in latter components of the mixed-methods study. This was to avoid any labelling effects that might arise by connotation if the descriptive adjectives (i.e., mild, moderate, and severe) were used.

5.4 Discussion

The results demonstrate that the combined use of Rasch and cluster analyses, alongside psychometric techniques, can create distinct and plausible descriptions in RA. This approach allows researchers to develop health states that are based on patient-level data but meet pre-specified needs of their study design. For example, to reduce respondent burden in the completion of complex valuation tasks, x number of states composed of n items can be constructed.

This is believed to be the first study to combine Rasch and cluster analyses in the development of health states. Rasch analysis demonstrates clear advantages for this type of exercise over the potential use of factor analysis, which is more applicable for the multi-dimensional instruments rather than unidimensional instruments like the HAQ. It is also superior to qualitative approaches, which obtain information from a small number of individuals participating in interviews or focus groups, as it uses a much larger and more diverse sample set. Furthermore, a floor effect has been reported with the HAQ (Wolfe et al., 2004), such that a severely disabled individual can record a decent HAQ score. By fitting the HAQ items into a Rasch model, the floor effect can be reduced; this was achieved by selecting the widest possible range on the logit scale. Despite these advantages, subjectivity was introduced when ensuring that the final descriptions of the health states were sufficient for the purposes of the future empirical studies (i.e., comprehensible to respondents who might not be well informed about RA, and containing a combination of activities that address both upper- and lower-limb disability).

Rasch analysis is regarded as a tool that can be used to aid in the development of health states. The statistical techniques used in assessing the goodness-of-fit of the models provided guidance, but did not supersede judgements regarding the clinical nature of the condition. The items included in the RA descriptions need to describe clearly the full range of disability associated with this chronic condition. While members of the general population may tend to focus on the classic symptoms of RA, such as mobility (e.g., climbing steps), the purpose of these descriptions of the health states was to create a descriptive and well-rounded picture of individuals living with RA across three distinct states through a limited number of items. Thus, it was essential to include in the descriptions both everyday tasks (e.g., bending down, lifting a full cup, standing up from a straight and armless chair), and pain and discomfort.

This is not the first study to conduct Rasch analysis on the HAQ. To date, two other studies have addressed the use of Rasch analysis on the HAQ, although their overall objectives were different from this present study. Tennant et al. (1996) investigated the scaling of the HAQ and the fit of the data to the Rasch model. Similar to the findings of the present study, the item *liftcup* adequately represented the upper level of disability: those who have difficulty with this task, or find it impossible, have the severest form of the condition. Wolfe et al. (2004) also applied Rasch analysis to the HAQ to reduce the instrument down to ten items; however, they opted to use a revised version, the HAQ-II, rather than the original instrument so their results are not directly comparable to the ones described in this chapter. The HAQ-II contained new items: waiting in a line for 15 minutes, doing outside work, lifting heavy objects, and moving heavy objects. After Rasch analysis, the remaining six items of the HAQ-II were *ontoilet*, *opencar*, *walkflat*, *overhead*, *standup*, and *climstep*; of which, only *standup* and *climstep* remain in the reduced HAQ described in this chapter. For the four discrepant items between the two studies, all items except *walkflat* were excluded based on the poorly spread item level criterion; *walkflat* was eliminated because of potential collinearity with *climstep* when the latter remained in the RA health state description.

The HAQ items comprise four out of five items in the developed health states. While the HAQ measures pain and discomfort, it was decided to not use the patients' responses pertaining to this domain. The continuous representation of pain on the HAQ may be more meaningful than categorical pain levels (e.g., no pain, moderate pain, and extreme pain) to physicians or rheumatologists monitoring RA patients over multiple time points. However, these ratings may carry less weight when appraised by individuals who may not be informed about RA itself. As pain is a symptom experienced by most patients living with RA (National Rheumatoid Arthritis Society, 2008), excluding this domain was considered detrimental to providing an appropriate description of the impact of RA on individuals' health. Therefore, to ensure that the health states were comprehensible and, more importantly, realistic, the decision was made to include the pain and discomfort domain, but rather than using the HAQ version thereof, to replace it by the dimension from the EQ-5D data collected from the same patients at the same time as the HAQ data.

In the EQ-5D, the pain and discomfort dimension is described by three levels. However, for this thesis, it was felt that the definition of these levels did not provide enough sensitivity to describe the three health states needed for subsequent empirical studies. As such, another level was created to represent "you have mild pain and discomfort"; this was used for the RA state that is mild (i.e., State X).

As with any study, this one has limitations. There is a potential for misclassification as the HAQ score was used as a proxy to determine the RA severity of the patients. While this may seem like an adequate approach, it has been reported that this instrument could elicit a floor effect (Wolfe et al., 2004), with the result that the severe patients can be represented by a HAQ score that underestimates the true severity of the condition has on their QOL (i.e., a lower HAQ score). However, across the different measures of QOL, which included EQ-5D index and EQ-5D VAS, the relationship between RA severity and the QOL measures at the aggregate levels is in the anticipated direction, suggesting that any misclassification of severity by HAQ score is likely minimal. In addition, the nature of how the information from the NDB was collected may be subject to self-reporting bias. It is possible that individuals may, either consciously or subconsciously, exaggerate the individual item score of the HAQ to please their physician and family members for fear that it may alter the current treatment regimen; the result would yield a better QOL (i.e., a lower HAQ score) than they should actually be.

This study used the data from 600 patients recruited through the NDB in Wichita, Kansas: the results presented in this chapter may not be generalizable. Although the sample encompassed all levels of disease severity, it may not be representative of the general RA population in the US, let alone in the UK, where the subsequent empirical studies will take place. The dataset itself contained responses from a third of patients with mild RA (i.e., HAQ score < 1), a third with moderate RA (i.e., HAQ score 1-2), and a third with severe RA (i.e., HAQ score > 2); thus, patients with severe RA were over-sampled. In reality, this is not representative of RA severity. This over-sampling ensured that distinct health states were constructed for respondents from the general population to value in the remaining mixed-methods components of this research project (described in Chapters Seven and Eight).

Furthermore, when the proportion of patients actually living in three defined health states were identified, 248 patients actually belonged in a very mild RA state. Because this very mild RA state was considered to synonymous to full health (i.e., no detriment in most of the item levels in the health state description), the decision was made to not have a health state that described a very mild form of RA. Excluding a very mild RA state may not portray a realistic picture of RA severity to the general population respondents. However, in terms of understanding the effect of disease adaptation information on their health state values (i.e., the primary objective of the overall thesis project), it was important to have states with some level of detriment present in the health state.

From the NDB website (National Data Bank for Rheumatic Diseases, n.d.), respondents are not offered direct incentives for participation but instead are offered a chance to win one of three $1,000 (US dollar) lotteries if the research questionnaire is returned within two weeks. This financial incentive may provide varying degrees of participation amongst patients in different social classes. However, as the dataset did not include information on socio-economic status variables (e.g., annual income, highest attained education levels), it was not possible to ascertain whether socio-economic status of the respondents varied significantly from that of the general RA population. This would be a concern only if either the distribution of HAQ scores, or the distribution of different clinical manifestations of RA, was related to social class. There is no evidence to suggest either of these events are probable.

5.5 Summary and Conclusions

Despite the potential limitations discussed above, the objective of the first component of the sequential mixed-methods study was achieved: three distinct yet credible RA states were defined. In addition to the description being representative of three different health states in RA, the constructed descriptions are believed to be meaningful. The results presented in this chapter demonstrate a methodology for reducing a dataset containing the individual HAQ scores to generate a framework for the RA health states. Overall, the combined use of Rasch and cluster analysis, with the results being assessed subjectively based on expert judgement, has proved to be an effective technique for identifying the most important items and levels for the construction of health states to be used in future valuation studies.

5.6 Tables and Figures

Table 5.1: Items in the HAQ[a]

HAQ Item	Domain	Description of Item
Dresself	Dressing & Grooming	Dress yourself, including tying shoelaces and doing buttons
Shampoo	Dressing & Grooming	Shampoo your hair
Standup	Arising	Stand up from a straight chair
Inbed	Arising	Get in and out of bed
Cutmeat	Eating	Cut your meat
Liftcup	Eating	Lift a full cup or glass to your mouth
Openmilk	Eating	Open a new milk carton
Walkflat	Walking	Walk outdoors on flat ground
Climstep	Walking	Climb up five steps
Washbody	Hygiene	Wash and dry your body
Tubbath	Hygiene	Take a tub bath
Ontoilet	Hygiene	Get on and off the toilet
Overhead	Reach	Reach and get down a 5-pound object (such as a bag of sugar)
Benddown	Reach	Bend down to pick up clothing from the floor
Opencar	Grip	Open car doors
Openjars	Grip	Open jars which have been previously opened
Fauceton	Grip	Turn faucets on and off
Runerand	Activities	Run errands and shop
Inoutcar	Activities	Get in and out of a car
Vacuum	Activities	Do chores such as vacuuming or yard work

[a] HAQ: Stanford Health Assessment Questionnaire.

Table 5.2: Characteristics of the study sample

Variables[a]	n	Mean	Standard Deviation	Minimum	Maximum
Age	600	61.6	12.8	16.8	89.9
Duration of RA (years)	600	16.7	11.7	0.7	75.8
HAQ	600	1.41	0.87	0	3.00
EQ-5D Index	600	0.67	0.22	-0.04	1.00
EQ-5D VAS	600	63.16	21.04	0	99
Female	474 (79%)				

[a] EQ-5D: EuroQol-5D; EQ-5D VAS: EuroQol-5D visual analogue scale; HAQ: Stanford Health Assessment Questionnaire.

Table 5.3: Characteristics of each severity group (n = 200 for each group)[a]

HAQ Score	Duration in Years (SD)	HAQ Score (SD)	EQ-5D Index (SD)	EQ-5D VAS (SD)
< 1.00	15.1 (10.7)	0.36 (0.32)	0.85 (0.11)	76.28 (17.51)
1.00 to 2.00	16.3 (11.6)	1.51 (0.33)	0.68 (0.17)	60.01 (18.79)
> 2.00	18.7 (12.6)	2.35 (0.22)	0.50 (0.21)	53.20 (19.74)

[a] EQ-5D: EuroQol 5D; EQ-5D VAS: EuroQol-5D visual analogue scale; HAQ: Stanford Health Assessment Questionnaire; SD: standard deviation.

Table 5.4: Frequency of responses for each item level of the HAQ[a]

HAQ Item	Number of Responses (%)				
	No Difficulty	Some Difficulty	Much Difficulty	Unable to Do	Missing
Dresself	261 (43.5)	242 (40.3)	68 (11.3)	25 (4.2)	4 (0.7)
Shampoo	329 (54.8)	155 (25.8)	49 (8.2)	46 (7.7)	21 (3.5)
Standup	246 (41.0)	243 (40.5)	91 (15.2)	14 (2.3)	6 (1.0)
Inbed	277 (46.2)	239 (39.8)	63 (10.5)	5 (0.8)	16 (2.7)
Cutmeat	325 (54.2)	169 (28.2)	80 (13.3)	20 (3.3)	6 (1.0)
Liftcup	363 (60.5)	159 (26.5)	59 (9.8)	7 (1.2)	12 (2.0)
Openmilk	224 (37.3)	189 (31.5)	108 (18.0)	63 (10.5)	16 (2.7)
Walkflat	284 (47.3)	211 (35.2)	75 (12.5)	26 (4.3)	4 (0.7)
Climstep	226 (37.7)	196 (32.7)	118 (19.7)	51 (8.5)	9 (1.5)
Washbody	349 (58.2)	196 (32.7)	43 (7.2)	8 (1.3)	4 (0.7)
Tubbath	178 (29.7)	113 (18.8)	58 (9.7)	238 (39.7)	13 (2.2)
Ontoilet	345 (57.5)	215 (35.8)	33 (5.5)	5 (0.8)	2 (0.3)
Overhead	209 (34.8)	177 (29.5)	93 (15.5)	118 (19.7)	3 (0.5)
Benddown	272 (45.3)	212 (35.3)	79 (13.2)	28 (4.7)	9 (1.5)
Opencar	340 (56.7)	185 (30.8)	57 (9.5)	15 (2.5)	3 (0.5)
Openjars	235 (39.2)	252 (42.0)	86 (14.3)	24 (4.0)	3 (0.5)
Fauceton	368 (61.3)	177 (29.5)	46 (7.7)	2 (0.3)	7 (1.2)
Runerand	241 (40.2)	209 (34.8)	96 (16.0)	50 (8.3)	4 (0.7)
Inoutcar	242 (40.3)	269 (44.8)	76 (12.7)	8 (1.3)	5 (0.8)
Vacuum	138 (23.0)	184 (30.7)	128 (21.3)	146 (24.3)	4 (0.7)

[a] HAQ: Stanford Health Assessment Questionnaire.

Table 5.5: Correlations between the HAQ item and domain scores[a]

HAQ Item	Domain	Spearman's rho
Dresself	Dressing and Grooming	0.911
Shampoo	Dressing and Grooming	0.825
Standup	Arising	0.947
Inbed	Arising	0.839
Cutmeat	Eating	0.813
Liftcup	Eating	0.760
Openmilk	Eating	0.967
Walkflat	Walking	0.841
Climstep	Walking	0.968
Washbody	Hygiene	0.585
Tubbath	Hygiene	0.990
Ontoilet	Hygiene	0.589
Overhead	Reach	0.956
Benddown	Reach	0.757
Opencar	Grip	0.761
Openjars	Grip	0.915
Fauceton	Grip	0.684
Runerand	Activities	0.789
Inoutcar	Activities	0.972
Vacuum	Activities	0.972

[a] HAQ: Stanford Health Assessment Questionnaire.

Table 5.6: Summary of the Rasch goodness-of-fit statistics for the eight HAQ domains[a]

Domains	Overall Item-Trait χ^2	DF	Item-Trait p-Value	Mean Item Fit (SD)	Mean Person Fit (SD)	PSI
Dressing and Grooming	46.61	7	0	0 (0.41)	-1.75 (2.34)	0.82
Arising	30.17	6	0.00004	0 (0.55)	-2.00 (2.73)	0.80
Eating	59.85	18	0.000002	0 (1.51)	-3.06 (3.22)	0.91
Walking	44.04	9	0.000001	0 (1.04)	-2.36 (3.29)	0.88
Hygiene	25.70	23	0.32	0 (2.50)	-1.76 (2.38)	0.84
Reach	31.27	7	0.00055	0 (1.93)	-0.94 (2.45)	0.82
Grip	62.82	17	0	0 (0.50)	-2.57 (2.57)	0.87
Activities	43.58	7	0	0 (2.78)	-0.70 (2.77)	0.76

[a] DF: degrees of freedom; HAQ: Stanford Health Assessment Questionnaire; PSI: person separation index; SD: standard deviation.

Table 5.7: Individual HAQ item fit for the items still under consideration (shown in bold) (Model M8)[a]

HAQ Item[b]	Location	SE	Fit Residual	DF	χ^{2c}	p-value
Liftcup	**1.62**	**0.09**	**-0.91**	**485.1**	**12.8**	**0.17**
Opencar	**1.26**	**0.08**	**-1.66**	**493.6**	**7.4**	**0.60**
Inoutcar	**1.04**	**0.09**	**-2.35**	**491.7**	**10.5**	**0.31**
Cutmeat	**0.89**	**0.08**	**-1.06**	**490.7**	**5.51**	**0.79**
Ontoilet	0.77	0.10	-0.37	494.5	13.1	0.16
Fauceton	0.73	0.10	0.84	489.8	5.53	0.79
Standup	**0.66**	**0.08**	**-0.24**	**490.7**	**7.79**	**0.56**
Washbody	0.65	0.10	-3.40	492.6	18.8	0.03
Walkflat	**0.58**	**0.08**	**0.31**	**492.6**	**11.3**	**0.26**
Benddown	**0.50**	**0.08**	**1.30**	**487.9**	**5.20**	**0.82**
Openjars	**0.41**	**0.08**	**1.61**	**493.6**	**18.96**	**0.03**
Shampoo	0.02	0.09	-1.50	476.6	7.47	0.59
Inbed	-0.10	0.09	-0.29	481.3	5.63	0.78
Climstep	**-0.24**	**0.07**	**0.53**	**487.9**	**17.7**	**0.04**
Dresself	-0.49	0.09	-3.17	492.6	16.2	0.06
Openmilk	-1.37	0.09	0.62	481.3	14.0	0.12
Overhead	-1.76	0.09	-2.17	493.6	9.2	0.42
Tubbath	-2.41	0.09	1.41	485.1	17.2	0.05
Vacuum	-2.77	0.09	-0.53	492.6	4.65	0.86

[a] DF: degrees of freedom; HAQ: Stanford Health Assessment Questionnaire; SE: standard error.

[b] Runerand excluded from this analysis

[c] DF for χ^2-test: 9.

Table 5.8: HAQ item fit for the items still under consideration (shown in bold) (Model M9)[a]

HAQ Item[b]	Location	SE	Fit Residual	DF	χ^{2c}	p-value
Liftcup	**1.74**	**0.09**	**-0.03**	**477.2**	**14.1**	**0.12**
Inoutcar	**1.23**	**0.09**	**-2.72**	**483.7**	**17.7**	**0.04**
Ontoilet	0.94	0.10	-0.62	486.5	13.8	0.13
Fauceton	0.88	0.10	1.88	481.8	9.7	0.38
Standup	**0.82**	**0.08**	**-0.88**	**482.8**	**5.0**	**0.84**
Washbody	0.82	0.10	-3.32	484.6	20.4	0.02
Walkflat	**0.75**	**0.08**	**-0.09**	**484.6**	**11.7**	**0.23**
Benddown	**0.66**	**0.08**	**0.90**	**480.0**	**4.7**	**0.86**
Shampoo	0.18	0.09	-1.58	468.8	9.22	0.42
Inbed	0.07	0.09	-0.51	473.5	4.25	0.89
Climstep	**-0.08**	**0.08**	**-0.59**	**480.0**	**10.6**	**0.30**
Dresself	-0.32	0.09	-2.90	484.6	17.9	0.04
Openmilk	-1.19	0.09	1.73	473.5	10.6	0.30
Overhead	-1.59	0.09	-1.32	485.6	10.5	0.31
Tubbath	-2.26	0.09	0.88	477.2	12.3	0.20
Vacuum	-2.64	0.09	-0.80	484.6	8.18	0.52

[a] DF: degrees of freedom; HAQ: Stanford Health Assessment Questionnaire; SE: standard error.

[b] Runerand, Openjars, Opencar, and Cutmeat excluded from this analysis; bolded items represent items still under consideration for inclusion in final health states

[c] DF for χ^2-test: 9.

Table 5.9: Individual item fit for the reduced HAQ (Model M10)[a]

HAQ Item	Location	SE	Fit Residual	DF	χ^{2b}	p-value
Liftcup	0.80	0.09	2.99	337.2	23.6	0.003
Inoutcar	0.33	0.09	-2.12	337.2	26.9	0.03
Standup	-0.05	0.09	-0.94	337.2	4.13	0.85
Benddown	-0.20	0.08	0.38	337.2	14.5	0.07
Climstep	-0.90	0.08	-0.93	337.2	4.00	0.86

[a] DF: degrees of freedom; HAQ: Stanford Health Assessment Questionnaire; SE: standard error.
[b] DF for χ^2-test: 8.

Table 5.10: Individual item fit for the HAQ items to be used in the health state descriptions (Model M11)[a]

HAQ Item	Location	SE	Fit Residual	DF	χ^{2b}	p-value
Liftcup	0.87	0.08	2.46	316.2	18.4	0.01
Standup	0.04	0.08	-0.77	321.3	6.5	0.48
Benddown	-0.11	0.08	0.11	319.1	11.3	0.12
Climstep	-0.80	0.07	-1.19	318.4	8.12	0.32

[a] DF: degrees of freedom; HAQ: Stanford Health Assessment Questionnaire; SE: standard error.
[b] DF for χ^2-test: 7.

Table 5.11: Summary of the Rasch-eliminated HAQ items[a]

HAQ Item	Domain	Item Level Collapsed	DIF – Characteristic and Split	Failed to meet Rasch Goodness-of-Fit Criteria
Dresself	Dressing & grooming	With much difficulty and unable to do		
Shampoo	Dressing & grooming	With much difficulty and unable to do		
Inbed	Arising	With much difficulty and unable to do		
Cutmeat	Eating			Failed
Openmilk	Eating	With much difficulty and unable to do		
Walkflat	Walking			Failed
Washbody	Hygiene	With much difficulty and unable to do		
Tubbath	Hygiene	With much difficulty and unable to do		
Ontoilet	Hygiene	With much difficulty and unable to do		
Overhead	Reach	With much difficulty and unable to do		
Opencar	Grip			Failed
Openjars	Grip		Male vs. female	Failed
Fauceton	Grip	With much difficulty and unable to do		
Runerand	Activities			Failed
Inoutcar	Activities			Failed
Vacuum	Activities	With much difficulty and unable to do		

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[a] DIF: differential item functioning; HAQ: Stanford Health Assessment Questionnaire.

Table 5.12a: Results from the *k*-means cluster analysis

HAQ Item	**HAQ Item Level**[a]				
	Cluster 1	**Cluster 2**	**Cluster 3**	**Cluster 4**	**Cluster 5**
Three Clusters					
Standup	0	1	2	---	---
Liftcup	0	1	1	---	---
Climstep	0	1	2	---	---
Benddown	0	1	2	---	---
Pain and discomfort	1	1	2	---	---
Four Clusters					
Standup	0	1	1	2	---
Liftcup	0	1	2	2	---
Climstep	0	1	0	2	---
Benddown	0	1	1	2	---
Pain and discomfort	1	1	1	2	---
Five Clusters					
Standup	0	1	1	1	2
Liftcup	0	0	1	1	2
Climstep	0	1	1	2	3
Benddown	0	1	1	1	2
Pain and discomfort	1	1	1	1	1

[a] Levels in the Stanford Health Assessment Questionnaire (HAQ): 0 = no difficulty with the task, 1 = some difficulty with the task, 2 = much difficulty with the task, and 3 = inability to perform the task. Levels for the pain and discomfort dimension in the EQ-5D: 0 = no pain and discomfort; 1 = moderate pain and discomfort; and 2 = extreme pain and discomfort.

Table 5.12b: Results from the *k*-means cluster analysis (excluding the pain and discomfort)

HAQ Item	HAQ Item Level[a]				
	Cluster 1	Cluster 2	Cluster 3	Cluster 4	Cluster 5
Three Clusters (excluding the pain and discomfort domain in the k-means algorithm)					
Standup	0	1	2	---	---
Liftcup	0	1	1	---	---
Climstep	0	1	2	---	---
Benddown	0	1	2	---	---
Four Clusters (excluding the pain and discomfort domain in the k-means algorithm)					
Standup	0	1	2	2	---
Liftcup	0	0	1	2	---
Climstep	0	1	2	3	---
Benddown	0	1	1	2	---
Five Clusters (excluding the pain and discomfort domain in the k-means algorithm)					
Standup	0	1	1	2	2
Liftcup	0	0	0	1	2
Climstep	0	1	2	2	3
Benddown	0	1	1	1	2

[a] Levels in the Stanford Health Assessment Questionnaire (HAQ): 0 = no difficulty with the task, 1 = some difficulty with the task, 2 = much difficulty with the task, and 3 = inability to perform the task.

Table 5.13: Frequency of level responses for the pain and discomfort dimension of the EQ-5D[a,b]

	No Pain and Discomfort (%)	Moderate Pain and Discomfort (%)	Extreme Pain and Discomfort (%)
All Respondents	64 (10.7)	418 (69.7)	118 (19.7)
Very Mild RA	58 (23.4)	183 (73.8)	7 (2.8)
Mild RA	2 (1.2)	134 (82.2)	27 (16.6)
Moderate RA	2 (1.9)	60 (55.6)	46 (42.6)
Severe RA	0	26 (52.0)	24 (48.0)
Missing	2 (3.1)	15 (3.6)	14 (11.9)

[a] Rheumatoid arthritis (RA) grouping by cluster analysis.
[b] EQ-5D: EuroQol-5D.

Table 5.14: Final rheumatoid arthritis health state descriptions

Health State X[a]	Health State Y[b]	Health State Z[c]
You have **some** difficulty bending down to pick up clothes from the floor	You have **some** difficulty bending down to pick up clothes from the floor	You have **much** difficulty bending down to pick up clothes from the floor
You have **some** difficulty climbing up 5 steps	You have **much** difficulty climbing up 5 steps	You are **unable** to climb up 5 steps
You have **no** difficulty lifting a full cup or glass to your mouth	You have **some** difficulty lifting a full cup or glass to your mouth	You have **much** difficulty lifting a full cup or glass to your mouth
You have **some** difficulty standing up from a straight and armless chair	You have **much** difficulty standing up from a straight and armless chair	You have **much** difficulty standing up from a straight and armless chair
You have **mild** pain and discomfort	You have **moderate** pain and discomfort	You have **extreme** pain and discomfort

[a] The HAQ items for this health state is based on cluster two of the four-cluster model.
[b] The HAQ items for this health state is based on cluster three of the four-cluster model.
[c] The HAQ items for this health state is based on cluster four of the four-cluster model.

Table 5.15: Patient characteristics of the final health states[a]

Cluster Groups	n[b]	Mean	Standard Deviation	Minimum	Maximum
Mild Rheumatoid Arthritis					
Age[c]	163	61.9	12.6	16.8	88.3
Duration of RA (years)[d]	163	16.1	12.3	1.3	61.1
HAQ[e]	163	1.59	0.58	0.13	3.00
EQ-5D[f]	163	0.66	0.16	0.17	1.00
EQ-5D VAS[g]	163	59.6	18.10	15.00	98.00
Female	127 (78%)				
Moderate Rheumatoid Arthritis					
Age[c]	108	61.2	14.1	23.3	89.6
Duration of RA (years)[d]	108	17.3	10.5	0.7	44.2
HAQ[e]	108	2.17	0.29	0.88	2.75
EQ-5D[f]	108	0.52	0.18	0.20	0.85
EQ-5D VAS[g]	108	52.3	18.32	13.00	99.00
Female	92 (85%)				
Severe Rheumatoid Arthritis					
Age[c]	50	63.1	12.4	31.4	89.9
Duration of RA (years)[d]	50	20.4	15.0	3.0	75.8
HAQ[e]	50	2.51	0.25	2.00	3.00
EQ-5D[f]	50	0.39	0.20	-0.04	0.75
EQ-5D VAS[g]	50	45.1	21.55	5.00	92.00
Female	43 (86%)				

[a] EQ-5D: EuroQol 5D; EQ-5D VAS: EuroQol-5D visual analogue scale; HAQ: Stanford Health Assessment Questionnaire; RA: rheumatoid arthritis.
[b] n = 31 missing, n = 248 in 'very mild rheumatoid arthritis' state
[c] one-way ANOVA results: F = 0.72, p = 0.54
[d] one-way ANOVA results: F = 2.35, p = 0.07
[e] one-way ANOVA results: F = 285.03, $p \leq 0.001$
[f] one-way ANOVA results: F = 188.32, $p \leq 0.001$
[g] one-way ANOVA results: F = 64.76, $p \leq 0.001$

Figure 5.1a: An example of appropriately ordered item levels (i.e., each level is distinctly in its place)

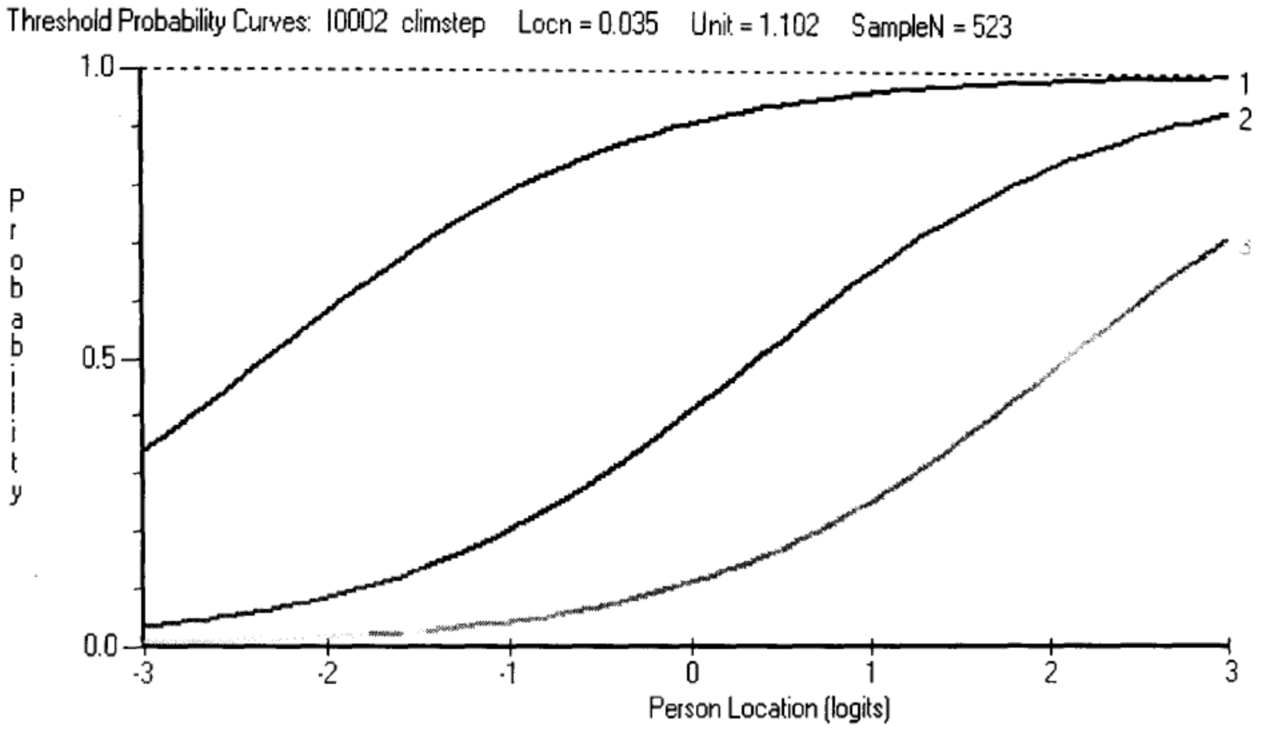


Figure 5.1b: An example of disordered item levels (i.e., the 3rd level is in between the 1st and 2nd levels)

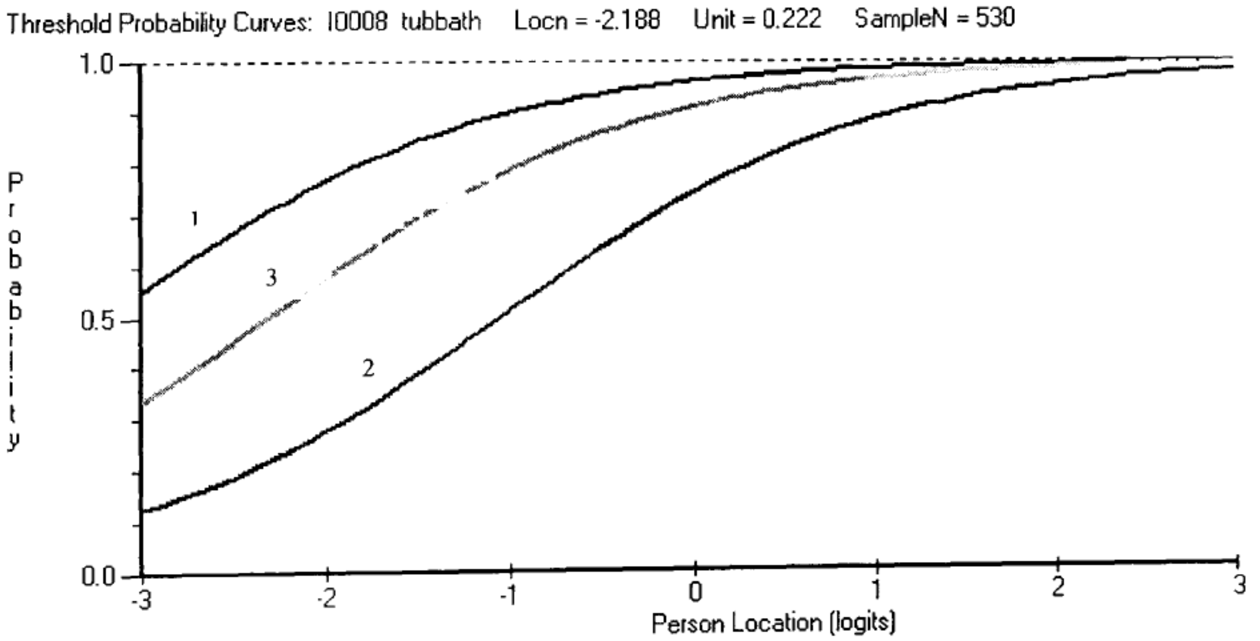


Figure 5.1c: An example of poorly spread item levels (i.e., the 2nd and 3rd levels are too close together to be discrete item levels)

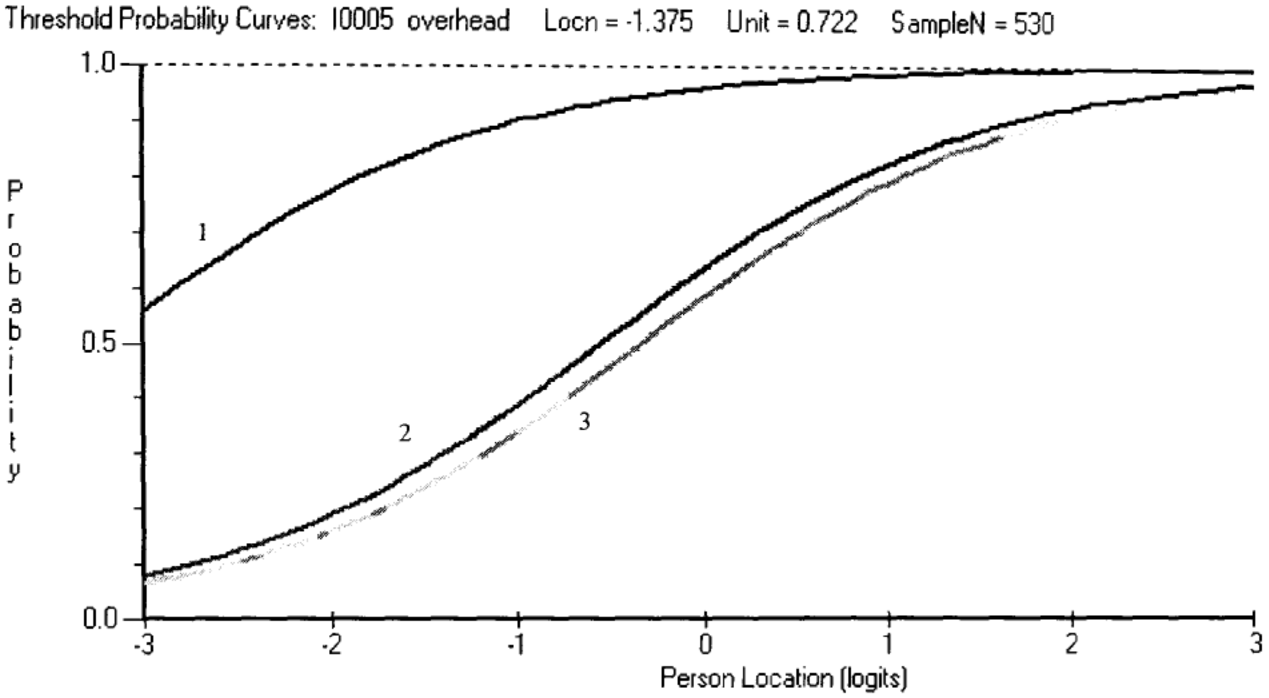


Figure 5.1d: An example of a level lying too close to 0% probability (i.e., the 3rd level is not readily discernible)

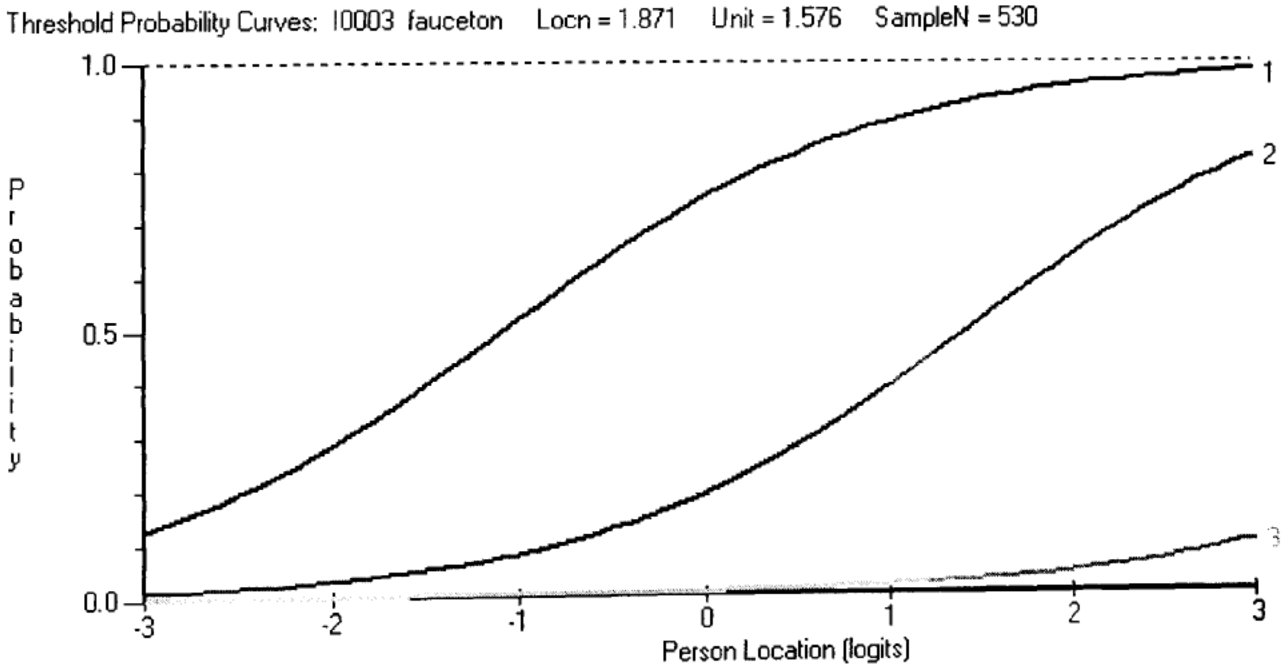
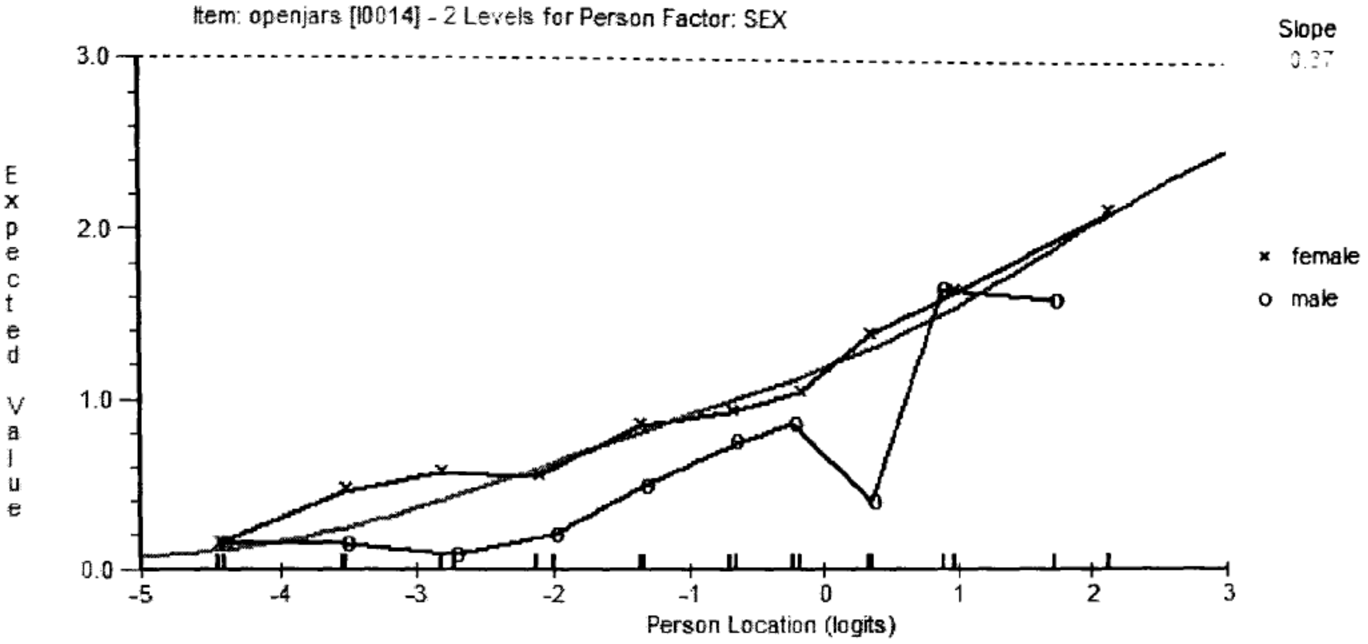


Figure 5.2: Item characteristic curve for *openjar* item stratified by sex



Chapter Six

The Design of the Qualitative and Quantitative Components

The preceding chapter described the first of the three components of this sequential mixed-methods study. Using previously collected patient-level data, Rasch and cluster analyses were used to generate three health states describing different severities of rheumatoid arthritis (RA). These states were constructed for use in the final two components of the mixed-methods study design (for details refer to Section 4.2); the results from which are described in Chapters Seven and Eight. The first of the remaining components explored the effect disease adaptation information may have on individuals' perceptions of RA using qualitative interviews (Chapter Seven). The final component determined whether or not information about disease adaptation was effective at altering the respondents' initial health state values, as well as identifying the factors that contributed to these changes (Chapter Eight).

The remaining two components of the mixed-methods study, described above, required the collection of responses from members of the general population. As both of these components investigated the effects of disease adaptation information on individuals' perceptions and health state values, the two studies used very similar designs to meet their objectives. Where slight differences between the design and implementation of the studies used in the final two components occur, this will be described in the pertinent chapters. The fundamental aspects common to both designs include the participant groups, the valuation tasks, the adaptation exercise, and the patient values presentation. This chapter aims to describe, in detail, these elements of the study. The decision to describe these elements here as a prelude to the chapters describing the remaining two components is intended to avoid over repetition of this material in the subsequent results chapters.

6.1 Study Participant Groups

A key feature for the remaining two components of the mixed-methods study is the intervention aimed at informing the respondents about disease adaptation, hereafter referred to as the 'adaptation exercise'. It was initially intended that members of the general population participants be recruited and interviewed individually to provide valuations for RA states both before and after undertaking the adaptation exercise. However, individual interviews run the risk of participants adjusting their valuations in an attempt to please the interviewer; this is known as prevarication or intervention bias (Hiebert and Nordin, 2006). Similarly, there is concern that an interviewer may subconsciously coerce the respondents to change their values in order to obtain positive research results; this is referred to as interviewer bias (Last, 2001). To avoid these potential interviewer effects, the general population respondents were subdivided into two groups, which undertook different health state valuation pathways, as will be discussed in more detail below. While this increased the required sample size, at least for the final quantitative component, it was necessary to ensure that the results represented the unbiased influence of the adaptation exercise on health state values.

The respondents were randomly allocated into the 'Initially Uninformed Group' and the 'Informed Group' (Figure 6.1, page 131). Both groups completed the health state valuation exercises (described in Section 6.2) and underwent the adaptation exercise (described in Section 6.3). The groups were differentiated by the fact that the Initially Uninformed Group completed a set of health state valuations before being presented with the adaptation exercise, while participants in the Informed Group undertook the adaptation exercise prior to completing any health state appraisals. The main group used to measure the effect of the disease adaptation information was the Initially Uninformed Group, for whom before and after adaptation exercise values were available. The Informed Group was created to identify potential interviewer effects, such as prevarication bias (Hiebert and Nordin, 2006) and interviewer bias (Last, 2001), as described above; this group thereby helped to identify the true effect of the intervention. By matching the characteristics of the respondents in both groups, a comparison between the values elicited after the adaptation exercise in both the Initially Uninformed and Informed Group was able to evaluate the impact the interviewer may have had on respondents' health state values; this is discussed further in Chapter Eight. To utilize the full potential of having a second group, the Informed Group also examined the influence of viewing health state values provided by patients actually living in the described health states (described in Section 6.4).

Individuals in the Initially Uninformed Group first valued the health states (refer to Section 6.2). This was followed by an adaptation exercise, where they listened to audio-recordings of patients discussing adapting to life with RA (discussed in greater detail in Section 6.3). After the adaptation exercise, individuals in the Initially Uninformed Group once again valued the same states using the same valuation methods that they had done previously. Individuals in the Informed Group, on the other hand, underwent the adaptation exercise before valuing the health states for the first time. After the first valuation, they then underwent a 'patient values presentation', where they were shown health state values provided by patients living in the same health state (Section 6.4); they were also shown their own values from the first valuation. The implications of their values were explained at this time. After this presentation, they were asked to repeat the health state valuations.

Individuals who participated in the qualitative and quantitative components were offered £10 cash for their time or the opportunity to donate the £10 to the Arthritis Research Campaign[11]. While it is acknowledged that the provision of a financial incentive may result in differing levels of participation amongst individuals in different socio-economic backgrounds, for this project an incentive was felt to be necessary to encourage participation. As the target sample was composed of members of the general population, and the interview process was expected to take a significant amount of time to complete (on the order of one hour), the decision was made to offer an incentive to ensure that a reasonable number of participants could be recruited in a timely fashion. Furthermore, without a small monetary incentive, the integrity of the study results could have been jeopardized by participants whose motive for involvement was a particular interest in health issues, either as patients or as family members and/or friends of patients.

6.2 Valuation Tasks

Respondents completed a series of valuation exercises (a copy of the questionnaire booklet is presented in Appendix A.4, page 247). First, all respondents provided a self-report of their health. Then the respondents in both groups completed two identical series of visual analogue scale (VAS) and time trade-off (TTO) exercises.

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[11] The Arthritis Research Campaign raises funds to promote medical research into the cause, treatment and cure of arthritic conditions; to educate medical students, doctors and allied healthcare professionals about arthritis; and to provide information to the general public. Further information is available from: http://www.arc.org.uk/.

Whether the individuals completed these valuation tasks at the start of the interview or after the adaptation exercise depended on their group allocation, as discussed in Section 6.1 (Figure 6.1, page 131).

6.2.1 Self-Report of Own Health

As an initial evaluation, the respondents provided a self-report of their own health by rating themselves against the individual items of the health state description. These items were: difficulty with bending down to pick up clothes from the floor, difficulty climbing up five steps, difficulty lifting a full cup or glass to one's mouth, difficulty standing up from a straight and armless chair, and the presence of pain and discomfort (Table 5.14, page 111). This particular exercise served to familiarize the respondents with the items in the health state descriptions to be appraised in subsequent valuation tasks; it was completed only once by the participants.

6.2.2 Visual Analogue Scale

The respondents first rated six health states – full health, own current health, dead, and the three constructed RA states (Table 5.14). The states were written on individual cards, which were shuffled by the interviewer. The health state on top of the pile was shown to the respondent to rate first; this was repeated until all health states were seen by the respondent. This process ensured that the order in which the respondents valued these states was randomized for each individual. The states were scored on a single vertical VAS, graded from zero to 100, which represented the worst and best imaginable states, respectively. The results provided an indication of the individual's ordinal rankings of the health states and the intensity of those preferences (Drummond et al., 2005). The respondents were not explicitly told that they were valuing states pertaining to RA. Instead, as discussed in Section 5.3.6, the RA states were labelled as States X, Y, and Z (i.e., mild RA, moderate RA, and severe RA) to avoid any pre-conceived ideas respondents may have had regarding the condition.

To enable easy comparison of the health state values obtained from the various elicitation methods, the VAS values were standardized. This operation allowed the values describing the respondent's current health state ('Your Own Health') and the three RA states to be anchored at zero, representing 'Dead', and one, representing 'Full Health' (The EuroQol Group, 1990):

$$Value_{Standardized} = \frac{(Unstandardized\ Health\ State\ Value - Dead\ Value) * 100}{Full\ Health\ Value - Dead\ Value}. \quad (6.1)$$

For example, if an individual valued a RA state at 70 on the VAS with anchors at 100, representing full health, and at 20, representing dead, then the standardized VAS value would be 62.5. Applying the above equation to the VAS values permits health states to be considered to be worse than dead from the perspective of the participant. The standardized values can be divided by 100 to enable comparisons with those values obtained using the time trade-off approach (described in the following section).

6.2.3 Time Trade-Off

The final component of the valuation tasks was a self-completed bottom-up titration time trade-off (TTO) exercise (Gudex, 1994) in which the three derived RA states were valued. As with the VAS exercise, the order in which the participants valued the RA states was randomized; this was dependent on what version of the questionnaire booklet the participants received. The TTO was administered using a self-completed version, rather than an interviewer-administered version with a time board. The self-completed version was selected to reduce the amount of time required to complete the entire interview process; using an interviewer script and time board would likely have increased the time required to obtain the values for the three RA states from the respondent. To ensure that the respondent understood the task, the interviewer provided the respondent with a practice question which they worked through together. The interviewer was also available to address any question the respondent may have had when completing the TTO exercises.

Respondents were presented with a choice of two scenarios and asked to make a trade-off between length and quality of life (QOL). In the case of a TTO valuation for an impaired state considered to be better than dead, one of the scenarios consisted of living in the hypothesized state for a fixed period of time: 25 years was used for this research. The other scenario – the trading scenario – consisted of varying lengths of time (*t*) in full health, never being affected by the disease. Both prospects were followed by death. The value of *t* was varied in the trading scenario until a point of indifference between the two scenarios was observed. The value for the hypothesized health state considered to be better than dead was calculated as:

$$Health\ State\ Value_{States\ better\ than\ dead} = {}^{t}\!/_{25}\,, \qquad (6.2)$$

where *t* is the number of life years for which a respondent was indifferent to living in the hypothesized health state and living in full health. So, for example, if an individual indicates that 20 years is the point of indifference between the two scenarios, then the health state value would be 0.8 (20/25).

In the case of states considered to be worse than dead, a different procedure was used to calculate the value of the health state. Using the protocol outlined in the Measurement and Valuation of Health (MVH) study (Dolan, 1997), when respondents deemed an impaired state to be worse than dead, they were presented with two scenarios, different from those outlined in the previous paragraph. The first scenario was to live in the impaired health state for *t* years followed by full health for $(25-t)$ years; the second scenario to die immediately. The value for a state worse than dead was determined by:

$$Health\ State\ Value_{States\ worse\ than\ dead} = -\left(25 - {}^{t}\!/_{t}\right). \qquad (6.3)$$

While it may not seem realistic for respondents to consider a scenario which has them living in an impaired health state for a specified length of time followed by complete recovery from the symptoms, this order forces respondents to consider being in an RA state for t number of years and to think specifically about the sacrifice being made (Gudex, 1994). There is concern that the respondents may not consider the trade-off appropriately if the ordering of the time spent in the impaired state and full health were reversed. Respondents, for example, might consider prematurely ending their life once they had enjoyed the time in full health, before the impaired state to be valued begins. Such reasoning, of course, would not lead to a valid assessment of their true desire to avoid living in a given health state.

One of the main concerns with the formula to calculate values for states worse than dead (Equation 6.3) is that the denominator is no longer a fixed number like that in the case for health states better than dead (Equation 6.2). Furthermore, these values can become very negative. For example, with a 25-year time horizon, the minimum value for states worse than dead would reach -24 (-24/1), if trade-offs were limited to whole years. As a result, the common practice is to transform these values so that the negative values fall in the range of [-1,0] (Patrick et al., 1994):

$$Transformed\ Health\ State\ Values_{States\ worse\ than\ dead} = -\left(25 - {}^{t}\!/_{25}\right). \qquad (6.4)$$

Patrick et al. (1994) advise that the negative values for states worse than dead, once transformed, can no longer be interpreted to be on the same scale as those for states better than dead. This can make comparison and aggregation of positive and negative values problematic.

The TTO approach was used as the valuation technique for this thesis because individuals tend to understand numbers, in terms of life years, better than percentages, in terms of risk probabilities (i.e., the standard gamble approach) (Drummond et al., 2005). Therefore, the TTO was chosen to alleviate the cognitive burden placed on the respondents. A 25-year time horizon, which is greater than the conventional ten-year time frame (Dolan, 1997), was chosen for the TTO exercises. The purpose of using a greater number of years was to ensure enough sensitivity to assess any changes that might arise in the subsequent valuations, and to avoid easy calculation of the implied values by the respondents when completing the same exercises the second time. However, it is recognized that the longer time horizon may make it difficult for older respondents to make realistic trade-offs between length of life and QOL.

Administration of the Valuation Exercises

Prior to recruiting participants for the final two components of this mixed-methods study, the Ph.D. candidate (for the qualitative component) and the interviewer (for the quantitative component) went through the entire protocol several times to ensure that she was fully aware of how to conduct the TTO, calculate the health state values, and present this information back to the respondents (refer to Appendix A.5, page 268, for the protocol for the qualitative component and Appendix A.6, page 279, for the protocol for the quantitative component). As the Ph.D. candidate was the interviewer for the qualitative component, mock interviews were conducted with willing students and staff members (mostly academic) at the School of Health and Related Research. For the quantitative component, an interviewer from Sheffield Hallam University was employed. Even though the interviewer had previous experience of administering VAS and TTO exercises, the Ph.D. candidate trained the interviewer to ensure that the interviews were being conducted to her specifications. First, the Ph.D. candidate went through the entire protocol with the interviewer and presented all the questionnaires that were used; the interviewer was requested to read through the protocol. The second session involved the interviewer going through the interview protocol with the Ph.D. candidate acting as the 'respondent'; any changes to delivery of the questions were discussed at this point. Then the interviewer repeated the protocol with a member of staff from Sheffield Hallam University with the Ph.D. candidate present as witness to this 'interview'. The interviewer was requested to practise the protocol with family members prior to conducting interviews with members of the general population. The interviewer was also advised to contact the Ph.D. candidate whenever any questions may arise from the interviews. The Ph.D. candidate also accompanied the interviewer for random visits throughout the data-collection phase. This allowed the candidate to ensure that the interview process was not changing over time.

The cognitive complexity of the TTO exercise meant that respondents needed to be trained in the TTO method before conducting the first valuation exercise. To achieve this, all the respondents completed the practice question (Appendix A.4, page 247). For this question, the interviewer instructed the respondent to look at the first row of the TTO table (i.e., "would you prefer to live in Health State A for 25 years or Full Health for 0 years?"). If the respondent preferred to live in the impaired health state than to die immediately, the interviewer instructed the individual to proceed with the remaining questions starting from the bottom of the table (i.e., "would you prefer to live in Health State A for 25 years or Full Health for 25 years?"). Once the respondent had recorded their preference for this question, the interviewer advised the respondent to move upwards on the table and indicate their preference. This process was continued until all the rows of the TTO exercise were completed. If the respondent experienced any difficulty (i.e., uncertainty over which response they preferred) at a certain question, the interviewer instructed the individual to start from the top of the table (i.e., the second question: "would you prefer to live in Health State A for 25 years or Full Health for 1 year?") and move downwards on the table until the respondent indicated a preference for all the rows on the TTO exercise.

Although the interviewer did not need to determine the value for the health state for the practice question, calculations were needed for the subsequent three TTO exercises (i.e., State X, State Y, and State Z); this was done for both the Initially Uninformed and Informed Groups in the qualitative component but for the Informed Group *only* in the quantitative component prior to the presentation of personal and patient values. For a completed TTO exercise, As, Bs, and equal sign(s) may be present; however, at times, an equal sign may be absent from an individual's response. If only one equal sign was presented amongst a series of As and Bs, the interviewer was instructed to use this as the point of indifference in the determination of the health state value (Equation 6.2). In a case where the individual did not place an equal sign on any of the rows of the TTO exercise (i.e., only As and Bs are present), the interviewer used where the respondent changed from As to Bs as the point of indifference. For example, if a respondent indicated As from 0 to 17 years in Full Health and Bs from 18 to 25 years in Full Health, the indifference point used was 17.5 years. In a case where the individual used a series of equal signs in their response, the interviewer was instructed to use the midpoint of all the equal signs to calculate the health state value. For example, if a respondent placed equal signs for 17 to 19 years in Full Health, the interviewer used 18 years as the indifference point; if a respondent placed equal signs for 18 and 19 years in Full Health, the interviewer used 18.5 years as the indifference point. These same indifference points were used for the quantitative analysis of the results.

The indifference point for each of the three RA states was used in the calculation of the health state values (Equation 6.2). These were entered in a table to be presented to the respondent (an example used for the Informed Group in the quantitative component, is shown in Appendix A.7, page 298). To ensure that the respondents understood their values for the RA states, the interviewer informed them that each of the 'scores' was measured on a scale anchored from zero to one, where the lower bound represented a preference to live in a dead state and the upper bound represented a preference to live in full health; a higher score implies a more desirable health state to live in and, therefore, a lower score implies a less desirable health state. In addition to the value for each of the three RA states, the number of years the respondent was willing to forego in order to have full health was also presented; this was determined by subtracting the indifference point from 25.

The preceding discussion focused on the more common situation of respondents. If, however, the respondent preferred to die immediately rather than spend any time in the impaired health state (i.e., giving a response of 'B' for the first row of the TTO exercise), the interviewer instructed the individual to complete the specific question using the states worse than dead scenarios on the following page of the questionnaire booklet. The respondent was asked to look over the different scenarios and to take note that Choice A now referred to some time spent in the impaired state (i.e., Health State A) followed by some time in Full Health. The interviewer instructed the respondent to first indicate a preference for the choice in the bottom row (i.e., "would you prefer to live 0 years in Health State A followed by 25 years in Full Health, or to die immediately?") and then to move upwards until all the rows in table had a response.

While the actual values used in the quantitative analysis for these 'worse than dead' states were determined using Equation 6.4, the interviewer did not use this equation when calculating the values to present back to the respondents. Instead, the interviewer informed the respondent that they provided a value of less than zero for this health state. The interviewer further explained that this 'score' implied that the respondent felt that living in this particular state would be worse than being dead;

this was presented to the respondent in a tabular format (Appendix A.7, page 298). The rationale for this simplified calculation was to streamline the process for the interviewer to calculate values for states worse than dead to ensure that the overall time of the interview process was minimized.

6.3 Adaptation Exercise

Individuals in the Initially Uninformed and Informed Groups underwent the adaptation exercise but, as discussed earlier, the order in which a respondent completed the intervention differed between the groups (Figure 6.1, page 131). The adaptation exercise comprised of two parts: (i) common symptoms of RA and general limitations a patient may experience; and (ii) selected information obtained from the *Health Talk Online* website[12] (DIPEx Health Experiences Research Group, 2008). This award-winning website features videos and audio-recordings from a wide range of real patients and covers patient experiences with nearly 50 health conditions including cancers, heart disease, mental illness and other chronic health issues. The website is intended for use by patients, their care-givers, family, and friends. It is also used for teaching and training by a wide variety of health professionals.

For the second stage of the adaptation exercise, excerpts of interviews from RA patients of varying age and disease duration were extracted from the *Health Talk Online* website. It should be noted that, although the case study for this thesis was RA, the disease under investigation was simply referred to as ‘arthritis’ in the presentations to the respondents. The intention of this generalized labelling was to avoid cognitively overburdening the study participants with scientific definitions of the various forms of arthritis. The interviewer did have the clinical definitions of both RA and osteoarthritis readily accessible if the participants questioned the distinction between the two terms. As such, the terms ‘arthritis’ and ‘RA’ are used interchangeably in the following description of the adaptation exercise, to reflect the language used when speaking directly with the participants.

While the main purpose of the adaptation exercise was to promote concepts of disease adaptation through the use of patient interviews retrieved from the aforementioned website, the participants were initially asked if they knew (i) what the common symptoms of arthritis were and (ii) whether they knew anyone who suffered from arthritis. The aim of these questions was to encourage individuals to

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[12] Previously known as the *Personal Experiences of Health and Illness* website.

think about life with arthritis and, more importantly, to ease them into the adaptation exercise. Each participant listened to three excerpts from patient interviews – hereafter referred to as 'recorded scenarios' to differentiate them from the interviews conducted in the qualitative component – which highlighted the health and lifestyle implications of living with RA, with an emphasis on disease adaptation. Because of confidentiality issues, the names of the patients were not provided on the *Health Talk Online* website; thus, the patients were given fictitious everyday names so that the study participants could easily differentiate one from another.

Ideally, a mixture of male and female recorded scenarios would have been used in the adaptation exercise. However, there were only limited recordings relating to adaptation to RA available on the *Health Talk Online* website. Therefore, the decision was made to use the best messages highlighting different aspects of disease adaptation regardless of the gender of the patients. It is important to note that the constructed adaptation exercise focuses strictly on adaptation rather than response shift (refer to Section 2.1.1 for a detailed description of disease adaptation and response shift). To fully examine the impact of response shift on health state values, it would be necessary to not only encourage the participants to consider themselves in the hypothetical health state but also to encourage them to consider life in the health state relative to QOL of other patients (i.e., response shift). Due to the complexity involved with promoting this concept of social comparison, it was decided that the concept of response shift will not be explored in this thesis.

6.3.1 Recorded Scenario One

The first recorded scenario was that of 'Lisa', a 30-year patient who had been diagnosed as having RA for one year at the time of the recording. The full transcript of the recording is:

> I didn't let anyone know how bad it was. You put a front on. It wasn't until I got indoors that I'd do the little weeping and the wailing kind of thing [laughs]. So yeah, I don't, I don't think they really knew, like, as I say, my Mom didn't know until we'd gone to [the] Zoo, how bad I was. And she was really, really shocked. 'Cuz I just didn't tell, you know, I'd just got on with it. Struggled, I didn't, you know, I didn't cope with it. I struggled. But as far as everyone else was aware it wasn't as bad as, you know, obviously for [daughter's name] and my husband, they didn't really know how bad it was. So I did cope with, I could go to Hollywood, couldn't I? I could be in Hollywood. But no, I did, I did really, yeah, yeah, I did cover it.

> I think one instance we'd gone to, we'd gone out with my brother-in-law and all our families and I was, just sat down normally. I was sat in a club kind of thing, you know, sat down having a drink and it was just like, "I've got to go to the toilet" and it took me about five minutes, to get up, to get up and get out of the chair. And you know people were going, "We didn't realize you were that bad". 'Cuz I just couldn't get my body to do anything.

This recorded scenario was selected because it highlights the struggles a patient may face when initially presented with arthritis or, for that matter, any health condition. Specifically, Lisa attempted to cope with RA by masking her symptoms from her family and friends.

After hearing Lisa's recorded scenario, the participants were asked a series of questions: (i) can you summarize the information you just heard and (ii) have you, or someone close to you, been in a similar situation to the one that the patient described; if not, can you imagine yourself acting in the same manner as the patient? After the participant answered these questions, the Ph.D. candidate's interpretation of the recorded scenarios was provided by the interviewer. The respondent was then asked if he/she had any additional comments to add. This same format was repeated with the two other recorded scenarios extracted from the *Health Talk Online* website. The topic guide of the adaptation exercise is presented in Appendix A.5, page 268.

6.3.2 Recorded Scenario Two

The second recorded scenario of the adaptation exercise is that of 'Ann'. She was in her fifties and had been living with diagnosed RA for four years. This excerpt was selected because it provided the respondents, who may not have illness experience, with a tangible example of having to make lifestyle changes when dealing with a significant life event or, in this case, an impaired health state. The full transcript of the recording is:

> But, and then I think it was about two years ago now I started swimming and that has just been fantastic. Because that is something I can do and I do it five days a week, every morning. I started off it, doing, it was this time of year, October, I got into the pool and I could do 35 lengths and I thought by Christmas I want to swim a mile and at Christmas I did. I was doing my 64 lengths in the hour.
>
> And now there's a new pool opened, and the same group of people go, and we all sort of, I mean they're not all sufferers, some just go because they enjoy going but we all sort of support each other, if you like, and I haven't been for two days this week so I'm already in trouble.

> But I can swim now for about an, well I could swim for two hours if I wanted to but I don't because I have other things to do, but I, I have found that that has helped and my consultant, you know, just sees me, says, "ah, my swimmer". You know, he's, he's really impressed that of the you know, the way I've sort of dealt with it. I didn't think, "Ah, my life has ended, I'm never going to be able to do anything". I just thought "Well OK, this is what it is and I'm not going to let it beat me, you know". So I don't, I try to do everything as I did before, but in moderation and that seems to have worked quite well so far. I do still have bad days and sometimes the medicine upsets me.
>
> But I would say, in general, I feel better now than I did, you know, sort of four or five years ago.

6.3.3 Recorded Scenario Three

The final recorded scenario in the adaptation exercise is that of 'Patricia', who was in her late seventies and had lived with diagnosed RA for over 40 years. This excerpt was chosen because it illustrates the general concept of disease adaptation. Importantly, it summarizes the information contained in the earlier recorded scenarios: from being initially in denial when diagnosed with a health condition (i.e. Lisa's scenario) to realizing that changes are required to accommodate the health condition (i.e. Ann's scenario). Patricia's interview also shows that the individual is able to learn what gives her pain relief. The full interview transcript is:

> As I said earlier on, there are three ways you can deal with arthritis and I've found this out personally when I first started this. You can be very angry and fight it. That only lasts for a certain time because the only one that's getting hurt is you. 'Cuz the more of a temper and, and that you get in the more you create, "Ooh, that hurts", sort of thing.
>
> The other thing is you can give in right from the beginning and you can say, "I can't do that". And let everybody else do it for you and give no thought to the fact that they've got their lives to live and they shouldn't be feeling that way that they've got to do it for you.
>
> And the third thing is to come terms with it and don't live against it, live with it. And when you get a bad pain just sit, whatever suits you. If you get a bad pain and painting the wall gives you relief, go and paint the wall. If you find, like me myself, the only way to get over it is to just sit quietly and rest and it will go.

6.3.4 Deliberation Period

After hearing each of the recorded scenarios, the respondents were encouraged to reflect upon, debate, and discuss this new information. All participants were asked what they felt about the information presented. The Initially Uninformed Group members in the qualitative component were specifically asked if this information would have been beneficial before completing their first valuation task. They then did their second (and last) valuations. Those in the Informed Group completed their first health state valuations after undergoing the adaptation exercise. Individuals in the Informed Group then underwent the patient values presentation, described below, before completing the second valuations.

6.4 Patient Values Presentation

Another way to inform the general population about the possibility of disease adaptation is to present them with patient values for the health states they valued. As this thesis examined the formation of informed general population values and how they might impact health state values, obtaining actual patient values was not an objective of this project. Instead, previously published results (Tijhuis et al., 2000) provided an indication of how patients, living in different severities of RA, valued their health states. Patient values were presented to individuals in the Informed Group prior to their second valuation. Tijhuis et al.'s (2000) study assessed the psychometric properties – feasibility, reliability, and validity – of the TTO approach in a group of patients living with RA. Specifically, they wanted to explore the ability of the TTO values to relate to different levels of HAQ scores. To investigate this, patients' TTO values of their current health were compared with their self-reported HAQ scores (Table 6.1, page 131).

Using the published results, a linear relationship was developed to explain how the HAQ score predicts the TTO value. This was achieved by running a linear regression using the average for each range of HAQ scores reported in the study by Tijhuis et al. (2000) – for example, for a range of 0 to 0.75, a score of 0.375 was used in the modelling – and the reported TTO values. Since only three RA states were needed for the valuation studies, and given the fact that there was no significant difference between the TTO values for the two most severe HAQ levels (i.e., both levels reported TTO values of 0.64), an average HAQ score of 2.255 was used to represent a severe form of RA. This led to the derivation of the following equation:

$$TTO = 0.889 - 0.103 * (HAQ\,score)\ , \tag{6.5}$$

This analysis was conducted using SPSS version 14 for Windows (SPSS, Chicago, IL, USA).

For each of the three constructed RA states for the present study, a HAQ score was determined. This was achieved by summing each of the HAQ item scores, where zero, one, and two represented no, some, and much difficulty in performing the specific task, and three represented an inability to perform the task, and taking an average. (The scoring of the HAQ is described in greater detail in Section 5.2.1.) The HAQ scores for the three RA states were determined to be 0.75, 1.50, and 2.25; as mentioned previously, higher scores imply greater disability. These scores may be considered to be crude because the health state description contained the pain and discomfort dimension from the EQ-5D. Since pain and discomfort was not a part of the HAQ instrument, it was not included in the calculation of the HAQ score; thus, the resulting score may be placed on the health state. These HAQ scores were substituted into the regression equation (Equation 6.5) to obtain modified TTO values, which were presented to individuals in the Informed Group (Table 6.1, page 131).

There are disadvantages to using patient TTO values from another published study, but it was considered to be a more realistic alternative than presenting fictional patient values to the individuals (e.g., adding a unit of 0.1 to the respondent's health state values). Because the previous results (Tijhuis et al., 2000) were modified using regression techniques to align them with the health states developed specifically for this thesis, a numerical illustration of how patients valued their life with RA was created using the 25-year time horizon.

In addition to having a being presented with patient values derived from the TTO, the Informed Group was also shown their own initial TTO values for the three RA states. They were given the opportunity to reflect on the information before completing their second, and final, valuation.

6.5 Summary

This chapter described the four fundamental components of the final qualitative and quantitative studies that complete this sequential mixed-methods study design. These components were the respondent group, valuation tasks, adaptation exercise, and patient values presentation. Participants were allocated into one of two groups: the Initially Uninformed Group and the Informed Group. The purpose of having two groups was to assess the presence of any interviewer effects; evaluating the influence of an intervention with a single group runs the risks of prevarication and interviewer biases. All respondents provided a self-report of their health, and valued a series of health states using the visual analogue scale and time trade-off approaches. All respondents were informed about disease adaptation by an adaptation exercise, which consisted of three audio-recordings of patients discussing adaptation; they were encouraged to discuss and reflect upon this information. Finally, individuals in the Informed Group were further informed about adaptation by a presentation of patient values for the RA states they valued.

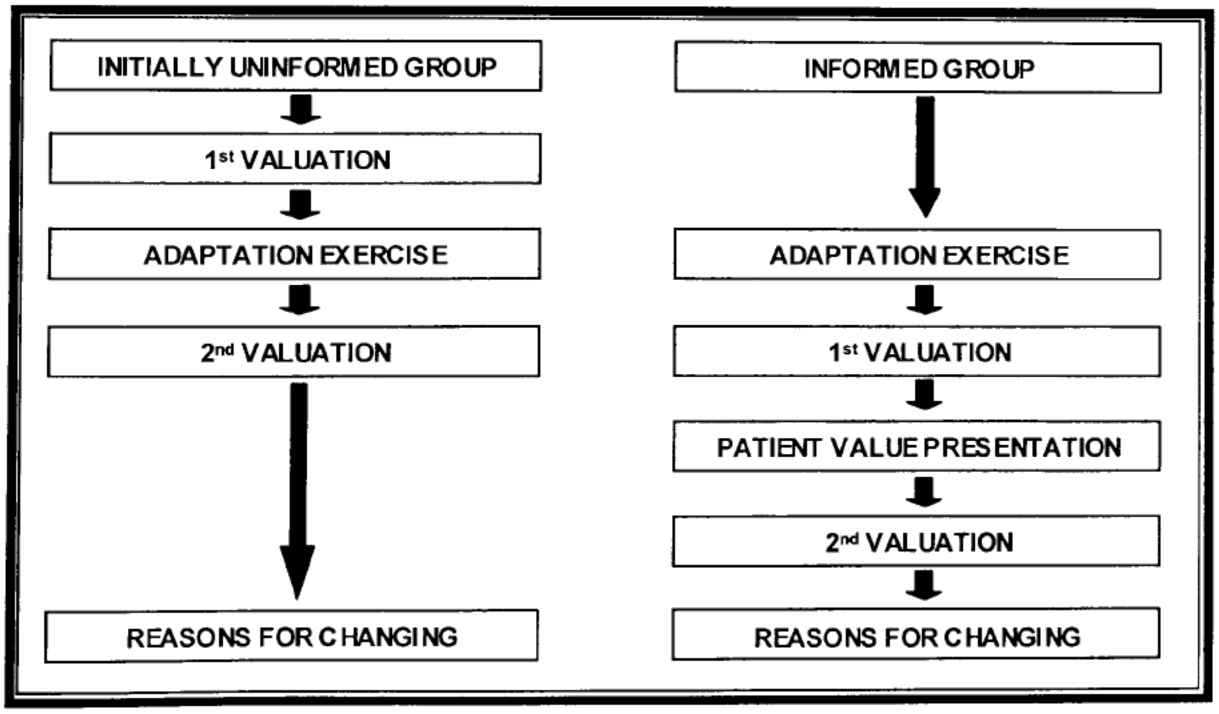
6.6 Tables and Figures

Table 6.1: Time trade-off (TTO) values within different categories of the Stanford Health Assessment Questionnaire (HAQ)

HAQ Score[a]	TTO Median (Range)[a]	TTO Values used for the Patient Values Presentation
0.00 – 0.75	0.87 (0.30 – 1.00)	0.81
0.76 – 1.50	0.74 (0.03 – 1.00)	0.73
1.51 – 2.25	0.64 (0.04 – 1.00)[b]	0.66
2.26 – 3.00	0.64 (0.05 – 1.00)[b]	0.66

[a] Information taken from Tijhuis, G.J., Jansen, S.J.T., Stiggelbout, A.M., Zwinderman, A.H., and Hazes, J.M.W. (2000). Value of the time trade off method for measuring utilities in patients with rheumatoid arthritis. *Annals of Rheumatic Diseases*, **59**, 892-897.
[b] Analysis of variance: $p < 0.01$ (in comparison with 0.87).

Figure 6.1: The design of the qualitative and quantitative components



Chapter Seven

Component Two: A Qualitative Exploration of How Disease Adaptation Information Affects the General Population's Perceptions of Rheumatoid Arthritis*

7.1 Introduction

Developing distinct and comprehensible rheumatoid arthritis (RA) states for individuals to value, as described in Chapter Five, is one aspect of getting accurate valuations from the general population. However, there is still a need to determine how best to inform general population respondents about the health states they are valuing. If members of the general population underpredict their ability to adapt – or even ignore the possibility of adapting – to an impaired state (Kahneman and Snell, 2000), the resulting value may not be a true estimation of their preference for living in that health state. This has significant implications when these values are incorporated in cost-effectiveness analysis, as demonstrated in Chapter Two.

The description of health states can probably never be complete or perfect, but the better the description, the more accurate and reliable the outcome will be. It is obviously essential that respondents do not overlook the effect of illness on common or important life functions. Perhaps less obvious, but of comparable importance, is that the descriptions provide respondents with information about the magnitude and the nature of adaptation experienced by patients over time. There is some evidence that general population respondents appear to be receptive to information about disease adaptation (Damschroder et al., 2005; Ubel et al., 2005). While the findings from these previous studies have demonstrated that respondents do alter their health

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* A version of this chapter is published as McTaggart-Cowan, H., O'Cathain, A., Tsuchiya, A., and Brazier, J. (2009). A qualitative study exploring the general population's perceptions of rheumatoid arthritis after being informed about disease adaptation. School of Health and Related Research: Health Economics and Decision Science Discussion Paper, 09/02.

state values, there is an absence of evidence regarding how disease adaptation information influences individuals' perceptions of the impaired health states.

This chapter describes the second component of the sequential mixed-methods study presented in this thesis. Specifically, the current chapter aims to meet three objectives in addressing the broad question of how information pertaining to disease adaptation affects the general population's perceptions of RA. The first objective was to pilot-test the key features of the study design, including the adaptation exercise and patient values presentation (both described previously in Sections 6.3 and 6.4, respectively) on a sample of participants recruited from the general population. The second objective was to identify the aspects of disease adaptation information that have the greatest influence on perceptions of RA. The final objective was to develop a questionnaire that encompassed the main reasons for why individuals may or may not have altered their values when appraising RA states. The developed questionnaire will be used in the final component of the sequential mixed-methods study (described in Chapter Eight).

7.2 Methods

To understand how disease adaptation information impacts an individual's perceptions of RA, I undertook a qualitative methodological approach based on individual face-to-face interviews conducted with members of the general population[13].

7.2.1 Study Participants

A member of the staff of the Faculty of Health and Wellbeing[14] at Sheffield Hallam University and I identified members of the general population to participate in this study. Individuals in the city centre of Sheffield, a large industrial city in the north of England, were purposively sampled to maximize variation with respect to age and gender. Maximizing this variation was important because health behaviours are known to differ by age and gender (e.g., Deeks et al., 2009). While other factors, such as individual's experience with illness, may also affect responses, it was ethically difficult to obtain such information when approaching prospective study participants. Although an exact count was not recorded, approximately 50 individuals were approached in the city centre; of these, 12 agreed to participate in the study.

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[13] As noted in Chapter Four, a first person voice is used throughout this chapter to reflect upon my influence on the qualitative research process, as per the norms for qualitative research.

[14] Formerly the Centre for Research and Evaluation.

Due to the specificity of the research question, 12 participants were considered sufficient to: (i) examine how disease adaptation information may affect individuals' perceptions of RA; and (ii) develop a questionnaire based on these results (Guest et al., 2006). The participants ranged in age from under 20 to over 50; seven were female and five were male; two individuals currently had arthritis; and six of the group had experience with illness (Table 7.1, page 157).

Ideally, responses from an equal number of male and female participants would have been obtained; however, the decision was made to conduct interviews with those individuals who were most willing to participate in order to ease the recruitment process and to obtain data in a timely manner. If I had felt that theme saturation – the point at which an investigator has gained sufficient data to feel confident that an understanding of the phenomenon has been achieved (Denzin and Lincoln, 2000) – had not been obtained with the 12 initial interviews, then I would have conducted more interviews, while ensuring an equal gender distribution amongst the respondents.

7.2.2 Interview Process and Questions

I conducted face-to-face semi-structured interviews with respondents from the general population. The interview process incorporated various valuation exercises and discussions (Figure 6.1, page 131). The study participants were randomly sorted into two groups: the 'Initially Uninformed Group' and the 'Informed Group'. The purpose of having two groups was to alleviate the potential for interviewer effects, as previously discussed in more detail in Section 6.1. The use of two groups is of greater importance for the final quantitative component of this study, as will be illustrated further in Chapter Eight. However, to pilot-test the key features of the study design, the two groups were retained. The difference between the two groups was that individuals in the Initially Uninformed Group undertook the adaptation exercise between the first and the second valuation exercises. For individuals in the Informed Group, the adaptation exercise occurred before the first valuation; a second valuation was elicited after new information – patient values for three health states of similar RA severity – was presented.

Participants in the Initially Uninformed Group first valued a series of health states, including the three RA states of different severities using a visual analogue scale (VAS) and a self-completed bottom-up titration time trade-off (TTO) exercise (Gudex, 1994). A thorough description of the methods used in, and the ordering of, the valuation tasks are presented in Section 6.2. While the participants completed valuation exercises, their health state values are not presented in this chapter. The main reason for this decision is that the values themselves do not provide any additional insight into understanding the influence disease adaptation information may have on the general population's perceptions of RA. Furthermore, analyzing the results from the purposively sampled participants in this study would not lead to statistically reliable results. However, it was important that the participants underwent the valuation exercises, to encourage them to reflect upon the impact of living in a given health state and to pilot-test the key features prior to the large-scale quantitative study (to be described in Chapter Eight).

For individuals in the Initially Uninformed Group, the first valuation was followed by the adaptation exercise. In this, I first asked the participants if they knew what the common symptoms of arthritis were and whether they knew someone who suffered from arthritis[15]. They were then presented with information pertaining to patients living with RA obtained from the *Health Talk Online* website (DIPEx Health Experiences Research Group, 2008). These were in the form of three audio-recordings of interviews from RA patients – hereafter referred to as recorded scenarios. The interview excerpts highlighted different aspects of adaptation: 'Lisa' described the trials of initially being diagnosed with RA; 'Ann' discussed changes she made to accommodate her RA; and 'Patricia' reflected on her process of adapting to RA (the transcripts of the recorded scenarios can be found in Section 6.3).

After playing the first of the recorded scenarios, I asked a series of open questions: (i) can you summarize the information you just heard and (ii) have you, or someone close to you, been in a situation similar to what the patient described; if not, can you imagine yourself acting in the same manner as the patient? I used probing questions to encourage participants to elaborate on their responses. I then asked them to reflect upon, debate, and discuss this new information (the topic guide of the adaptation exercise is presented in Appendix A.5, page 268). The same questions were repeated after the participant heard each of the remaining two recorded scenarios. After the adaptation exercise, the participants in the Initially Uninformed Group repeated the same valuation tasks that I had given to them at the start of the interview.

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[15] As noted in Chapter Four, during discussion with the participants, the disease under investigation was generally referred to as 'arthritis', rather than as rheumatoid arthritis. This attempted to avoid cognitively overburdening the participants with the definitions of the various forms of arthritis. I had clinical definitions of both rheumatoid arthritis and osteoarthritis readily accessible if the participants questioned the distinction between the two terms; however, these were not used in any of the interviews. As such, the terms 'arthritis' and 'RA' are used interchangeably throughout this chapter to reflect the language used with, and by, the participants.

Participants in the Informed Group, on the other hand, underwent the adaptation exercise *before* valuing the health states by VAS and TTO (as shown in Figure 6.1, page 131). First, they listened to the patients' recorded scenarios and answered the open questions, as outlined above. After the adaptation exercise and the first valuation, participants in this group were presented with patient values for the health states they had just valued (see Section 6.4 for details of the patient values presentation). In addition to viewing patient values, they were also shown their personal TTO values for the RA states they had valued. These Informed Group participants were then asked to reflect upon the differences, if any, between their values and those of the patients'. After the patient values presentation, the individuals were then asked to repeat the initial valuations by VAS and TTO.

At the close of the session (i.e., after completing the second valuation), personal health state values over the course of the interview, along with the patient values, were presented to *both* the Initially Uninformed and Informed Groups. All participants were asked to discuss why their valuations changed or did not change between the first and second valuations.

All the face-to-face interviews were conducted in a meeting room in the University of Sheffield. Probes, or follow-up questions, were also used to allow me to respond to unanticipated comments made by the participants; hence, not every interview was conducted with exactly the same questions. On average, the entire interview session – including the valuation tasks – took about 80 minutes; the longest lasted approximately 110 minutes. Digital audio-recordings of all the interviews were collected. At the end of each interview, I recorded field notes in a diary; contents included suggestions to improve my interviewing abilities (e.g., 'allow more time so the participant can answer the questions properly' and 'you can give signs of encouragement to the participant without speaking') and any recurrent topics brought forth by the participant during the interview (e.g., 'the concept of pain was a big issue for him' and 'the presence of support was important to her').

The interviews were conducted during February and March, 2008. The University of Sheffield Ethics Committee approved the study protocol (copies of the ethics certificates are shown in Appendix A.8, page 299). All individuals were informed about the study prior to giving consent to participate (the consent form is presented in Appendix A.9, page 301).

7.2.3 Data Analysis

I transcribed the early interviews verbatim while recruitment for other study participants was still ongoing; this provided me with the opportunity to critique my interviewing abilities and my topic guide, and to improve upon them in subsequent interviews. In-depth analysis of the results occurred only after all 12 interviews were conducted. Each interviewee was then renamed according to the first letter of the participant's given name. Interview transcripts were prepared, with any identifiable characteristics mentioned during the interview (e.g., names of family members and doctors) removed. The transcripts were imported into NVivo (version 7.0), a computer-assisted, qualitative data analysis software package (QSR International, Doncaster, Australia). This program was used to manage the data and to help facilitate analysis.

Framework Analysis

I used the framework approach to analyze the qualitative interviews. Framework analysis is often used for applied or policy-relevant qualitative research (Ritchie and Spencer, 1994). The use of framework analysis as an approach to analyze the interviews allowed me, as a researcher, to address the aim of this study. This approach, in particular, enabled me to explore concepts which I felt were fundamental to the research question, and concurrently facilitated the emergence of other themes from the interviews. This approach is used in a manner that allows the coding framework to materialize from the data, as well as being informed by existing scientific knowledge of the issue under study. The five key stages that were applied to the interviews are:

1. Familiarizing
2. Identifying a thematic framework
3. Indexing
4. Charting
5. Mapping and interpreting

Each of these stages is described below.

Stage 1: Familiarizing

For the first stage of analyzing the qualitative data, I listened to the interviews several times during the transcription process. After completing the interview transcriptions, I listened to them all again to check the accuracy of the transcripts. I believed that I achieved full immersion of the data, as I was able to relate the participants' voices to the specific interview transcripts I was reading. I then read each transcript in its entirety several times and noted any recurrent themes that would aid in explaining how disease adaptation information affected the general population's perception of RA. In addition, I summarized each interview; this enabled me to keep the interviews in case formats, as well as gain important information without referring to the entire interview during the later steps of the framework analysis.

Stage 2: Identifying a Thematic Framework

It is usual in framework analysis to identify themes and sub-themes. However, since I had previously studied the constant comparison method (Strauss and Corbin, 1998), which is another qualitative approach to exploring and identifying issues that may arise from a research question, I incorporated this method in identifying the thematic framework. Using constant comparison, I summarized each line of the transcript in an attempt to gain a deeper understanding of the interviews. I then clustered similar summaries together to form sub-themes. The sub-themes were then organized into higher-order, main themes (e.g., the summaries 'the need to deny having arthritis' and 'do not want to burden others' belonged in the sub-theme of 'ability to hide vulnerability', which I subsequently incorporated into the main theme of 'attitudes toward coping'). From these different levels of themes, I produced flow diagrams in an attempt to understand any connections that might arise between themes.

During this process I also reviewed literature from the fields of health economics and health psychology, which were potentially relevant to my analyses in understanding how disease adaptation information affects individual's perceptions of RA; this step is common in framework analysis. By searching the literature, I discovered that components of the theory of planned behaviour provided an explanation for an individual's desire to change their attitudes towards aspects of health, such as attaining control of the illness and conducting behaviour in a particular way (Armitage and Conner, 2001). In addition, I had found that some study participants expressed greater understanding towards the patients in the adaptation exercise compared to others; as a result, I also investigated the literature about empathy (e.g., Davis, 1983).

All the information to be used in the identification of a thematic framework – the content of the interviews, the developed flow diagram, and the literature about the theory of planned behaviour and empathy – was discussed with Dr. Alicia

O'Cathain[16]. Due to my previous exposure with the constant comparison approach, I found myself breaking the data from the interviews into pieces that were, at times, too small for framework analysis. As a result, my interpretation of the interviews had focused on small details rather than on the global picture of the information vocalized by the participants. During this discussion, we identified some important concepts that have been overlooked in the initial review (e.g., personality characteristics of the participants). After reflection on our discussion and re-reading the transcripts, I developed a thematic framework based on the issues emerging from the transcripts and the information found in the literature.

Stage 3: Indexing

I indexed all transcripts using the codes from the constructed thematic framework. Coding is an umbrella term for the process of organizing qualitative data to assist in its retrieval and interpretation. This process highlights any complexities or contradictions arising from the transcripts. Not all text fragments had to be coded if they were not relevant to the research question. More than one code, if needed, could be applied to the same portion of the transcript. However, if any part of the transcript that was pertinent to the research question did not fit the framework, new codes were developed. These codes were incorporated into the existing framework and used in coding the remaining transcripts. When all the transcripts were coded, the framework was re-assessed, such that unassigned codes were eliminated and duplicate codes were combined into a single code (e.g., 'do not give in to the new change' and 'fight any presented change' were combined as the latter code). The new thematic framework (presented in Appendix A.10, page 304) was then used to re-code the interviews once again to ensure that the final codes were consistently applied to all the transcripts.

Stage 4: Charting

After applying the thematic framework to all the transcripts, the next step was to build up the picture of the whole dataset by considering the range of attitudes and experiences for each issue. The data was 'lifted' from their original context and rearranged according to the appropriate main themes identified from the thematic framework (Spencer and Ritchie, 1994).

In an attempt to develop an even greater understanding of the interviews, I created charts, which are essentially tables with headings and subheadings drawn from the

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[16] A member of the Ph.D. advisory panel, who specializes in mixed-methodology research.

thematic framework; doing this provided me with an overview of the data. Table 7.2 (page 158), for example, shows the perspectives of three participants in the 'attitudes toward coping' theme. By summarizing the content in each of the participant's interviews pertaining to the three sub-themes (i.e., 'ability to hide vulnerability', 'making lifestyle changes', and 'available support') of the main theme, I was able to compare the perspectives that existed within each participant as well as to observe the range of perspectives across participants.

Stage 5: Mapping and Interpreting

Throughout the data analysis process, I was continuously developing and revising my flow diagrams. This both aided my understanding of the complex data and provided a mechanism to clearly present my research findings to my advisory panel.

To aid in the interpretation of the research findings, I use quotes from the participants as an illustration in this thesis; this also allows the reader to more readily evaluate the conclusions I have drawn from the data (Spencer and Ritchie, 1994). The quotes have been presented for the most part verbatim in an attempt to not alter the 'flavour' of the language used by the participants themselves. However, interjections (e.g., "ah", "uh", "like", and "you know") were removed and, where necessary, words were inserted, marked off by square brackets, to provide a greater comprehension of what was being said during the interviews (e.g., "take their mind off it" became "take their mind off [the arthritis]"). The quotes have been kept as brief as possible, while highlighting the main points I wanted to address; in all cases, the context has been maintained.

In attempt to develop a greater understanding of the impact disease adaptation information may have on general population's perceptions of RA, I considered the participants' reflections about the recorded scenarios alongside their direct answers to the questions about changing values. If participants' comments regarding the recorded scenarios were not assessed, then their interpretations of how they felt about people with arthritis, illness itself, and general life outlook would be neglected. I expected that all of these factors would be influenced by the disease adaptation information presented to the participants.

7.3 Findings

From the interviews, I identified five themes relating to how disease adaptation information could influence members of the general population's perceptions of RA (Figure 7.1, page 163):

- Attitudes toward coping;
- Views on life with arthritis;
- Previous life events;
- Personalities; and
- A desire to live a long life.

Each of these themes is discussed further in the following section.

7.3.1 Attitudes Toward Coping

Upon reflecting on the information presented in the adaptation exercise, the participants began to realize that they could adjust to life with arthritis. Specifically, they started to understand that they could cope with an impaired health condition by making lifestyle changes to accommodate RA; by drawing on support from others to ease difficulty associated with RA; and by covering up any vulnerability related to having RA.

Making Lifestyle Changes

The second and third recorded scenarios of the adaptation exercise illustrated that a person living with arthritis can continue to live a fulfilling life. Upon hearing Ann's recorded scenario and how she kept herself physically fit by setting goals at the swimming pool, the participants expressed a range of positive emotions. These ranged from the opinion that what she has accomplished was "inspirational" and "impressive" to the fact that "she was going to fight [arthritis] all the way" (David, Informed Group). Because of Ann's message, participants began to recognize that a meaningful life and a reasonable level of fitness are achievable despite the impaired health state associated with arthritis.

The participants acknowledged that the first step is to "come to terms" with the fact that you have arthritis. While getting to this stage of acceptance "would take a while", it depends on the characteristic of the person "because some people do dwell on things and mope around, and some people just get on with it" (Christine, Informed Group). In addition, participants recognized that a more active process is involved in getting on with life once diagnosed with arthritis. Referred to as either adapting or coping, it is a stage of "necessity" because participants recognized that lifestyle changes need to be made in order to "accommodate" living with an illness.

> Because if you didn't [adapt to arthritis], you'd drive yourself mad with it, I think. For the sake of your own [...] mental health more than anything [...] you have to find some sort of way around it or you [would] just be miserable constantly, wouldn't you? (Simon, Initially Uninformed Group)

Participants with experience of chronic illnesses, compared to those who had minimal experience, seemed more open-minded about altering the activities that they currently enjoyed. They recognized that there is a need to know your "capabilities".

> If someone said to me I couldn't go [horse] riding, I'd be gutted. But 'aving said that, then I'd think, "well, I could go driving the 'orses, or something like that". So I would look for other options. I wouldn't stop everything. [...] I'd still go and mess with 'orses anyway and be around them. So I'd still have that contact, which that's what I'd like anyway. (Alice, Informed Group)

Alternatively, those with limited illness experience felt that making changes would be difficult and not being able to do the same activities as beforehand would be "frustrating".

> I would really struggle imagining making changes to life. I appreciate that I would probably have to but I find [making changes] really difficult to imagine. (Robert, Informed Group)

In addition to changing activities, participants recognized that, over time, a patient would begin to learn more about their illness. From Patricia's recorded scenario, participants began to understand that one could manage arthritis and educate oneself about pain relief. One participant, in particular, demonstrated a take-charge attitude to expedite the progress and illustrated a desire to obtain control of her health.

> I'll research the condition [as to] whether you could make dietary changes or [learn] what exercises is safe to do without over exerting your joints... (Rose, Informed Group)

Most of the study participants agreed that the adaptation exercise was "interesting" and "informative". The recorded scenarios further provided the participants with the evidence that, if patients can adapt to life with arthritis, then they should be able to cope with arthritis if they were placed in that situation.

> I think it was hearing those three people speak and realizing that it isn't actually the end of the world to be diagnosed [with arthritis]. [...] You can find ways of coping and be positive. [...] I guess I was

> thinking it was really bad to start with but then heard them: [arthritis] might not be that bad. (Karen, Initially Uninformed Group)

For those in the Informed Group, the presentation of the patient values, in addition to the recorded scenarios, provided them with concrete and conclusive evidence that an individual can cope with living with arthritis.

> When I looked at [the patient values], it made me think that I was really mardy[17] and being pathetic basically [...]. I was just thinking that I wanted to do everything and that if you can't do everything you want, your life ends. And looking at facts, [...] you do still have a life; so it made me think. [...] I could still have this, I could still have that. Am I really prepared just 'cuz of my discomfort – extreme pain, or whatever – prepared to give up? (Alice, Informed Group)

Available Support

Another factor that affected whether the participants believed that they could cope with living with arthritis was their personal attitudes towards available support, whether provided by family, friends, or their own spirituality. This provided some participants, after a period of reflection and deliberation, with a sound reason to alter their initial perceptions of RA because of the belief that the support they had available could help them adapt more quickly to the illness. For example, participants who were close to their families would welcome the help they offered whereas others expressing a more independent approach to life were too "proud" to acknowledge the need for support and preferred to deal with things on their own. The latter view was adopted most often by some individuals in an attempt not to "worry others" in what they regarded as their "own battle to fight".

> I believe we come into this life on our own; we leave this life again on our own. We're very alone at those two critical times [...] and you have to learn to deal with that situation yourself [and ...] the support of family, friends, or your loved ones will just need to be given a rest. You can't rely on other people to see you through that pain. You need to interpret that pain yourself. [...] It's important to have to deal with these things on your own, I believe. That way, if you become too reliant on other people, you could become a burden to family [and] friends. It doesn't help your situation [... and] you have two lots of people suffering. (Doug, Initially Uninformed Group)

The recorded scenario provided by Ann in the adaptation exercise mentioned that she had a network of friends – both sufferers and non-sufferers of arthritis – at the swimming pool. The participants recognized that having a "wide circle of friends"

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[17] The definition of 'mardy' is 'to be moody' (Bartsch-Parker, E., Burgen, S., Crowe, R., Maolalaigh, R.O., and Watt, D. (1999). *British Phrasebook*. Australia: Lonely Planet Publications.)

would allow them to cope better. Specifically, one can “take their mind off [the arthritis]” (Alice, Informed Group) yet, at the same time, receive support from interacting with other people “in the same position” (Christine, Informed Group).

Another aspect that contributed to the participants’ attitudes towards the influence that available support might have on their ability to cope with an illness was their spiritual beliefs. One participant, when she was dealing with her health problems, “held on to the hope” (Sophie, Initially Uninformed Group) until she gained a full recovery. For another individual, “faith [played] a big part” in how she has coped with her health issues.

> In the early days we prayed about this and believed that God could heal and I believe that [my arthritis has] not progressed as quick, as I thought it might do. [...] I’ll be going to heaven and there will be no pain. (Joanne, Informed Group)

Ability to Hide Vulnerability

The first of the recorded scenarios in the adaptation exercise is that of Lisa, a recently diagnosed 30-year old RA patient. In this recorded scenario, she highlights the potential to hide arthritic symptoms from other people. After listening to Lisa, some study participants believed that, if they had arthritis they might not want to fully disclose everything about their condition to others. In fact, they recognized that this was a form of dealing with a chronic health condition, albeit a non-laudable coping mechanism. The participants believed that not vocalizing the “full extent” of the condition to people is a fact of “human nature”, such that other “people don’t want to see you miserable even if you are in pain” (Alice, Informed Group). By “masking” the disability or “vulnerab[ility]”, participants, who indicated that they would cover up their symptoms, chose to do so in an attempt to “be seen as normal – shall we say, in inverted commas – than having some kind of condition” (Simon, Initially Uninformed Group).

Interestingly, participants who specified a preference to cover up their symptoms were individuals with no direct experience with arthritis. Patients currently living with arthritis, on the other hand, did not hold this perception; instead, they viewed the need to appear normal as a result of how society perceives individuals with illnesses.

> [If you have arthritis] you don’t want people to think you’re weak or think anything’s wrong with you so you can appear normal to everybody else 'cuz you think people might think less of you. I think it’s nonsense [...] [You think] you might not be accepted in society

> because society doesn't accept something [...] out of the ordinary, away from the normal. Disability can carry that stigma, even though it's not your fault. (Clare, Initially Uninformed Group)

7.3.2 Views of Life with Arthritis

When the participants reflected on the content of the adaptation exercise, it helped them to explore their initial views of arthritis. Not surprisingly, these appeared to depend on their personal experience of arthritis. The interviews revealed that those study participants without arthritis experience were more receptive to the disease adaptation information than those who did have this experience.

Experience with Arthritis

When I asked the participants to describe the common symptoms of arthritis, many of the participants expressed pre-conceived ideas of what it would be like to live with this chronic condition. These impressions stemmed directly from interactions with patients with arthritis or indirectly from stereotypes of the disease; the latter may possibly have arisen from media portrayals. The level of arthritis experience affected the attitude individuals had towards the health condition. Those participants with first-hand experience – either themselves or someone close to them – believed that there are "worse conditions to have" than arthritis. However, due to the potential struggles patients may face with their arthritis, *inexperienced* participants envisioned an extreme form of the condition during their initial valuation of the RA states. Because of this idea, arthritis was described as "depressing" and "unfair". In this context, these individuals, compared to those with arthritic experience, expressed a lower preference to live in any arthritic health state before the adaptation exercise was administered.

> [Arthritis] looks quite stressful and frustrating. And you can see if they're [people with arthritis] walking – obviously they need walking aids – and they'll happily do it for a couple of minutes and you can tell they get tired and a little bit sore and it just progresses like that. It's not something I would like to have, to be honest. It seems it just one of those illnesses that seem to be really unfair. [...] Even though you're mentally fit and healthy, your body is just really struggling to keep up [with the arthritis] and move with your mental state, which, I think, is really frustrating for anyone. (Sophie, Initially Uninformed Group)

Of the 12 participants, two individuals had arthritis: Clare had osteoarthritis and Joanne had RA. Half of the participants had varied levels of arthritis experience with family members or friends. This resulted in some individuals coming into the study more informed about arthritis than others.

> [With arthritis], you go, "I'm as fit as a fiddle but I have arthritis". You don't class as an illness as such [...] that's how [...] me Dad [with arthritis] classed it, "ah, I'm fit. I've never been poorly in me life". (Alice, Informed Group)

An Older Person's Disease

The participants expressed negative impressions when the age of the patient with arthritis was considered. Upon hearing the first recorded scenario of the adaptation exercise, most participants had trouble believing that Lisa had already been diagnosed with arthritis at the age of 30; in fact, one participant questioned whether the recorded scenario was indeed "real". Most thought that arthritis only afflicted older people and considered the patient must have been "unlucky" if arthritis developed at an early age.

> I don't know how prevalent arthritis is at an early age, but if you're in your thirties, is it some kind of failing that you've got arthritis? And is there the support there for younger people? [...] I don't know. I've only really associated it with the elderly. (Rose, Informed Group)

Since most participants associated older patients with having arthritis, they felt that older patients were better suited to cope than younger patients because they were in the appropriate "age category" to have arthritis.

> Like in your thirties, you're definitely not meant to be getting any illnesses so any illness is a real shock [...] because it's not fair on a 30-year old. At 50 [... you're] used to be caring for people [...] and may have experience with people getting illness at that age, like breast cancer and stuff. So it's more of that generation that would be thinking about positive thinking: just trying to be practical and get through it for the sake of a kid. (Karen, Initially Uninformed Group)

However, after being informed through the adaptation exercise that arthritis could occur at any age, individuals began to understand more about the disease.

7.3.3 Previous Life Events

Reflecting on the information in the adaptation exercise enabled the participants to refer back to previous events in their own lives. In particular, previous direct or indirect experience with illnesses had a significant bearing on their attitudes toward the presented RA health states.

Experience with Illness

Aside from specific experience with arthritis, the participants' personal experiences with chronic illnesses appeared to enable them to understand the messages behind the recorded scenarios in the adaptation exercise better than participants without experience with illness. One participant, in particular, was able to juxtapose the content in Ann's interview to his own personal health battle.

> [Ann] was going to fight [arthritis] all the way. It's not letting it beat her. [Because of my celiac disease, I was told to] either [...] get rid of your bikes or well, sit in the house and feel sorry for me self, or you're going to try coping and keep going on bike. [...] I try to ride for as long as I can now so [my stomach] don't hurt. So I'm not letting [the celiac disease] beat me. (David, Informed Group)

In contrast, the current wellness status of healthy participants may have prevented them from fully grasping the concept of living with arthritis. This, in turn, may have prohibited them from completely understanding the message brought forth by the patients in the adaptation exercise.

> [When] you're going at the questionnaire, you're looking at from your own perspective, I guess. And from my own specific perspective, I'm in quite good health [...] So if you offer me their scores [patient values] without the questionnaire – I don't know. I think I'd still, at this stage, take my own scores because I don't have to cope with the condition. (Simon, Initially Uninformed Group)

In addition, the adaptation exercise prompted participants to recall illness events amongst family and friends. For example, one participant described how her father's fight to live longer despite being given only two days to live made her realize that there is more to life than full health. However, the influence of illness experience had the opposite effect on other individuals who associated severe illness with very poor quality of life.

> I saw my mother die of a stroke. [...] But she died really because she didn't want to live. She didn't want to live that type of life where she couldn't do anything for herself. Extreme pain has that affect on you; the only way it can affect. [...] You can forget moderate, slight pain but extreme pain, I don't think, you could forget about it. [...] My personal experience is extreme pain makes life not worth living. And if life isn't worth living, what is the point of it? Yeah, I'm not one of these people who keep people living forever in extreme pain. I mean, to me, that's just cruel. If people want to die let them die. (Clare, Initially Uninformed Group)

Not as Painful as First Perceived

All the participants discussed the concept of pain. Those with minimal or no experience with arthritis – or with any illness for that matter – described the assumed pain as "horrible", "constant", and "difficult to accept". Because of these negative opinions, participants considered arthritis to be a severe health condition that they would fear to have.

> I'm not good with dealing with pain and I value life the way it is quite a lot. I don't think I would value [life] nearly as much if I was in a painful discomfort. (Robert, Informed Group)

In addition, a number of participants, when making their assessments, focused on the label, 'extreme pain or discomfort', of the severe health state description. The connotation that arises from the word 'extreme' made many people apprehensive of living in that health state for a long period of time.

> I couldn't be able to live with that [...] The word, I think, extreme pain just seems to me that you notice it all the time. (Darryl, Initially Uninformed Group)

After hearing the recorded scenarios, the participants gained further insights about what it is like to live with arthritis. At the start of the session, participants thought that the pain experienced when living with arthritis would be quite severe but after the adaptation exercise, participants began to realize that mild and moderate pain would not be as horrible as they initially thought.

> I think listening to the recordings, it made me realize that there was – and actually talking [it] through myself, as well – there was some life with moderate pain. With extreme pain, I would say, there is very little life. If you just have moderate pain, you can get on with your life. There is a life to live. (Clare, Initially Uninformed Group)

7.3.4 Personalities

From their interviews, aspects of the participants' personalities were revealed which, in turn, appeared to affect their reactions to the messages highlighted in the adaptation exercise. This theme highlights how the participants talked throughout their interviews. While they did not overtly state their personality traits, it was apparent that their outlook on life played a major role in how they would face a significant change to their lives. For example, some individuals answered the open questions with upbeat personalities, while others used a lot of negative language.

Attitudes toward a Life Change

I observed a range of different approaches in how the participants would deal with the news that they had arthritis. Participants indicated that they would feel "angry" initially because they would not be able immediately to come to terms with this life-changing event. However, individuals with positive attitudes recognized that there is a need to "investigate" what changes could be made to improve their life.

Furthermore, they were more likely to accept the fact that, while they might not be able to do the same things as they did before they developed arthritis, they needed to make the best of the situation that they were given.

> I can relate to [Ann's interview] because sometimes I think, "I can't achieve this particular thing". [...] I can always surprise myself at the end of day thinking that, "I have achieved and I've done it with ease and I've actually enjoyed the experience". So what it proves you can never dismiss any situation, [no matter] how unlikely it may look at the time. (Doug, Initially Uninformed Group)

Individuals with negative outlooks on life felt that it would be a "struggle" and a "challenge" to find a substitute for an activity that they currently loved. In cases where moderation would enable an individual still to partake in activities that they enjoyed, one individual, in particular, recognized that this modification would, in theory, work for some people but not for him.

> I think it would be difficult for me personally. [...] I think I would probably over do [things] more [...] I [would] find it difficult, I think, to moderate. (Simon, Initially Uninformed Group)

Empathetic People

It was evident that some participants were able to empathize with the patients in the recorded scenarios more than others. This was not simply related to attitudes to coping or past life events as discussed earlier. These individuals seemed more able to put themselves into the position of the patients after hearing the recorded scenarios.

> [The information presented in the audio-recordings has] broaden my mind. I think 'cuz [arthritis is] something that didn't affect me directly. [...] After listening to the [patients ...] my outlook kind of changed. Though [arthritis] is a severe illness, it is not the end of the world. Basically, there are still plenty of opportunities for you still and there's numerous things you can do. And, yeah, basically, I think I was naïve to it and thought that it was a more horrific than it was. I mean, I'm not saying that I'm not discounting anyone at all. I know it's a serious illness but there are plenty of things there are still available for you to do. (Sophie, Initially Uninformed Group)

The degree to which the participants empathized with the individuals in the recorded scenarios was further demonstrated when the participants consistently used the fictional names of the patients throughout their responses in the interviews. This indicated that these participants were engaged with the entire adaptation exercise and, therefore, may have been more influenced by the patients' messages.

> Lisa were more in the fed up stage and angry stage, which you're going to be at that age, I think. [...] I should imagine the longer you

> have it the more you learn to manage it and know what gives you a bit relief. [...] I thought Lisa was quite sad and then Patricia and Ann were like, "it's not the end of your life. It's not the end of your world. [...] You 'ave to put up with it. You can have a quality of life and things". [...] So that's what I sort of got from them. (Alice, Informed Group)

7.3.5 Desire to Live a Long Life

Reflecting on the adaptation exercise helped participants recognize that, even if living with a disability, the number of life years was very important in their assessments. Specifically, reflecting on their personal health state values and comparing them with those of the patients' affected the opinions of some of the individuals in the Informed Group because they realized they had a desire for a greater number of life years. This was a result of wanting to achieve certain milestones in life, such as "want[ing] to see everybody and [seeing my children] grow up" (David, Informed Group). Some individuals realized that they would be giving up so much of something they really wanted and, therefore, accepted that they could live with a poorer quality of life in return for a longer life.

> [Arthritis] is obviously manageable, if you like; you can put up with it. [...] [Giving up] 18 years [of my life to not have arthritis]: I must 'ave had a mental block. (Alice, Informed Group)

7.4 Reasons to Change Questionnaire

The objective of the qualitative interviews was to understand how disease adaptation information might affect the general population's perceptions of RA (Figure 7.1, page 163). While the study participants did not explicitly state that, for example, they opted to increase their health state values because they empathized with the patients' messages after hearing the recorded scenarios in the adaptation exercise, I had to extrapolate what they were saying to how it might affect their values for RA states. These hypotheses provided me with an outline of how the survey, the Reasons to Change Questionnaire (RCQ), was to be designed. This questionnaire was developed as a tool to obtain in-depth information in a quantitative interview environment (to be described in Chapter Eight) where participant numbers were too large to allow individual interview transcription and analysis. The intention of the RCQ was to provide greater insight into the factors that contribute to participants' changing their health state values after being provided with disease adaptation information. The results from administering this questionnaire are discussed in Chapter Eight.

7.4.1 Instrument Development

The items in the RCQ are presented in Table 7.3 (page 159); the actual questionnaire is displayed in Appendix A.11 (page 306). Each item was constructed to describe important aspects of each sub-theme identified from the framework analysis in the qualitative interviews (described in Section 7.3). Where possible, the description of the items was derived using the language of the participants (e.g., item 1.f – 'taking part in today's session has helped me to realize that arthritis is not "just a part of getting old"').

Two lists, one of 25 and one of 26 items, were drawn up to represent the sub-themes to be addressed. The extra item (item 2.c) – 'my opinions about arthritis changed after seeing the patient scores' – was intended only for those individuals in the Informed Group because they had the opportunity to view the patient values. The items were clustered into three groups: (1) values affected by the disease adaptation information (items 1.a to 1.m); (2) values affected by how the disease adaptation information was presented (i.e., the recorded scenarios, the interviewer, the patient values presentation) (items 2.a to 2.b for the Initially Uninformed Group and items 2.a to 2.c for the Informed Group); and (3) values not affected by the disease adaptation information (items 3.a to 3.j). Items belonging in the same sub-themes were clustered together; this was to ensure ease of comprehension for the respondents when completing the questionnaire.

Three items in the questionnaire – items 3.h, 3.i, and 3.j – assessed the individual's understanding of the valuation task. The inclusion of these items – at least the ones pertaining to the understanding of the valuation exercise – was due to one individual in the qualitative study (Robert), who did not grasp the concepts of the health state values when I provided an explanation for his values for the three arthritic states, relative to what the patients had provided. Although he mentioned that he would change his values to match the patient values, he, in fact, changed his second TTO values in the negative direction because he thought that the patients perceived their lives with arthritis to be much worse than he did. When he realized the error, he stated that knowing that the patients thought that having arthritis was "more bearable than I had, [... my] results would be reasonably similar the second time" (Robert, Informed Group).

Questions pertaining to an individual's experience with arthritis were not included in the RCQ. Instead, this information was captured in the section describing the respondent's demographic characteristics in the valuation booklet (shown in Appendix A.4, page 247). The primary reason for not including this sub-theme in the RCQ was that more specific information could be attained through the demographics section. In that section, individuals were asked it they currently have arthritis and, if they did, the type of arthritis it was (e.g., RA, osteoarthritis, psoriasis, etc.). They were also asked if they knew anyone that suffers from arthritis and, if they did, what their relationship was with this person.

A number of response sets were considered for the RCQ. Initially, a binary option – 'yes' or 'no' – was considered for the sake of designing a simple questionnaire and to avoid overburdening the respondents, who also had to complete the series of valuation exercises and undergo the adaptation exercise in the large-scale study. However, with the binary option, the intensity of the individual's responses would not be assessable. Therefore, the item responses were measured using a five-point response scale, which ranged from 'strongly agree' through 'neither agree nor disagree' to 'strongly disagree'. The decision to use a five-point scale was to provide enough choices to the participants, without overburdening them, yet ensure sensitivity to differentiate between the individuals' responses when used in statistical analysis of the subsequent large-scale study (Chapter Eight).

7.4.2 Hypotheses

For each item on the RCQ, an *a priori* hypothesis was derived based on the findings from the qualitative interviews; these hypotheses are presented in the right-hand column of Table 7.3 (page 159). These hypotheses enabled me to consider whether or not the participant's response to each item was expected to yield a change – and the direction of this change – in the health state values. For example, if the participant agreed with the fourth RCQ item (i.e., item 1.d), 'taking part in today's session has helped me to realize that you still could have a good quality of life when living with arthritis', a change in health state values in the positive direction would be expected in the subsequent quantitative study. It was expected that being informed about disease adaptation would encourage participants to consider that a meaningful life is still possible with RA. However, the first RCQ item (i.e., item 1.a), 'taking part in today's session has helped me to understand more about the disease of arthritis' may lead to different outcomes. It is not obvious how health state values may change because this depends on the level of knowledge the participant entered the study with. Specifically, individuals without any arthritis experience may be affected positively or negatively by the disease adaptation information, whereas individuals with arthritis were expected to not change their health state values because they were already informed.

7.5 Discussion

The aim of this phase of the thesis was to gain a greater understanding of the effect disease adaptation information may have on members of the general population's perceptions of RA. Based on results from the qualitative interviews, the adaptation exercise and the patient values presentation were observed to influence the individual's: (i) coping attitudes; (ii) view on life with arthritis; (iii) previous life events; (iv) personality traits; and (v) desire to live a long life. These five themes, although described as distinct entities, are, in fact, highly interrelated. For example, an individual's coping attitudes may be affected by past life events, which, in turn, may be affected by the individual's personality traits. The themes, and their corresponding sub-themes, were incorporated into the RCQ, an instrument designed to evaluate individuals' reasons for changing – or, conversely, not changing – their health state valuations after being provided with disease adaptation information. This questionnaire was to be administered in the large-scale valuation study described in Chapter Eight.

The results from this qualitative component demonstrated that the use of recorded scenarios, a reflection and deliberation period, and a patient values presentation were well received by respondents from the general population. Respondents appeared to engage with the messages in the recorded scenarios and seriously and conscientiously considered the open questions of the adaptation exercise, and the valuation exercises. The use of the adaptation exercise appeared to be effective at informing participants about the possibility of adapting to life with RA, as some of them reconsidered their initial perceptions. The quantitative impact of the disease adaptation information on the individuals' values cannot be determined in this qualitative study. These values are assessed on a larger, statistically meaningful, sample in the valuation study reported in Chapter Eight.

The present study was not the first to conduct an adaptation exercise (for a discussion of previous adaptation exercises, refer to Section 2.3.1); at least three studies have incorporated adaptation exercises to aid the general population in their valuation of hypothetical health states, including Ubel et al. (2005) and Damschroder et al. (2005, 2008). The current work was the first to utilize recorded scenarios from real patients to promote concepts of disease adaptation and to assess the influence of this. Previous adaptation exercises adopted introspective methods by encouraging respondents to think back to previous challenging life events and to consider how their emotions towards that particular event lessened over time. The current adaptation exercise used a combination of both extrospective and introspective approaches: the general population respondents recognized that patients do adapt to a specific chronic illness (i.e., RA) over time and translated this information into their own health state valuations if they wished to do so.

While the intent of the recorded scenarios for the current study was primarily to inform the participants about the eventual possibility of adapting to a specific health condition, it also served to personalize the health states. Giving everyday names to the patients in the recorded scenarios appear to have encouraged some participants to empathize with the patients whose information was contained in the adaptation exercise. Empathy is broadly defined as the ability of an individual to react to the experiences of another individual (Davis, 1983). More specifically, empathy is also referred to as "the capacity for knowledge of the emotional state of another person, regardless of one's own emotional state" (Girón and Gómez-Beneyto, 1998). What emerged from the interviews was that the individuals' degree of empathy seemed to be related to their personality traits and their experiences with illness.

The theme representing individual's personalities aligned with the components of the theory of planned behaviour. This theory, in general, describes the principal determinants of an individual's behaviour (Ajzen, 1988). Individuals tend to engage in behaviours that are evaluated positively, since social pressure encourages them to perform in that manner, and in behaviours that are perceived to be relatively easy to perform. The attributes of the theory of planned behaviour supported the information contained in some of the interviews, such as the individual's need to achieve a level of perceived control of the illness (i.e., make changes to adapt to an illness) and the need to conduct behaviour in a particular way (i.e., the desire to be viewed as normal and without a disability).

The results from this current study also support the claim that to get full benefit from using an adaptation exercise it may be necessary to provide a reflection and discussion period. Providing the participants with the opportunity to think and talk about the information presented in the adaptation exercise allowed them to clarify their preferences before valuing the RA states; this coincides with the way individuals formulate decisions in reality. As discussed previously in Section 2.3.1, this approach has been evaluated empirically by several studies (Dolan et al., 1999;

Stein et al., 2006) but the results to date have been inconclusive. It is speculated that if I had presented the three recorded scenarios, without time for deliberation, the themes identified to explain the impact of disease adaptation information on RA perceptions would differ from those identified in the present study. Without deliberation, I believe that the participants would not have had the opportunity to reflect upon the new information and hence would have had more difficulty incorporating that information into the subsequent valuations of the impaired health states.

7.5.1 Study Strengths and Limitations

The results from this study contribute to the current literature about the construction of informed general population values. However, the use of interviews to collect data placed some limitations on this study: in particular, responses represented the opinions of the participants *at the time* of interview only (Denzin and Lincoln, 2000).

The use of patient recorded scenarios was a novel technique in communicating the concept of disease adaptation. However, the order in which recorded scenarios were presented may have affected the participants' impressions of RA. The results may have been biased when starting with a negative recorded scenario (i.e., Lisa's interview) and then improving the messages in each of the subsequent recorded scenarios (i.e., Ann and Patricia's interviews). This potentially may have encouraged the participants to improve their initial perceptions of health states.

The sampling method may have also affected the study results. By recruiting participants in the city centre of Sheffield, the research may not be transferable to the general population in terms of health status. For example, individuals in ill health would be less likely to walk along the High Street compared to more healthy individuals. Furthermore, by depending on individuals who agreed to participate, the results may be subject to volunteer bias. Therefore, the obtained study sample may not be transferable to the full range of the general population.

7.5.2 Reflexivity

Reflexivity is an important process to ensure the quality of qualitative research (Denzin and Lincoln, 2000). This study may have been limited by my developing experience as a qualitative researcher. My still-developing skills as an interviewer were revealed in the first few interviews through occasionally awkwardly phrased questions, unnecessarily rigid adherence to the interview topic guide, and sporadically missed opportunities to follow up on interesting points. As the study progressed, I improved in these areas and obtained quality data to assist in the development of a questionnaire used to evaluate the rationale for an individual to change their values for the RA states after being informed about disease adaptation.

Qualitative researchers also need to acknowledge their bias toward the research topic to ensure transparency in how the conclusions were drawn. Denzin and Lincoln (2000) explain, "all research is interpretive; it is guided by a set of beliefs and feelings about the world and how it should be understood and studied". Although I attempted to be unbiased, I may have had an underlying desire for the study participants to change their values, especially for the first few interviews. This may have impacted on how I asked the questions in the first interviews. However, since I transcribed the interviews while recruitment for study participants was ongoing, I was able to modify my delivery of the interview questions in cases where I detected an apparent bias. Even when I displayed any desire for wanting the participants to change their health state values, this subsided once the interviews were underway; I became engrossed with what the participants said in the interviews and wanted to determine the answer to my research question regardless of whether the results were positive or negative.

7.6 Summary and Conclusions

This study provided an understanding of the effect disease adaptation information had on members of the general population's perceptions of RA. Based on the results from the qualitative interviews, the effect of disease adaptation information on the individual's perceptions of RA was influenced by their: (i) attitudes toward coping; (ii) views about life with arthritis; (iii) previous illness experiences; (iv) personality traits; and (v) desire to live a long life.

The results from this qualitative component were incorporated into the final quantitative component, described in Chapter Eight, in the form of a questionnaire. The RCQ was specifically designed to evaluate an individual's reasons for changing – or, conversely, not changing – their health state values. Administration of this questionnaire should enable a quantitative assessment of the factors that influence members of the general population to change their health states after being informed about disease adaptation.

7.7 Tables and Figures

Table 7.1: Characteristics of the study participants

Participant[a]	Gender	Age Group	Has Arthritis?	Has Illness Experience?
Initially Uninformed Group				
Clare	F	> 50	Yes	Yes
Darryl	M	20-29	No	No
Doug	M	> 50	No	Yes
Karen	F	20-29	No	No
Simon	M	30-39	No	No
Sophie	F	20-29	No	Yes
Informed Group				
Alice	F	40-49	No	Yes
Christine	F	20-29	No	No
David	M	40-49	No	Yes
Joanne	F	> 50	Yes	Yes
Robert	M	< 20	No	No
Rose	F	30-39	No	No

[a] Not participant's real name.

Table 7.2: Charting example for 'Attitudes Toward Coping' theme

Participant	Ability to Hide Vulnerability	Making Lifestyle Changes	Available Support
Alice	She's in denial about her knee pain. She doesn't want to burden others with her problems. She feels that covering feelings is a common thing humans do. Covering up is a sign that you're scared of the outcome.	She felt that even if you've accepted arthritis, you can still have bad days. She recognizes that adaptation and disease management takes awhile. She would be okay with making some changes in live (being with horses) and knowing the limitations of your body; however, she feels as if it would be unfair to do so.	She felt that a support of friends is very important, especially a need to be with non-sufferers so that you can forget that you have arthritis.
David	He has hid stomach pains (celiac disease) from others because it is his own battle to fight and not to appear weak. He felt that masking the true appearance is commonly done.	He recognized that you need to come to terms with your illness at first but disease adaptation takes a while. He knows that he can't dwell on his problem because it will get him stressed.	
Joanne	She related to Lisa, in that she suppressed pain and symptoms from others. She felt that covering up feelings is a common thing humans do as it is a sign that you're scared of the outcome	She felt that accepting that you have an illness is a big part	She recognizes that the need for friends as support is very important. Faith and prayers are a big part in her life.

Table 7.3: Components of the Reasons to Change Questionnaire

Theme	Sub-Theme	Individual Item (Variable name)	Item Number	Hypothesis
Views of Life with Arthritis	Experience with arthritis	Taking part in today's session has helped me to understand more about the disease of arthritis. (UNDARTHDIS)	1.a	If a person understands more about arthritis than before, he is more likely to change his values. Change can occur in either direction.
		Taking part in today's session has helped me to understand more about what it is like to live with arthritis. (UNDARTHLIFE)	1.b	If a person understands more about what is like to live with arthritis, he is more likely to change his values. Change can occur in either direction.
		Taking part in today's session has helped me to realize that I now know as much as patients do about what it is like to live with arthritis. (UNDARTHPAT)	1.c	If a person now realizes that he knows as patients do about living with arthritis, he is more likely to change his values. A positive change is expected.
		Taking part in today's session has helped me to realize that you still could have a good quality of life when living with arthritis. (GOODQOL)	1.d	If a person now realizes that a good quality of life is achievable with arthritis, he is more likely to change his values. A positive change is expected.
		Taking part in today's session has helped me to realize that there are worse diseases to have than arthritis. (WORSEDIS)	1.e	If a person now realizes that there are other diseases worse than arthritis, he is more likely to change his values. A positive change is expected.
	An older person's disease	Taking part in today's session has helped me to realize that arthritis is not "just a part of getting old". (NOTOLD)	1.f	If a person now realizes that arthritis can occur at any age, he is more likely to change his values. Change can occur in either direction.
Previous Life Events	Not as painful as first	Taking part in today's session has helped me	1.g	If a person now realizes that pain is

	perceived	to realize that living in pain is not always a horrible thing. (PAINOK)		not always a horrible thing, he is more likely to change his values. A positive change is expected.
A Desire for a Long Life		Taking part in today's session has helped me to realize that I would rather live longer with arthritis so I can spend more time with my family and friends. (FAMTIME)	1.h	If a person now realizes that spending more time with family and friends despite being in an impaired health state, he is more likely to change his values. A positive change is expected.
Attitudes toward Coping	Ability to hide vulnerability	Taking part in today's session has helped me to realize that that having arthritis does not have to make me look vulnerable or weak. (NOTWEAK)	1.i	If a person now realizes that having arthritis does not have to make him look weak, he is more likely to change his values. A positive change is expected.
		Taking part in today's session has helped me to realize that I can cover up the signs of arthritis to appear normal (COVERUP)	1.j	If a person now realizes that he is able to cover up his symptoms to appear normal, he is more likely to change his values. A positive change is expected.
	Available support	Taking part in today's session has helped me to realize that people can cope with having arthritis by themselves. (SELFCOPE)	1.k	If a person now realizes that he can cope with arthritis by himself, he is more likely to change his values. A positive change is expected.
		Taking part in today's session has helped me to realize that family and friends can help people cope with arthritis. (FAMCOPE)	1.l	If a person now realizes that he can cope with arthritis with support from family and friends, he is more likely to change his values. A positive change is expected.
	Making lifestyle changes	Taking part in today's session has helped me to realize that that I could cope with arthritis because patients cope with it. (PATCOPE)	1.m	If a person now realizes that he can cope because patients cope, he is more likely to change his values. A positive change is expected.

Pertaining to the Disease Adaptation Information		My opinions about arthritis changed after hearing the recordings. (RECORDING)	2.a	If a person's opinions about arthritis changed after hearing the recordings, he is more likely to change his values. A change can occur in either direction.
		My opinions about arthritis changed after talking to the interviewer. (TALKING)	2.b	If a person's opinions about arthritis changed after taking to the other group members, he is more likely to change his values. A change can occur in either direction.
		My opinions about arthritis changed after seeing the patient scores. (PATSCORE)	2.c[*]	If a person's opinions about arthritis changed after seeing the patient scores, he is more likely to change his values. A change can occur in either direction.
Personality	Empathetic people	In general, I feel that if I had to, I think that I can imagine living with arthritis for the rest of my life. (IMAGINARTH)	3.a	If a person can imagine living with arthritis for the rest of his life, he is more likely to change his values. A change in either direction is expected.
		In general, I feel that if I had to, I can "put myself in other people's shoes" and see things from their point of view. (OTHERSHOE)	3.b	If a person is able to put himself in other people's shoes and see things from their point of view, he is more likely to change his values. A positive change is expected.
		In general, I feel that it's unfair for me to value a patient's life because I don't really know what it's like to live with arthritis. (UNFAIR)	3.c	If a person thinks it's unfair for him to value a patient's life because he doesn't understand arthritis fully, a small amount of change or no change is expected.
	Attitudes to a	In general, I feel that I	3.d	If a person has a

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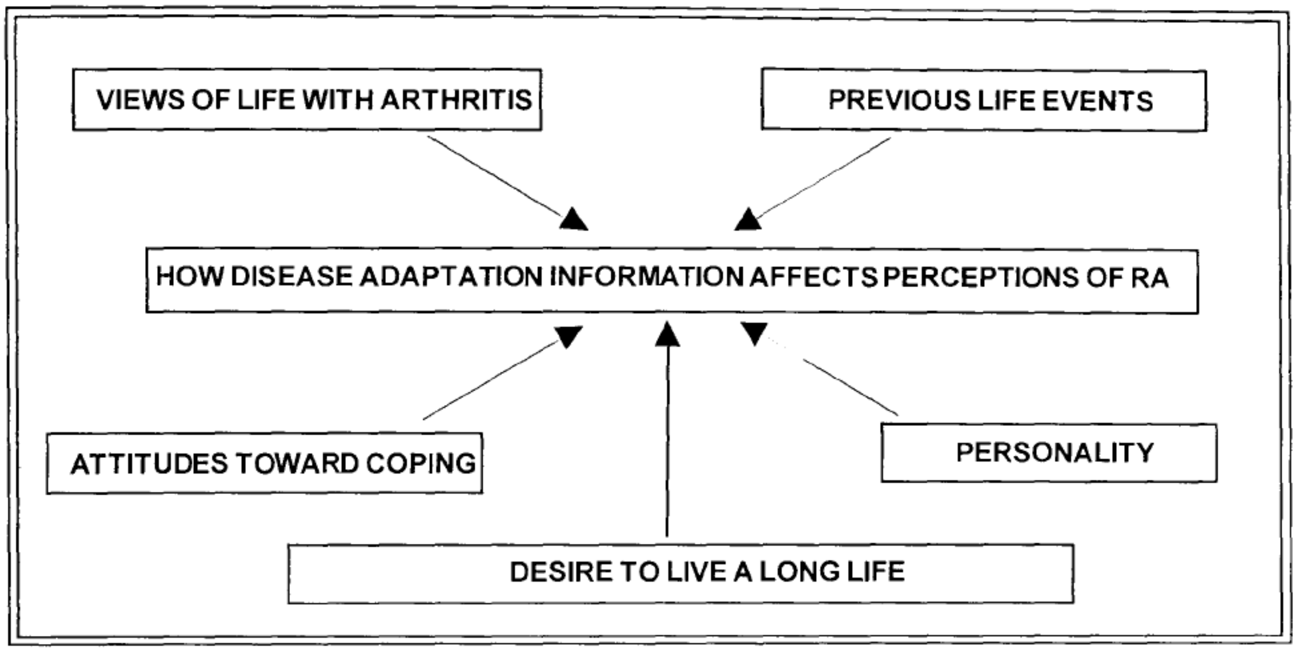
[*] Item 2.c is included for those individuals participating in the Informed Group; otherwise this item will be eliminated for the Initially Uninformed Group.

	life change	have a positive outlook on life. (POSOUTLOOK)		positive outlook, he is more likely to change his values. A positive change is expected.
		In general, I feel that I am the type of person that can adapt to change. (ADAPT)	3.e	If a person can adapt to change, he is more likely to change his values. A positive change is expected.
Previous life events	Experience with illness	In general, I feel that I personally know what it is like to have a health problem. (HLTHPRBSELF)	3.f	If a person has a health problem, he is more likely to change his values. A positive change is expected.
		In general, I know what it is like to have a problem through a family member or a close friend with a health problem. (HLTHPRBOTH)	3.g	If a person knows of someone with a health problem, he is more likely to change his values. A positive change is expected.
Increased understanding of the valuation task		In general, I feel that I understood the first valuation exercise. (FIRSTEXER)	3.h	This question is for researcher's interest.
		In general, I feel that I understood the second valuation exercise. (SECDEXER)	3.i	This question is for researcher's interest.
		In general, I feel that I had a difficult time deciding how long I wanted to live with arthritis on the valuation exercises. (TRADEOFFIDFF)	3.j	This question is for researcher's interest.

Figure 7.1: Themes that affect an individual's perceptions of RA after being informed about disease adaptation



Chapter Eight

Component Three: A Quantitative Assessment of the Influence of Disease Adaptation Information on Values for Rheumatoid Arthritis States

8.1 Introduction

The previous chapter used the results from qualitative interviews to demonstrate how the provision of disease adaptation information affected general population respondents' perceptions of rheumatoid arthritis (RA). However, due to the small number of individuals purposively sampled to participate, the health state values from the previous study were not statistically meaningful. As such, a large-scale follow-up study was conducted to assess quantitatively the magnitude of change in health state values after the respondents were informed about disease adaptation using the previously developed adaptation exercise.

This chapter reports the findings from the third, and final, component of the sequential mixed-methods study. It first establishes the extent to which general population respondents change their health state values after being informed about disease adaptation. The magnitude of change is tested by a series of statistical tests of association. Second, the factors which cause an individual to change their initial values are investigated. This second objective explores the rationales identified from the previous chapter using the Reasons to Change Questionnaire (RCQ). The items from this instrument were subjected to principal components analysis before being evaluated using regression techniques.

8.2 Study Design

This study follows the design described in greater detail in Section 6.1; the following summarizes it and identifies details specific to the quantitative component of this work. Respondents from the general population were recruited and randomized into two groups: an Initially Uninformed Group and an Informed Group (Figure 6.1, page 131). The main group used to measure the effect of the adaptation exercise was the Initially Uninformed Group. The Informed Group was created to identify potential interviewer effects, such as prevarication bias (Hiebert and Nordin, 2006) and interviewer bias (Last, 2001), thereby helping to identify the true effects of the adaptation exercise.

Individuals in the Initially Uninformed Group first valued the three RA states derived using Rasch and cluster analyses, as discussed in Chapter Five (descriptions of the health states are presented in Table 5.14, page 111), along with full health, current health, and dead states. This was followed by an adaptation exercise, where they listened to recorded scenarios (e.g., audio-recordings) of patients discussing adapting to life with RA, and then were encouraged to discuss and reflect upon the information in the recorded scenarios. After the adaptation exercise, they once again valued the same states as they had previously. Individuals in the Informed Group, on the other hand, underwent the adaptation exercise before valuing the health states for the first time. Then they were shown patient values for the states they had previously valued (i.e., the 'patient values presentation'), along with their personal values for the health states; the implications of these values were explained. After this presentation, they were asked to repeat the health state valuations. At the end of the session, both the Initially Uninformed and Informed Groups were given the RCQ to complete.

8.2.1 Objective One: To Determine the Effectiveness of Disease Adaptation Information in Altering Health State Values

The first objective of this study was to assess whether informing respondents about the possibility of adaptation was effective in changing their initial values for RA states. Four null hypotheses, described below, were explored (Table 8.1, page 200). The statistical methods used to test the hypotheses are fully described in Section 8.3.4.

Null Hypothesis One: The Adaptation Exercise Has No Effect in Altering Health State Values

Testing this null hypothesis establishes whether the adaptation exercise is ineffective in changing respondents' health state values. This was done using the responses provided by the Initially Uninformed Group only. Comparing their first and second valuations within the same health state evaluates the impact the adaptation exercise may have on the individuals' valuation of health states.

Null Hypothesis Two: The Patient Values Presentation Has No Effect in Altering Health State Values

Testing this null hypothesis establishes whether the patient values presentation is ineffective in changing respondents' health state values. This was done using the responses provided by the Informed Group only. Comparing their two valuations – one done before, and the other after their exposure to patient values – assesses whether there was no effect on the health state values after provision of the patient values presentation, when preceded by the adaptation exercise.

Null Hypothesis Three: The Interviewer Has No Effect in Altering Respondents' Health State Values

Comparing the second value by the Initially Uninformed Group and the first value by the Informed Group tests whether the interviewer had any effect on the inflation of the second values provided the former group. Identifying prevarication and interviewer biases was the principle motivation for having two groups of participants in the study design, as discussed previously in Section 6.1. Evaluating the effect of an intervention – in this case, the adaptation exercise – with a single group runs the risk of individuals changing their health state values to please the interviewer (i.e., prevarication bias) (Hiebert and Nordin, 2006). Similarly, there is concern that the interviewer could inadvertently coerce the respondents into changing their initial values (i.e., interviewer bias). This effect must be evaluated if the true impact of the adaptation exercise on health state valuations is to be identified.

Null Hypothesis Four: The Adaptation Exercise is No More Effective in Altering Health State Values than the Patient Values Presentation

The impacts of the adaptation exercise and of the patient values presentation on the health state values were compared. This was achieved by comparing the magnitude of change between the two values of the Initially Uninformed Group and between the two values of the Informed Group, while recognizing that the difference between the latter group's values is the combined effect of the patient values presentation and the adaptation exercise.

8.2.2 Objective Two: To Determine the Factors That Influence an Individual to Change Their Values

The second study objective is to explore the factors that influenced individuals to alter their initial health state values. In addition to conventional individual characteristics (e.g., sex, age, illness experience), responses to the RCQ (shown in Appendix A.11, page 306) – developed based on the findings from the qualitative study (described in Section 7.3) – were used to shed light on what other factors may influence individuals to change their initial health state values. Responses to the RCQ were subjected to principal components analysis to reduce items to smaller numbers of components; this process is described in Section 8.4.4.

8.3 Data Collection

The aim of the data collection process was to obtain responses from members of the general population, subdivided into the Initially Uninformed and Informed Groups. The respondents were selected so that their age-sex distribution was representative of the general population of adults in the United Kingdom (UK).

The Faculty of Health and Wellbeing at Sheffield Hallam University conducted the interviews for this study. This organization, which has worked with The University of Sheffield's School of Health and Related Research on a number of previous valuation studies, undertook the sampling and management of the interviews; the Ph.D. candidate entered, collated, and analyzed all the raw data. The recruitment and interviews were conducted following the Ph.D. candidate's specifications. The interviewer assigned to this study was experienced in the use of both the visual analogue scale (VAS) and time trade-off (TTO) approaches. Over the course of the data collection period, the Ph.D. candidate accompanied the interviewer for 7% (14/200) of the interviews to check the quality and consistency of the interviews being conducted. This also provided the Ph.D. candidate with the opportunity to obtain a sense of both how the respondents were being approached to participate in the study and how the interviews were being conducted.

8.3.1 Sampling and Study Participants

To obtain a representative sample of the general population, respondents were sampled using the AFD Names and Numbers version 3.1.25 (AFD Software Limited, Ramsey, Isle of Man, UK). This software provides access to a comprehensive list of names and addresses of over 39 million people living in the UK. While the interviews were supported by two project grants, the funds were, nonetheless, limited. Thus, only two towns in the County of South Yorkshire were sampled for this study: Barnsley and Rotherham. They were selected for convenience due to their proximities to the interviewer's home; these towns, however, were similar to the national statistics in terms of age and presence of long-term illness (UK Statistics Authority, 2009). As this was a methodological study, the fact that the interviews were conducted locally rather than achieving a wide geographical distribution was not considered to be a serious limitation to the results.

Various neighbourhoods in Barnsley and Rotherham, representing different socio-economic classes, were identified by postal codes. A random sample of households within the selected neighbourhoods were sent a letter by post, inviting them to take part in the research study and informing them that an interviewer was to call at their homes (a copy of the invitation letter is displayed in Appendix A.12, page 308); the letter also included a leaflet providing information about the study (shown in Appendix A.13, page 309). When the interviewer arrived at an identified household, she asked the person who answered the door if he/she, or someone else in the household, would be interested in participating in the study. If an agreement was made, an interview was arranged in the home at a time that was convenient for the participant. All the participants were required to sign a consent form before participating in the study (shown in Appendix A.14, page 311).

The interviewer was instructed to arrange the sampling such that an equal number of individuals with similar distributions of sexes and ages were selected for the Initially Uninformed and Informed Groups. She was given a spreadsheet with the number of men and women in seven age groups to be recruited for this study; as mentioned above, the gender and age distribution aligned with the UK population demographics obtained from the National Census (UK Statistics Authority, 2009). By keeping a record of the individuals' characteristics (e.g., sex and age) and having it readily accessible, the interviewer was able to easily allocate the respondents into either the Initially Uninformed Group or the Informed Group while ensuring similarity across the groups. When the interviewer approached an interested individual, she asked for his/her age; this was to avoid oversampling certain groups. If, however, oversampling was an issue, the interviewer asked whether there was someone else present in the household who would be willing to participate in the study instead. At times, the interviewer had to turn down willing individuals, and households, to ensure that certain sex-age groups were not over-represented in the study sample.

At the end of the interview, the interviewer completed five questions (described in Appendix A.15, page 312). These questions were designed to evaluate the interviewer's impressions of the participant's understanding of the valuation tasks and the adaptation exercise, and the participant's involvement during the interview session. Each question was measured on a five-point Likert scale, ranging from strongly agree to strongly disagree. (The interviewer topic guide is presented in Appendix A.6, page 279.)

The interviews were conducted between October 2008 and January 2009. The University of Sheffield Ethics Committee approved the study protocol (copies of the ethics certificates are provided in Appendix A.8, page 299).

8.4 Data Analyses

To meet the study objectives outlined in Section 8.2.1, the dataset was subdivided according to the group allocation of the respondents. The dataset was not pooled together because the first and second valuations of the two groups have different implications. For example, the first values by the Initially Uninformed Group were not subjected to the adaptation exercise while the first values by the Informed Group were; thus, aggregating these values would not be meaningful.

All analyses described in this section were conducted using SPSS version 14 for Windows (SPSS, Chicago, IL, USA). Statistical significance for all tests was defined as $p \leq 0.05$.

8.4.1 Description of the Study Sample

The participants were characterized on the basis of their sex, age range, education level, employment status, illness experience, and, as a proxy for current health status, a self-rated EuroQol-5D (EQ-5D) index. Categorical variables are presented as the proportion of the sample within each group while continuous variables are presented as means and standard deviations (SDs). Independent t-tests and χ^2 tests evaluated whether differences existed between the demographic variables and group allocation of the individuals. The responses to the interviewer questions, in terms of frequency, are reported.

8.4.2 Identification of Consistent Responses

Some of the study participants provided inconsistent responses when all their health state values were considered. Before comprehensively assessing the results, it was important to appraise the extent of these inconsistencies and to remove responses that were strictly inconsistent to ensure that the statistical results were meaningful.

Assessing the health state values at an individual level evaluated the logical consistency of the responses. It was hypothesized that respondents would prefer fewer and milder symptoms rather than more frequent and more severe symptoms. Therefore, for a given round of valuations (e.g., first valuation or second valuation) and for a given valuation technique (e.g., VAS or TTO), responses that were considered to be strictly logically inconsistent were:

1. any health state rated higher than Full Health; and
2. health states pertaining to RA not rated in the order of State X $\succ$ State Y $\succ$ State Z (i.e., mild RA $\succ$ moderate RA $\succ$ severe RA) where '$\succ$' represents a greater preference for one state over another.

The description of the three RA states is presented in Table 5.14 (page 111).

If a respondent demonstrated logically inconsistent health state valuations for both the VAS and the TTO, then this respondent was removed from the dataset and their responses were excluded from all analyses. However, if a respondent demonstrated inconsistency for only, say, the TTO, then the VAS values was retained. In addition, if a respondent indicated that the values for any two, or all three, RA states were equal (i.e., State X = State Y, State Y = State Z, or State X = State Y = State Z), then his/her responses for a particular valuation method was excluded. The resulting sample therefore contained only strictly logically consistent responses which, hereafter, is referred as to as the 'more strict response set'.

A second approach was also undertaken on the full dataset which allowed for some minor logically inconsistent responses to remain in the dataset. Responses demonstrating 'Your Own Health' higher than Full Health on the VAS and responses demonstrating that any two, or all three, RA states were given approximately equal values (i.e., within 0.04 units – a difference of one life-year) by TTO (i.e., State X $\approx$ State Y, State Y $\approx$ State Z, or State X $\approx$ State Y $\approx$ State Z) were kept for future analyses. The purpose of this approach ensured that only the most severely inconsistent responses were removed. Thus, the remaining dataset was believed to be more statistically powerful (i.e., more responses retained) than the more strict response set that was outlined above. The resulting sample is denoted as the 'less strict response set'.

Responses for both the less strict and the more strict response sets were characterized in terms of sex and age, and compared to those obtained for the complete set. The health state values for the three datasets – complete set, less strict response set, and more strict response set – were characterized in terms of means and SDs. For each health state, the absolute size of change between the two values was compared:

$$Change_{njm} = Value_{2_{njm}} - Value_{1_{njm}}\ , \tag{8.1}$$

where n is the respondent, j is the health state, m is the valuation method, and $Value_1$ and $Value_2$ are the health state values individuals provided during their first and second valuations, respectively.

The Dataset to Be Used in Subsequent Analyses

The use of effect sizes provided a means to determine which of the three datasets – complete set, less strict response set, and more strict response set – should be used in subsequent statistical analyses. Calculating the effect size can provide information on the strength of the relationship between any observed effects independently from the numbers in the groups. This measure is determined by:

$$Effect\,Size_{jm} = MD_{jm} \Big/ \sqrt{\frac{SD_{1_{jm}}^{\ 2} + SD_{2_{jm}}^{\ 2}}{2}} \tag{8.2}$$

where j is the health state, m is the valuation method, MD is the mean difference between two health state values and SD_1 and SD_2 are the SDs of the first and second health state valuation, respectively. By using Cohen's (1988) criteria, the calculated effect sizes can be described as small, medium, or large. An effect size between 0.2 and 0.3 may be considered a 'small' effect, 0.4 to 0.7 a 'medium' effect, and 0.8 to 1.0 a 'large' effect. If the effect sizes for the different datasets remained the same when assessing the health states, then the dataset with a higher number of responses should be used to ensure greater statistical power since removal of the inconsistent responses did not affect the results. However, if the effect sizes differed, a decision had to be made between using a dataset with the least number of inconsistent responses or a dataset with a large enough sample to obtain reliable statistical results.

When the dataset to be used in subsequent statistical analyses was chosen, the health state values were characterized as means and SDs, and as percentiles (i.e., 5%, 50% (median), and 95%). These summary statistics provided a check for the validity of the responses. Individuals, on average, should have valued the health states such that the means were ordered as: Full Health $\succ$ Dead, and State X $\succ$ State Y $\succ$ State Z. The percentiles provided information about the distribution about the responses: differences between the mean and median for any state suggested skewed responses. For each health state, the number of individuals valuing them negatively (i.e., states worse than dead) was also reported.

The TTO values for respondents from the Informed Group were investigated in greater detail. The frequency of these values, relative to being higher or lower than the patient values that were presented to them, was reported. This aimed to assess whether viewing patient values had a direct impact on the Informed Group's second valuations.

8.4.3 Testing the Four Null Hypotheses

A series of statistical tests of association was conducted (Table 8.1, page 200). These tests were intended to address the null hypotheses previously set forth in Section 8.2.1.

In determining the size of the sample to be used in the study, there needed to be enough observations to detect differences between the valuations both within the same group and between different groups. It was proposed that the sample size should have an 80% power to detect a mean difference (*MD*) of 0.05 between health states, assuming a common *SD* of 0.10 when using an independent t-test at the 5% significance level:

$$Sample\ Size_{per\ group} = 2 * \left[\frac{(z_A + z_B)(SD)}{MD} \right]^2 , \qquad (8.3)$$

where z_A is 1.96 and z_B is 0.84. This calculation indicated that a minimum sample of 63 individuals in each group was needed; however, a total of 200 individuals were recruited to ensure that the minimum sample size was retained while allowing for the removal of logically inconsistent responses.

Null Hypothesis One: The Adaptation Exercise Has No Effect in Altering Health State Values

Paired t-tests were conducted to compare the first and second valuations of the same health state by the same respondent in the Initially Uninformed Group. If no statistically significant differences were observed between the values, this indicated that the adaptation exercise had no impact on the individual's valuations.

Null Hypothesis Two: The Patient Values Presentation Has No Effect in Altering Health State Values

Paired t-tests were also conducted on the health state values provided by the individuals in the Informed Group. If no statistical difference was detected between the first and second values for each health state, this indicated that the presentation of the patient values had no influence on the valuations.

Null Hypothesis Three: The Interviewer Has No Effect in Altering Respondents' Health State Values

Conducting independent t-tests between the Initially Uninformed Group's second valuation and the Informed Group's first valuation identified whether the interviewer had an effect on the respondents' health state values. If no statistically significant differences between these valuations were present, this would suggest that individuals in the Initially Uninformed Group had *not* increased their second values either in order to satisfy the interviewer or because the interviewer subconsciously encouraged individuals in this group to increase their initial values. If, however, significant differences were detected between the aforementioned values, then inflation in the Initially Uninformed Group's second values may have been due to interviewer effects.

Null Hypothesis Four: The Adaptation Exercise is No More Effective in Altering Health State Values than the Patient Values Presentation

The magnitude of change between the health state values was assessed for both the Initially Uninformed and Informed Groups (Equation 8.1). Change was described using means and SDs. Independent t-tests assessed whether no difference existed between the aggregate change values of the Initially Uninformed and Informed Groups. If there was a statistically significant difference between the change values, and the change between the two valuations by the Initially Uninformed Group was greater than those provided by the Informed Group, then the adaptation exercise would have a greater effect than the patient values presentation, when preceded by the adaptation exercise.

8.4.4 Individual-Level Analysis of Change

While the preceding section described statistical tests to determine the impact of disease adaptation information on health state values, there is also a need to explore the factors that influenced individuals to alter their initial health state values. Using linear regression techniques, a relationship can be described between the continuous change in health state values for both VAS and TTO methods and one or more explanatory variables. Possible explanatory variables may include both standard individual characteristics (e.g., sex, age, and illness experience) and responses to the RCQ. The resulting multivariate linear regression function is represented by a linear combination of more than one regression coefficient. The coefficients indicate the magnitude of the change in the health state value for every one-unit increase for a given explanatory variable, while holding all other variables in the model constant.

To examine this change, individual-level data was explored. The reason for this is that an individual-level analysis is more statistically powerful than at the aggregate level. Since there were three RA states valued by two methods (i.e., VAS and TTO), six change values were used per respondent.

Individual-level analysis was conducted using responses only from the Initially Uninformed Group. The rationale behind this decision to evaluate only the Initially Uninformed Group responses was that the RCQ may not have been capable of detecting the reasons for changes detected for respondents in the Informed Group. While the valuations provided by the Informed Group were primarily to test the presence of interviewer effects, its secondary purpose was to evaluate the effect of the patient values presentation. However, the independent effect of the patient values presentation could not be evaluated since it was preceded by the adaptation exercise. The adaptation exercise may have altered the Informed Group's initial opinions regarding the health states they valued and the RCQ was not able to capture this change.

First, before any modelling was conducted, the dependent variable had to be tested to determine whether or not it was normally distributed. Then the Initially Uninformed Group's responses to the RCQ were subjected to principal components analysis in an attempt to reduce the items in the instrument to a smaller number of principal components to be included as explanatory variables in the regression modelling. Bivariate analysis was performed using the derived principal components and conventional demographic variables (e.g., age and sex). Once these steps were completed, multivariate linear regression modelling was conducted.

Test for Normality

The intention was to develop a multivariate linear regression model to predict the factors which cause an individual to change their initial health state values. This model requires that the dependent variable – in this case, the continuous change in values – be normally distributed. To examine whether this assumption was met by the dependent variable, two plots were constructed: a histogram and a quantile-quantile plot. A histogram is a graphical display of tabulated frequencies. If it is observed to be a bell-shaped function with a peak at the mean, then the responses to the dependent variable are considered normally distributed. A more useful tool for assessing normality is the quantile-quantile plot. It is a scatterplot with the quantile of the scores on the horizontal axis and the expected scores (assuming a normal distribution) are plotted on the vertical axis. A plot of these scores against the expected normal scores should reveal a straight line; curvature of this line indicates departure from normality. If, however, a non-normal distribution was detected, the dependent variable could be transformed using a mathematical function (e.g., natural logarithm) to ensure the responses are normally distributed before conducting a multivariate linear regression. It is important to note that if the dependent variable was transformed, care must be taken in interpreting the meaning of the explanatory variables.

Principal Components Analysis

Before regression techniques were applied to determine the factors that influence respondents to change their valuations, the items of the RCQ were subjected to principal components analysis (PCA) to reduce the large number of items to a smaller number of domains. PCA is a statistical technique which aims to simplify complex sets of data by transforming possibly correlated variables into a smaller number of uncorrelated variables; these are referred to as principal components, or components (Kline, 1994). The first principal component accounts for the largest proportion of the variability in the dataset while each succeeding component accounts for as much of the remaining unexplained variability as possible. This technique was used to reduce the 25 items of the RCQ to a more reasonable number so that fewer tests were undertaken in the regression model.

The first step in the PCA was to examine the inter-correlation between the items. If any items of the RCQ did not correlate well with other items ($r < 0.20$) (Field, 2005), then these items were removed, as some correlation between items was needed to identify principal components. Similarly, items were excluded if they were too highly correlated ($r > 0.80$) with other items (Field, 2005); removal of these items alleviated the potential for multicollinearity. Within each set of items that demonstrated either low or high correlation, an item was removed one at a time and the R-matrix was assessed. The item that was chosen to be excluded should have resulted in R-matrix with the highest determinant (i.e., greater than 1×10^{-5}). Then the inter-correlation between items was re-assessed until the remaining items were moderately correlated with each other ($0.20 < r < 0.80$).

The second step was to assess whether the dataset was suitable for PCA through the use of the Kaiser-Meyer-Olkin (KMO) measure of sampling adequacy and Bartlett's test of sphericity. The KMO test statistic assesses the patterns of correlations in a given dataset. A KMO value of zero indicates that the correlation patterns are widely spread such that the sum of partial correlations is large relative to the sum of correlations. A KMO value of one indicates that the patterns of correlations are compact and distinct, resulting in reliable components. An adequate value for the KMO test statistic is between 0.5 and 0.7 but ideally this value should be higher (Field, 2005). The Bartlett's test of sphericity examines whether the original correlation matrix is an identity matrix (e.g., all correlation coefficients are zero); some relationships between variables need to be present for PCA to be successfully applied to a given dataset. A significant Bartlett's test statistic implies that the R-matrix is not an identity matrix and hence PCA can be applied to the data.

The third step in the PCA determined the ideal number of principal components for the given dataset; this was accomplished by using the Kaiser criterion and by examining the Scree plot. The Kaiser criterion is based on the eigenvalues, which are determined during the extraction of the components. The eigenvalues associated with each factor represent the variance explained by that particular linear component. The eigenvalues of the factors after rotation are displayed. Rotation has the effect of optimizing the factor structure; Varimax rotation was chosen for this study because it is considered to be an excellent method of reaching orthogonal simple structure (Kline, 2004; Field, 2005). Alternatively, the Scree plot, describing the relationship between number of eigenvalues and number of components, can also be used to determine the number of suitable components. A point of inflection on the curve indicates the most efficient number of components; this, at times, can be difficult to detect based on the nature of the curve. If, however, a discrepancy between the number of components obtained from the Kaiser criterion and the Scree plot exists, then the items in each of the components from both methods need to be carefully assessed. In this study, when the Kaiser and Scree methods differed, the method that yielded components with the most logical groupings of items was preferred. The identified principal components were to be considered as potential explanatory variables in the regression modelling.

Bivariate Relationships

Once the principal components of the RCQ were identified, the next step was to identify the factors which cause an individual to change their initial values through a linear regression. However, before any modelling was conducted, the relationships between potential explanatory variables were assessed. Scatter plots were used to display the relationships between the observed change and the continuous explanatory variables. The strength of the correlation between the variables was examined by the R^2 value. The relationships between change and categorical variables, on the other hand, were identified using boxplots. Boxplots provide a means of graphically depicting groups in terms of the sample minimum, lower quartile, median, upper quartile, and sample maximum; these five summary statistics – known as Tukey's values – were reported.

Main Effects and Interaction Terms

Based on the results from the qualitative interviews, the following main effects were expected to influence whether an individual changed their health state values: sex, age, illness experience, current health status, RCQ component scores, RA state valued, and valuation method used. Of the main effects, the RCQ component scores were the only variables measured along a continuous gradient; the remaining variables were categorical and hence were dummy-coded.

From the main effects, interaction terms were created. These were derived from researcher judgement which was guided by the results from the qualitative component described in the previous chapter. It is likely that some of the RCQ components may interact with the information pertaining to an individual's experience with arthritis and chronic conditions, and current health status; some of the RCQ items specifically tapped into these constructs. Furthermore, it was expected that the RCQ components might interact individually with sex, RA state, and valuation method. Illness experience was expected to interact with an individual's current health status, as these variables correlated with each another. In addition to these two-way interactions, three-way interaction terms were constructed between RA states, valuation method, and the RCQ component scores.

Multivariate Linear Regression

Linear regression describes a relationship between one or more explanatory variables and the dependent variable which is, in this case, the change in health state values. The multivariate linear regression function is represented by a linear combination of more than one regression coefficient. The coefficients indicate the magnitude of the change in the predicted health state value for every one-unit increase for a given explanatory variable, while holding all other variables in the model constant.

Before multivariate linear regression was conducted, univariate analyses identified those variables which best explained the changes observed in the health state values. The explanatory variables were considered on their own in the model and their significance was evaluated using the t-test statistic. If a categorical variable was comprised of more than one level – for example, age group – the model fit was also assessed using the F-test statistic.

Using backwards regression, a model containing only the main effects was constructed. All variables were entered into the model, but only those that had a significance level of $p < 0.05$ were retained in the final model. After the main effects model was finalized, a second model was constructed to incorporate the interaction terms. These terms were manually entered into the model one at a time; this step was repeated until the best fitting model arose.

The F-test statistic was used to examine the overall significance of both models. A significant model implies that there is at least one regression coefficient statistically different from zero. The overall fit of the model was assessed using the R^2 value.

8.5 Results

8.5.1 Characteristics of the Study Participants

A total of 200 individuals participated in the study. To recruit this number of respondents, invitation letters were posted to 649 addresses in the Barnsley and Rotherham areas. Of these addresses, the interviewer approached 629 homes. Out of those households approached, 167 (27%) refused to participate and, after making at least three attempts, 175 (28%) were not in or no contact was made. The interviewer rejected 50 (8%) potential respondents because of concerns regarding their ability to complete the interview due to age or existing health conditions. A further 37 (6%) were excluded to avoid oversampling of certain age-sex groups. Interviews were conducted with the remaining 200 respondents, resulting in a response rate of 32%.

Table 8.2 (page 201) shows the characteristics of the study participants. The distribution of the individuals, in terms of age and sex, aligned with the data obtained the current census results (UK Statistics Authority, 2009). Within each group – Initially Uninformed and Informed Groups – there were 48 males and 52 females of varying ages. The two groups were similar in terms of marital status, education level, employment status, illness experience, and EQ-5D indices. The mean (SD) time for the participants to complete the interview was 46.9 (11.6) minutes.

The results obtained from the five interviewer questions are also reported in Table 8.2 (page 201). On average, the interviewer felt that the majority of the participants understood what was expected of them when completing the VAS and TTO exercises, and did not lose interest when valuing the health states that came later in the sequence. For the questions pertaining to 'the respondent gave a lot of thought to the decisions that he/she was asked to make' and 'the respondent was engaged with the patients' messages in the recordings', the interviewer response was skewed towards the 'positive' outcome, although the most frequent response was 'neither agree nor disagree'. For both questions, in only a few cases did the interviewer feel strongly that the participant had not carefully thought about their decisions or engaged with the patients' messages in the recorded scenarios.

8.5.2 Logically Inconsistent Responses

When applying the strictly logically inconsistent criteria for excluding inconsistent responses, 156 VAS values (79 responses by the Initially Uninformed Group and 77 by the Informed Group) and only 65 TTO values (35 responses by the Initially Uninformed Group and 30 by the Informed Group) remained. A large number of TTO responses were removed for the strict criteria because many of the respondents valued at least two of the three RA states similarly (i.e., within 0.04 units), such that State X $\approx$ State Y, State Y $\approx$ State Z, or State X $\approx$ State Y $\approx$ State Z. When the strictly logically inconsistent criteria were relaxed, 175 VAS values (84 responses by the Initially Uninformed Group and 91 by the Informed Group) and 179 TTO values (90 responses by the Initially Uninformed Group and 89 by the Informed Group) remained.

The age-sex distribution for all three datasets is shown in Table 8.3 (page 203). In general, when the inconsistent responses were removed for the VAS approach, the number of remaining responses on the more strict and less strict response sets were similar to those obtained for the complete set. However, the number of TTO responses remaining for the more strict response set was considerably lower than that obtained for the complete set. For example, there would only be 13 and 15 male respondents in the Initially Uninformed Group and Informed Group, respectively, compared to the 48 males respondents in each of the groups for the complete set. With the less strict set, these were increased to 43 in each group (out of a total number of 48 per group). Thus, using the less response set should generate responses that were representative of the general population, although the more strict response set would not.

Table 8.4 (page 204) presents the health state values for the complete, less strict, and more strict response sets. In general, similar values were observed for Full Health, Your Own Health, and Dead across the three datasets. Not surprisingly, given that the majority of TTO responses were removed for the more strict response set, the values differed from those obtained using the complete dataset. This was particularly noticeable for State Y and State Z (i.e., moderate and severe RA states). In general, the TTO values provided by the less strict response set were more closely aligned to the complete dataset than those from the more strict response set. For example, the first TTO valuation for State Z was 0.25 for both the complete and less strict response sets, while this state was valued at 0.10 for the more strict response set.

The effect sizes presented in Table 8.5 (page 205) showed that the adaptation exercise, in general, had a greater effect on the individual's health state values than did the patients values presentation, as the effect sizes were greater for the Initially Uninformed than for the Informed Groups. The only exceptions were for State X and State Y (i.e., mild and moderate RA states) when valued by TTO in the more strict response set (i.e., greater effect sizes were observed for the Informed Group when compared to the Initially Uninformed Group). It is noticeable that the interventions resulted in effect sizes that were greater when valuing the health states by VAS when compared to TTO. For example, when using the less strict response set, effect sizes of 0.41 and 0.24 were afforded for VAS and TTO, respectively, for State X.

Based on the results outlined in the preceding paragraphs, the decision was made to use the less strict response set in all subsequent analyses. This was due to a larger number of TTO values that remained after removing the most severely inconsistent responses when compared to the numbers that remained in the more strict response set (Table 8.3, page 203) and the fact that the effect sizes were relatively similar across all response sets (Table 8.5, page 205). While it is recognized that the complete response set could have been used in lieu of the less strict response set, the most inconsistent responses (which were indicative of respondents who had not understood the valuation tasks) were still eliminated in the least strict response set.

8.5.3 Aggregate Health State Values

With the decision made to use the 'less strict' response set for all statistical analyses, the relevant aggregate health state values for the Initially Uninformed Group and Informed Group could be determined, as are presented in Table 8.6 (page 206). The expected trends were observed, such that rankings for the aggregate VAS values demonstrated that individuals valued Full Health $\succ$ Your Own Health $\succ$ State X $\succ$ State Y $\succ$ State Z $\succ$ Dead. Similarly, the TTO valuations for the three RA states were logically ordered, with State X $\succ$ State Y $\succ$ State Z. When comparing the mean and median health state values (Table 8.6), the similar values suggested that the VAS values were symmetrically distributed. The means and medians for the TTO were dissimilar, suggesting that the TTO values were right-skewed; however, this was not expected to be a problem in the regression analysis because it was the changes in health state values, rather than the absolute health state levels, that were being modelled. In general, an individual's second attempt at valuing the RA states resulted in a positive change when compared to the first valuation.

This, however, was not the case for the Informed Group. For two health states, State X, when valued using the TTO, and State Z, when valued by the same group using the VAS resulted in the opposite trend (Table 8.6, page 206); in other words a negative change was detected. Also, State Y when valued by VAS demonstrated no change between the first and second valuation.

Table 8.7 (page 207) displays the proportion of individuals in the Informed Group that valued the RA states higher or lower relative to the values provided to them by the patient values presentation. When examining the second valuation (i.e., values subjected to the patient values presentation), the majority of respondents provided values higher than the patient values for States X and Y; for State Z, 57% of the respondents gave a value less than the patient value. When comparing the proportion of values relative to the patient values during the first valuation, the frequency of those providing health state values higher than the patient values was similar in magnitude to the second valuation. This implied that seeing the patient values, after having undergone the adaptation exercise, had a limited effect on changing an individual's health state values.

When assessing the second, and final, valuations, both the Initially Uninformed and Informed Groups provided similar values for all three RA states; only State X (i.e., mild RA) was valued slightly higher by the Initially Uninformed Group. It is interesting to observe that providing disease adaptation information (i.e., adaptation exercise and patient values presentation) encourages similar patient and general population values for all but the most severe RA state (i.e., State Z).

States Worse Than Dead

The proportion of individuals in the Initially Uninformed Group and in the Informed Group that valued health states negatively (i.e., states worse than dead) are reported in the right-hand column of Table 8.6 (page 206). While only a small number of participants valued States X and Y as being worse than dead, over a quarter of the sample valued State Z by TTO negatively: 31% in the Initially Uninformed Group valued this state negatively, whereas 26% in the Informed Group did for their first valuation. A slight reduction in numbers was observed when this state was valued during the second valuation.

Null Hypothesis One: The Adaptation Exercise Has No Effect in Altering Health State Values

Results from the paired t-tests showed that the Initially Uninformed Group changed their values for most health states (Table 8.8, page 208) (i.e., p-values ≤ 0.05). This provided evidence that the adaptation exercise was, in fact, effective in altering respondents' initial health state values. However, this result does not address the concern that the observed increase might be due to an interviewer effect; this is tested in null hypothesis three.

Null Hypothesis Two: The Patient Values Presentation Has No Effect in Altering Health State Values

The paired t-test revealed that the Informed Group showed statistically significant changes for the health state values for State Z when assessed using the TTO ($p < 0.01$) (Table 8.9, page 209). This demonstrated that the presentation of patient values, when the respondents were informed about adaptation through an adaptation exercise prior to the first valuation, played a limited role in altering health state values. This intervention influenced change only for the most extreme health state and for only one valuation method.

Null Hypothesis Three: The Interviewer Has No Effect in Altering Respondents' Health State Values

When independent t-tests were conducted to compare the second valuation of the Initially Uninformed Group and the first valuation of the Informed Group, the results indicated that there were no significant differences (i.e., no p-value ≤ 0.05) between these two values across all states (Table 8.10, page 210). The results demonstrate that individuals in the Initially Uninformed Group may have slightly inflated their values; however, this increase was not found to be statistically significant. This implies that any interviewer effect did not explain the differences found between the first and second valuations of the Initially Uninformed Group.

Null Hypothesis Four: The Adaptation Exercise is No More Effective in Altering Health State Values than the Patient Values Presentation

The magnitude of change between the first and second valuations (Table 8.11, page 211) for both groups was evaluated using independent t-tests. The results revealed that individuals in the Initially Uninformed Group increased their second valuation more than those in the Informed Group (i.e., positive mean values). The results from the between-group tests showed that a statistically significant difference arose between the two groups for the following states: State X (by TTO), State Y (by VAS), and State Z (by both VAS and TTO). The greater change in the health state values observed in the Initially Uninformed Group implied that the adaptation exercise had more of an impact on the individual's valuations than did the patient values presentation, at least when the latter was preceded by an adaptation exercise.

8.5.4 Principal Components Analysis

Inter-correlation between the Variables

The inter-correlation between RCQ items was assessed for the Initially Uninformed Group's responses using the less strict response set. Two pairs of items were highly correlated with each other ($r > 0.80$): 'taking part in today's session has helped me to understand more about the disease of arthritis' was highly correlated with 'taking part in today's session has helped me to understand more about what it is like to live with arthritis'; and 'my opinions about arthritis changed after hearing the recordings' was highly correlated with 'my opinions about arthritis changed after talking with the interviewer'. The exclusion of the latter item of each of these pairs resulted in a larger matrix determinant (i.e., greater than 1×10^{-5}) and the desired moderate inter-correlation values between all items ($0.20 < r < 0.80$), as discussed in Section 8.4.4.

Suitability of Principal Components Analysis for the Dataset

The PCA yielded KMO test statistics of 0.73, which exceeds the range of adequacy (0.5-0.7). The result from Barlett's tests of sphericity was statistically significant ($p < 0.01$), indicating that the R-matrix was not an identity matrix. The results from these two tests demonstrate that the less strict response set was suitable to be subjected to PCA.

The Number of Principal Components

Using the Kaiser criterion, seven principal components were identified (Table 8.12, page 212). The Scree plot, however, showed little indication of a point of inflection (Figure 8.1, page 216). The seven components identified by the Kaiser criterion related to the information extracted from the qualitative interviews, as described in Chapter Seven. The seven components that comprised the RCQ could be best described as: personality, information, recognition of coping strategies, opinions of arthritis, empathy, ease with the valuation exercises, and illness experience.

Overall, the items that comprised the components were logical. For example, the 'Empathy' component was comprised of the following RCQ items: 'in general, I feel that if I had to, I think that I can imagine living with arthritis for the rest of my life'; 'in general, I feel that it's unfair for me to value a patient's life because I don't really know what it's like to live with arthritis'; and 'in general, I feel that I can "put myself in other people's shoes" and see things from their point of view'. An exception was the component labelled 'Information'. It contained the item 'in general, I feel that I had a difficult time deciding how long I wanted to live with arthritis on the valuation exercises'. It was expected that this would belong in the 'Ease with Valuation Exercises' component but it did not. While this item did not seem to fit well, when compared to the other items in that component, it loaded highly on the 'Information' component at 0.74. Thus, there is likely an unexplained relationship between this item and the other 'Information' components that needs to be explored in the future. The other items that comprised the 'Information' component were 'my opinions about arthritis changed after hearing the recordings' and 'taking part in today's session has helped me to understand more about the disease of arthritis'.

With the seven principal components identified, they could then be used as potential explanatory variables in the regression modelling. This process is described in the following section.

8.5.5 Individual-Level Analyses

Test for Normality

The histograms for change in health state values from valuation one to valuation two for the Initially Uninformed Group produced a bell-shaped function, which implies that these values are symmetrically distributed (Figure 8.2, page 217). The quantile-quantile plot, Figure 8.3 (page 217), however, shows some curvature in the relationship, indicating a departure from normality. Several transformations were conducted – including natural logarithm, standardization, and Box-Cox – in an attempt to normalize the distribution. However, none of these transformations resulted in a quantile-quantile plot that was more linear than that shown in Figure 8.3. Unsuccessful transformation of non-normal TTO values has been reported before (Dolan and Roberts, 2002), although this previous study assessed health state level rather than changes in values. Based on these previous findings, and the fact that none of the transformations substantially improved the linearity of the quantile-quantile plot, the untransformed model was used for further analysis. However, it is recognized that the assumption of normality might not be fully met.

Bivariate Relationships

Scatter plots and boxplots (not shown, but summarized in Table 8.13, page 213) described the relationship between change and potential main effects variables to be included in the regression models. As discussed previously, scatter plots were used for continuous variables and boxplots for categorical variables. The strength of relationship between change and the RCQ component scores was negligible ($R^2 < 0.04$). Tukey's values from the boxplots for changes in health states values are shown in Table 8.13. In general, a greater change in health state values was detected for individuals who had no illness experience, who were youngest in age, and who had a good health status. When individuals valued the same health states for the second time, a greater change was observed for State Z when compared to States X and Y.

Univariate Analyses

Univariate analyses were conducted to assess what variables were statistically significant when included individually in a model addressing changes in health state values (Table 8.14, page 214). Results indicated that the change was influenced by age ($p < 0.01$), current health status ($p < 0.01$), experience with illness ($p < 0.02$), personality component ($p < 0.04$), information component ($p < 0.01$), and ease with the valuation exercises component ($p < 0.01$); the severity of the RA state ($p = 0.14$)

and the valuation method ($p = 0.14$) did not contribute to the observed change. For example, individuals who were more likely to change their values were younger in age, healthier, had no illness experience, had positive personalities, were receptive to the new information presented, and had no difficulty with the valuation exercises.

Multivariate Analyses

Two multivariate linear models were constructed, one consisting of only main effects and another consisting of main effects and interaction terms.

Main Effects Model

Table 8.15 (page 215) presents the main effects that influenced individuals to change their initial health state values. Consistent with the findings from the univariate analyses (Table 8.14, page 214), significant main effects included age, current health status, and ease with the valuation exercises. Although illness experience, personality, and information were found to be statistically significant when tested univariately against the dependent variable of change in health state values, these main effects were not significant in the multivariate model. Similarly, the recognition of coping strategies variable was not significant in the univariate model ($p = 0.10$) but had an effect when tested alongside age, current health status, and valuation exercise ease, such that those individuals who felt that they could cope were more likely to change their initial values.

The main effects model had a constant value of 0.106 units, which represented the baseline level of change amongst all participants. When completing the second valuation for the RA states, younger individuals (i.e., less than 30 years of age) were more likely to maintain the baseline level of change than older individuals. Individuals between 30-59 years increased their values by only 0.05 units (i.e., by taking the sum of the coefficients corresponding to the constant and variable representing 30-59 years, 0.106 + (-0.061)) and those over 60 had negligible change (e.g., 0.106 + (-0.102)). Change was also influenced by the individual's current health status. Individuals in good health (i.e., EQ-5D index > 0.95) and moderate health (i.e., EQ-5D index 0.65-0.95) improved the baseline levels of change to 0.17 (i.e., 0.106 + 0.062) and 0.12 (i.e., 0.106 + 0.017), respectively, while those in poorer health (i.e., EQ-5D index < 0.65) maintained the baseline level of change. Although the moderate EQ-5D group was not statistically significant, it was included in the model to ensure that all the EQ-5D levels were captured in the full model (i.e., excluding one level would inhibit the interpretation of the influence of this variable.

Individuals who had better recognition of coping strategies increased their initial values by 0.13 (i.e., 0.106 + 0.022), whereas those who found the valuation exercises easy increased their values by 0.13 (i.e., 0.106 + 0.025). A low R^2 of 0.05 was determined for this model.

Main Effects and Interaction Model

As described previously in Section 8.4.4, potential interaction terms (e.g., illness experience*current health status) were added one by one to the main effects model, as outlined above. Two interaction terms were found to be statistically significant (Table 8.16, page 215). With the addition of these two terms, the signs for the main effects remained the same and the magnitude of each coefficient for the main effect differed only slightly from the previous model. As such, the interpretation of the main effects in the interaction model is the same as without the interaction terms, as presented in the previous section, and hence is not discussed here. Instead, this section focuses on each of the interaction terms, as described below.

The results indicate that the inclusion of the two interaction terms slightly improved the overall fit of the model that explains changes in health state values ($R^2 = 0.09$). The first interaction describes an individual's health status and their willingness to accept the information presented in the adaptation exercise. An increase in values of 0.12 (i.e., by taking the sum of the coefficients corresponding to the constant and the interaction term, 0.091 + 0.027) was detected for individuals reporting a healthy status (i.e., EQ-5D index > 0.95) and a willingness to accept the disease adaptation information. The second term described the interaction between the health states being valued and how the health state was valued. An increase in values of 0.21 (i.e., 0.091 + 0.114) was detected when individuals valued the most extreme RA state (i.e., State Z) by the TTO method. The latter result was not unexpected since many of the participants, after being informed about their values for the extreme state and its implications, reported that they were surprised that they had been willing to give up that many years of life to avoid living with RA; this is consistent with the findings from the qualitative study discussed in Chapter Seven.

Combining the main effects and the interaction terms gives a regression model describing a change in an individual's health state values represented by:

$$\begin{aligned} \textit{Change in values} &= 0.091 - 0.059\beta_{age2} - 0.112\beta_{age3} + 0.062\beta_{healthy1} \\ &+ 0.012\beta_{healthy2} + 0.020\beta_{coping\ strategies} + 0.026\beta_{ease\ with\ valuation\ exercises} \qquad (8.4) \\ &+ 0.114\beta_{State\ Z*TTO} + 0.027\beta_{healthy1*opinions\ of\ arthritis} \end{aligned}$$

where β_{age2} and β_{age3} are the regression coefficients corresponding to respondents in the 30-59 and the over-60 age group, respectively; $\beta_{healthy1}$ and $\beta_{healthy2}$ are the regression coefficients corresponding to respondents reporting EQ-5D indices > 0.95 and 0.65-0.95, respectively; $\beta_{coping\ strategies}$ is the regression coefficient corresponding to the ability that respondents recognize coping strategies; $\beta_{ease\ with\ valuation\ exercises}$ is the regression coefficient corresponding to how easy the respondent found the valuation exercises; $\beta_{StateZ*TTO}$ is the regression coefficient for the interaction term corresponding to respondents valuing State Z by the TTO method; and $\beta_{healthy1*opinions\ of\ arthritis}$ is the regression coefficient for the interaction term corresponding to respondents reporting EQ-5D index > 0.95 and their improved understanding of arthritis.

Based on Equation 8.4, the person who is likely to increase their health state value by the largest margin is someone with the following characteristics: under 30 years of age, in good health (i.e., an EQ-5D index > 0.95), with recognition of coping strategies, who found the valuation exercises easy to complete, and had an improved opinion of arthritis. Such an individual would increase his/her initial health state values by 0.23 (i.e., summing the coefficients in Equation 8.4: 0.091 + 0.062 + 0.020 + 0.026 + 0.027). When this individual valued State Z by the TTO method, the health state value increased to 0.34 units (i.e., 0.23 + 0.114).

Also based on Equation 8.4, the person who is likely to increase their health state values by the smallest margin is an individual with the following characteristics: over 60 years of age, in poor health (i.e., an EQ-5D index < 0.65), with poor recognition of coping strategies, who found the valuation exercises difficult to complete, and did not have an improved opinion of arthritis. This individual will decrease his/her initial health state values by 0.02 units. (i.e., 0.091 + (-0.112), as all other coefficients in Equation 8.4 are zeros).

8.6 Discussion

The main finding from the final component of the mixed-methods study, as presented here, was that individuals can be effectively informed about disease adaptation. This was verified by alteration of their initial values for RA states following the use of an adaptation exercise. Furthermore, the study revealed that an individual's age, current health status (as represented by EQ-5D index), recognition of coping strategies, and ease with the valuation exercises influenced their willingness to alter their valuations. This section discusses the findings presented in this chapter; its broader implications are described in the following final thesis chapter.

8.6.1 Values for Health States

Overall, the values for RA states were rated in a logically consistent order; this was observed to be the case even when internally inconsistent responses were not removed from the sample (i.e., using the complete response set). This provided evidence that the participants, on average, comprehended what was asked of them during the valuation exercises. In turn, this ensured that the results presented were valid.

Administration of the adaptation exercise had an influence on the individual's initial valuation of the RA states. This was detected by statistically significant changes between the two valuations across all health states, as well as by the larger effect sizes, provided by the Initially Uninformed Group when compared to the Informed Group. On the other hand, the patient values presentation, when preceded by the adaptation exercise, had a minimal effect on the health state valuations; the only significant change was detected when individuals in the Informed Group valued State Z by TTO.

Although not a statistically significant result, a change in the negative direction was observed for State X using the TTO approach for the Informed Group. When these participants observed the patient values before the second valuation, they informed the interviewer that they were surprised that patients would be willing to give up 4.8 years of their lives (from a total of 25 years) to not have to live with a mild form of RA. Thus, it is assumed that the observed reduction in values was a result of incorporating the information gained from patient values presentation.

The concern that the interviewer may have influenced individuals to inflate their second health state values after hearing the patients' recorded scenarios was explicitly addressed in the design of the study. Results from the independent t-tests between the values subjected to the adaptation exercise (i.e., the second values by the Initially Uninformed Group and the first values by the Informed Group) show minimal differences. This demonstrates that the observed change in valuations by the Uninformed Group likely did not result from either a desire to please the interviewer or from subconscious urging from the interviewer. As a result, the impact of the adaptation exercise on the health state values can be effectively compared using the Initially Uninformed Group's values.

The results indicate that, when using the standardized VAS as the technique for valuation, individuals were more likely to provide lower values for life in various RA states when compared to using TTO. This result contributes to the current body of evidence stating that different valuation techniques yield different results (Brazier et al., 2007). The lower VAS values may be a result of the respondents not considering the duration of the health states when making their assessments (Robinson et al., 1997). Without having an 'end date', respondents may not have the opportunity to carefully consider the impact QOL had on their attitudes toward length of life. Alternatively, the TTO encouraged the respondent to think about time spent in the impaired health state in one-year increments. A "threshold of tolerability" may have also contributed to the higher TTO values. The value of the health states may have to fall below a certain point before respondents would be willing to give up any time at all on the TTO (Robinson et al., 1997). The possibility of this threshold is likely as 26 respondents (13% of the complete response set) opted to not trade any, or just one, life year(s) for all three RA states. While these non-traders were regarded to be strictly inconsistent responses, the decision was made to keep these responses in the analyses to ensure that the number of responses retained for further analyses would be statistically powerful.

The results revealed that the provision of disease adaptation information (i.e., adaptation exercise or the combination of adaptation exercise and patient values presentation) yielded similar values for the three RA states regardless of group allocation; with only State X (i.e., mild RA) being valued slightly higher by the Initially Uninformed Group (Table 8.7, page 207). When comparing the final TTO values to those obtained from patients, general population values, after being subjected to disease adaptation information, afforded similar patient and general population values for all but the most severe RA state (i.e., State Z). This difference in values may be a result of members of the general population having difficulties envisioning adaptation due to the severity of the condition.

8.6.2 Changes in Health State Values

The difference between the first and second valuations provided by the two groups revealed that the change in values of the Initially Uninformed Group was larger. This indicates that the patient values presentation, when the first valuation was preceded by an adaptation exercise, may have had less of an impact on altering individuals' initial health state values than did the adaptation exercise alone.

The valuation methods had a different effect on the individuals' values for the different RA states. The VAS had a greater effect on the individuals' valuation for State X, such that a larger change between the valuations was detected when compared to the results from the TTO. On the other hand, a greater change occurred when individuals valued State Z using the TTO method. This may be a result of the range of sensitivity associated with each of the valuation methods. The VAS seemed to be more capable of differentiating between responses at the upper end of the scale, whereas the TTO appeared to be more sensitive at the lower end. This difference in sensitivity may be a result the expiration date (as discussed in above in Section 8.6.1) imposed with the TTO method and the lack thereof with the VAS approach.

The results obtained from assessing change in health state values need to be interpreted with care especially in cases where individuals at first valued a state as being worse than dead and then, after they were informed about adaptation, their impression of the state improved to being better than dead. The reason for this concern is that states worse than dead were 'transformed' (Dolan et al., 1996), as described previously in Section 6.2.3. Although this transformation has been used elsewhere in the literature (Patrick et al., 1994), states worse than dead (i.e., negative values) can no longer be interpreted to be on the same scale as those for states better than dead (i.e., positive values). This conversion allows negative values to range from -1 to 0. If this transformation had not been done, the minimum value for states worse than dead would reach -24, if trade-offs were limited to whole years. Accordingly, the results may be an underestimation of the amount of change observed because two different scales were used. However, this may not be a significant concern as only 12/179 (7%) of the individuals initially appraised State Z to be worse than dead at first but then changed their valuation to a state better than dead for the second valuation.

8.6.3 Individual-Level Analyses

Using the responses from the Initially Unformed Group, the factors that affected changes in health state values were explored both univariately and multivariately. When the main effects were tested univariately, age, EQ-5D indices, and illness experience influenced change in health state values. Along with the aforementioned variables, three principal components from the RCQ were also identified to impact change: personality, information, and ease with the valuation exercise.

Consistent with the univariate results, the multivariate analysis revealed that individuals who were younger in age or who had better health (i.e., high or moderate EQ-5D indices) were more likely to change their initial values by a larger magnitude than those who were older and were in poorer health. The responses to the RCQ revealed that an individual's recognition of their ability to cope and their ease with the valuation exercises also contributed to the individual's willingness to alter their initial values. The inclusion of interaction terms only slightly improved the overall fit of the model. When valuing the most severe RA state by TTO, individuals tended to increase the second valuation by a greater magnitude when compared to the milder two states. This was not unexpected since many individuals, after being informed about their values for the extreme state and its implications, reported that they were surprised that they were going to give up that many years of life to avoid living with RA; this is consistent with the findings from the qualitative study. In addition, healthier individuals with an improved opinion of arthritis were more likely to change their values for the most severe RA state.

The change model has a low R^2 value. This low R^2 is not a substantial cause for concern since the objective of this analysis was to assess the relative effect of the different respondent characteristics on the valuations rather than to find a model that explained all the variance in the valuations. It is possible that the RCQ may not be tapping into the all the rationales individuals may have for changing their initial health state values.

8.6.4 Study Strengths and Limitations

This is believed to be the first study that assesses what factors may influence members of the general population to change their health state valuations after being informed about disease adaptation. Information about disease adaptation has previously been presented as adaptation exercises (Ubel et al., 2005; Damschroder et al., 2005 and 2008), as described in Section 2.3.1, but this current study was the first to utilize audio-recordings of patient interviews (i.e., recorded scenarios) to demonstrate how real patients live with, and adapt to, the condition. Interestingly, comparison of the results from the current work with those from the study by Damschroder et al. (2008), which also used the TTO approach, yielded different outcomes. Damschroder et al.'s (2008) study found that the administration of the adaptation exercise had no effect on the health state values; this is contrary to the results presented in this thesis. This discrepancy between the results may be related to the fact that Damschroder et al. (2008) used a generic adaptation exercise (i.e., think back to a previous different life event and assess how your emotions toward this event changed over time) to encourage respondents to consider disease adaptation when valuing health states pertaining to paraplegia, below-the-knee amputation, colostomy, and severe pain. An adaptation exercise of this type may not have encouraged the respondents to focus specifically on concepts of adaptation related to the health states they were valuing. In the current work, the adaptation exercise was condition-specific, such that respondents were aware that people with RA can adapt to their health condition over time. They could then choose whether to apply this information directly to their health state values.

Three RA states were used to explore the research question; however, this number of states may not provide as large a range of severities for respondents to value as would be desirable. Furthermore, the description of the health states was mostly based on the HAQ (Table 5.14, page 111). Although the pain and discomfort dimension from the EQ-5D was incorporated as well, the use of four of five items from the HAQ may have made the health states too limited in scope. While physical limitations are a key part to the RA condition, other aspects may need to be considered when selecting domains to be included in the health states, such as social functioning and mental health, which were not included in the current work.

The order in which the health states was presented to the respondents may have also had an effect on the valuations. Respondents presented with a scenario in which a bad state is followed by a good state may value the states differently from a scenario where a good state is followed by a bad state (Dolan et al., 1996). To avoid this, the order of which the respondent valued each health state was randomized so the order should not have a consistent effect, as mentioned previously in Sections 6.2.2 and 6.2.3. However, this effect may have contributed to person-to-person variability. While the effect of health state ordering could be empirically examined, such an analysis would be complicated by the fact that individuals completed two TTO exercises, with differing orders, during the interview session.

There is also potential concern that a labelling effect was present. When respondents in the Initially Uninformed Group valued the health states during the first valuation attempt, they were not told that the health states focused on RA, or arthritis for that matter. However, when they underwent the adaptation exercise, they were informed that the recorded scenarios pertained to patients living with arthritis. As a result, when they were asked to complete the valuation exercises for the second time, they had the label of 'arthritis' in their minds. Since individuals in the Informed Group completed the adaptation exercise first, they were informed the states pertained to arthritis for both valuation attempts. While identifying the health states as States X, Y, and Z attempted to avoid any preconceived ideas about RA, using the recorded scenarios reintroduced these labelling effects. The use of labels may have affected the individuals' aggregate values, and their corresponding changes. As shown in the literature review presented in Chapter Three, the use of labels may lead to lower health state values because respondents may introduce emotion and stereotype into their valuations but the impact of labelling on individuals' health state values may also produce an opposite effect. Empirical work have demonstrated that the use of health state labels do not consistently yield lower values. Gerard et al. (1993) showed that there were no significant differences between the values for health states with or without labels. Preliminary work by Robinson and Bryan (2001) found that health state labels produced a mixed effect. Using nine different health scenarios (e.g., breast cancer, vision impairment, low back pain, and asthma), they found that labels, in general, negatively influenced individuals' responses on the VAS and TTO (i.e., respondents provided a lower value for the labelled states); only the median TTO values for low back pain showed statistically similar values when labels were used. Except for breast cancer and diabetes, the median responses for the valuation technique of person trade-off were higher for scenarios where labels were used. The authors speculated that this finding may be attributed to the fact that non-labelled scenarios were initially viewed by respondents as being more severe than when the same scenario was presented to them with labels. As such, by not providing a label in this study, a respondent may have associated the health states with a more severe condition (e.g., multiple sclerosis) during the valuation process; however, upon being informed that the states pertained to RA, the respondent may have increased their values due to the realization that the states pertained to a 'relatively benign' diagnosis. In either of the aforementioned scenarios (i.e., decreased or increased values), the presence of the labelling effect may have confounded the impact of the adaptation exercise on general population values. In hindsight, individuals in the Initially Uninformed Group should have been informed that the health states that they were valuing for the first time pertained to arthritis to alleviate the potential for any labelling effects. Alternatively, the adaptation exercise could have been designed (e.g., by editing the recorded scenarios) so that the actual disease was never mentioned. By doing this, the adaptation exercise can still be regarded as specific because the patients' experiences with adaptation will be still pertain to the health state under investigation. It would be interesting to investigate the differences between these two methods in a future study.

For this study, condition-specific health states were used in the investigation of informed general population values. Conversely, however, the guidelines from the National Institute for Health and Clinical Excellence (NICE) advise that generic health states (i.e., health states composed of dimensions from the EQ-5D), rather than condition-specific ones, should be used in an economic evaluation framework (National Institute for Health and Clinical Excellence, 2008). The question then arises how best to inform respondents about the 'generic' patient. While introspective methods could be used to elicit disease adaptation information (e.g., Ubel et al., 2005), this may not be truly informing respondents about the adaptation to a 'generic' health state. It would therefore appear that, in looking to inform respondents about disease adaptation, researchers may have to opt for condition-specific health states.

Three audio-recordings from the *Health Talk Online* website (DIPEx Health Experiences Research Group, 2008) were used to inform the general population respondents about adaptation to RA in this study. While it was considered advantageous to use actual patients discussing how they have dealt with their health condition, the information available on the aforementioned website may have been biased towards the positive end as its primary intention is to provide educational and supportive material for patients. The recorded scenarios therefore may not have fully addressed the entire range of adaptation issues for the respondents to consider. The inclusion of Lisa's recorded scenario aimed to highlight the struggles a patient may face, and hence provide a more complete picture of life in the described health states. However, an astute respondent may recognize that this patient was in her first year of having arthritis and that after some time, she may begin start making changes to her life to accommodate her illness (as discussed in Ann's recorded scenario). As a result, the adaptation exercise used may have portrayed a distorted picture of adaptation in RA; especially since the severity of the patients' condition do not align with the health state description. This may have influenced the respondents to inflate their initial health state values because RA adaptation was described in a positive light. This issue may have been rectified by actually conducting individual face-to-face interviews with RA patients that have adapted positively and adapted negatively to their health condition, and reporting this information back to the respondents. However, due to the logistics of collecting patient-level data (especially the lengthy review process to receive NHS ethical approval), it was felt that presenting the audio-recordings available from the *Health Talk Online* website was the best option. Incidentally, the presentation of simulated patient data was also briefly considered (i.e., using actors to promote different aspects of adaptation) but there was concern over the ethical implications of falsely informing the respondents with this type of ‘patient’ data.

The preceding discussion introduces the issue of what type of information *should* be presented to the general population to inform them about adaptation. Specifically, should a normative approach be taken (i.e., information about fully adapted patients using laudable coping techniques) or should a more comprehensive range of patient views (i.e., incorporating patients with differing degrees of adaptation using a combination of laudable and non-laudable techniques) be included? By including all forms of adaptation, respondents can make the assessment as to how they want to incorporate this information into their valuations. Furthermore, this would allow the influence of specific elements of the adaptation process (previously described in Section 2.2.1) on an individual’s health state values to be examined. Specifically, it would be useful to identify the different adaptation elements associated with patients living in a particular health condition and to ask general population respondents to what extent they want to use these adaptation elements in their valuations. This would provide information on whether health state values increased because the general population respondents incorporated either positive or negative elements of adaptation into their assessments. This raises the question of whether the level of resources for treatment should be adjusted because respondents have incorporated negative elements of adaptation – for example, lowering their health expectations – into their valuations. Conversely, it is also important to identify what aspects of the provided information have resulted in respondents keeping the same health perceptions but simply changing their values, possibly due to a better understanding of the valuation task or a desire to ‘match’ patient values. Developing a greater understanding of how information may influence health state values is important before adaptation exercises are used to contribute to healthcare resource allocation decisions.

The recorded scenarios chosen for the adaptation exercise do not align with the RA health state descriptions; this was a result of information regarding the patient’s severity not being available on the *Health Talk Online* website (DIPEx Health Experiences Research Group, 2008). The fact that the messages portrayed in the adaptation exercise did not appear to align with item levels in the health states is important, as it meant that the respondents had to consider the possibility of adaptation when living in an impaired health condition, regardless of the severity of the state itself. By using an extrospective approach, the recorded scenarios informed the respondents that patients do adapt to a specific chronic illness (i.e., RA) over time and the respondents then translated this information introspectively into their own health state valuations if they wished to do so. If the health states of the patients in the recorded scenarios had been stated and aligned with the health states being valued, then the respondents would not have been able to really draw upon other factors – such as personality traits and views of RA and illness, as identified from the qualitative interviews – that were important for them to receive the disease adaptation information and then apply it to their health state values. However, this mismatch between health states and recorded scenarios may have falsely informed the respondents that, independent of severity, adaptation to RA is achievable. This, in turn, would result in an uninformed increase in values after the respondents underwent the adaptation exercise.

It is also important to note that how the health state values were calculated may have had an effect on the study results. If a respondent provided more than one equal sign as the point of indifference for a RA state, the midpoint of the equal signs was used as the health state value, as previously described in Section 6.3.2. This, however, may not provide an accurate representation of the respondents' value for a particular health state because it does not reflect any uncertainty around their responses. As such, it would be necessary to go back to the raw data to assess how the respondent provided their indifference point for the three RA states. The range over which equal signs were used for each respondent could be used to evaluate this uncertainty. This would provide more information about the resolution of the responses; however, the mean value would still need to be used in evaluating whether a statistically significant change in the health state value was observed. This range of uncertainty is particularly important when the Informed Group, during the patient values presentation, viewed patient values that were different to their personal values. In this case, it is possible that the realization of the differences might cause a respondent to increase the range of uncertainty in his/her response rather than directly altering it. However, the potential impact of this effect was not investigated as the sole influence of the patient values presentation on the individual's values could not be deciphered as it was preceded by the adaptation exercise. This would be an area for future work if there is further interest in using a patient values presentation to provide information regarding disease adaptation.

Although the objective of the RCQ was to gain information about the complex rationales for why respondents changed their health state values, it may not have tapped into all the constructs. While valid and reliable instruments that measure personality and empathy levels could have been used, the RCQ was felt to be more advantageous, as it was based directly on information gained from the qualitative interviews (described in Chapter Seven). Rather than using a series of different instruments that measured personality traits and illness experience, the RCQ minimized the additional burden placed on the respondents when completing the entire quantitative exercise.

Using an external organization to contract an independent interviewer to conduct the data collection provided an objective way to obtain health state results. If the Ph.D. candidate had conducted the interviews, her desire for people to change their values might have been evident to the participants; as such, the participants' desire to please might have been much greater. The Ph.D. candidate did accompany the interviewer over the course of the data collection process to ensure quality and consistency in the interviews being conducted, but the participants were not made aware that the extra personal attending was the lead researcher, lest this bias their answers in any way. Instead, they were told that the additional person in the room was present to observe the interviews for training purposes.

8.6.5 Generalizability

This study included 200 participants recruited from two cities in the County of South Yorkshire; the participants were equally divided into the Initially Uninformed and Informed Groups. Although the responses may not necessarily represent those of the general population in the UK, the study sample was heterogeneous and encompassed a distribution of age ranges and sex. As this study was methodologically focused, the intention was to use the results to inform future data collection methods rather than to apply the results in practice.

8.7 Summary and Conclusions

While the determination of informed general population values is important from a policy perspective, there is currently an absence of empirical studies that explore this issue. As a result, this study appraised whether the adaptation exercise alone, and/or the patient values presentation preceded by the adaptation exercise, were effective at altering the initial values members of the general population have for RA states. In conjunction with the testing of the interventions, the factors which may influence an individual to change their initial health state values were determined in this study.

The findings obtained from this final component of the sequential mixed-methods study demonstrated that informed general population values can help to overcome the limitations of using only patient values or 'uninformed' general population values. As mentioned in Chapter Two, patients have direct knowledge about what it is like to live in the impaired health state; however, the use of their values in a decision-making context is potentially problematic for several reasons. For example, patients may have a vested interest in better funding for their particular disease. In other cases, patients may have adapted to their health condition through negative adaptation (Menzel et al., 2002), raising the question whether the level of resources for treatment should be adjusted because patients have 'adapted' to their condition by, for example, lowering their health expectations. On the other hand, because healthcare in the UK is publicly funded, the implication is that values from members of the general population (i.e., the tax payers) should be used. However, if these respondents are uninformed about the health states under investigation, the resulting health state values may not actually represent their preferences for health states, and as a result may be sub-optimal for guiding healthcare decisions.

Before the adaptation exercise is considered to be used in practice, there is a need to determine the economic importance of implementing such an intervention within a valuation study. Specifically, the health state values need to be entered into an economic model to establish the impact it has on the incremental cost-effectiveness ratio (previously described in Section 2.1). This will provide a meaningful interpretation of the magnitude of changes in health state values from a decision-makers' perspectives (Walters and Brazier, 2003 and 2005).

In conclusion, the results indicated that the use of an adaptation exercise encouraged individuals to change their initial values for RA states. On the other hand, the patient values presentation had a negligible effect on further change for participants who had already been informed through the adaptation exercise. Statistical tests and regression models revealed that an individual's age and current health status had a significant effect on the magnitude of change in the health state values. The results from this study contribute new information to the debate as to whose values should be considered when making decisions regarding healthcare, and the way informed general population values should be obtained; this discussion will be pursued in the following chapter.

8.8 Tables and Figures

Table 8.1: Summary of statistical tests of association to be conducted

Test	Initially Uninformed Group		Informed Group		Implication	Statistical Test	Null Hypothesis
	1st	2nd	1st	2nd			
1	✓	✓			Effect of the adaptation exercise on the values	Paired t-test	No difference between the 1st and 2nd values in the Initially Uninformed Group
2			✓	✓	Effect of patient values presentation on the values	Paired t-test	No difference between the 1st and 2nd values in the Informed Group
3		✓	✓		Effect of the interviewer on the values	Independent t-test	No difference between 2nd value of the Initially Uninformed Group and 1st value of the Informed Group
4	✓	✓	✓	✓	Comparing the effect of the adaptation exercise with the effect of the patient values presentation	Independent t-test	No difference between the change in the two values of Initially Uninformed Group and the change in the two values of Informed Group

Table 8.2: Characteristics of the study participants[a]

	Count (%)[b]		
	Initially Uninformed Group (n = 100)	**Informed Group (n = 100)**	**National Census[c]**
Males			
Younger than 30 years	9 (9)	13 (13)	10
30-39 years	10 (10)	8 (8)	9
40-49 years	9 (9)	9 (9)	9
50-59 years	8 (8)	6 (6)	8
60-69 years	6 (6)	6 (6)	6
Older than 70 years	6 (6)	6 (6)	6
Total	48	48	
Females			
Younger than 30 years	9 (9)	10 (10)	10
30-39 years	9 (9)	9 (9)	9
40-49 years	10 (10)	9 (9)	9
50-59 years	9 (9)	8 (8)	8
60-69 years	9 (9)	9 (9)	7
Older than 70 years	6 (6)	7 (7)	9
Total	52	52	
Education level			
Primary school	0	1 (1)	
Secondary school	54 (55)	63 (54)	
A-levels	14 (14)	12 (12)	
University	17 (17)	13 (13)	
Other (e.g. college)	14 (14)	9 (9)	
Employment status			
Self employment	7 (7)	6 (6)	
Paid employment	49 (49)	44 (44)	
Unemployed	8 (8)	9 (9)	
Retired	20 (20)	27 (27)	
Looking after home	5 (5)	5 (5)	
Student	1 (1)	1 (1)	
Disabled/long-term sick	10 (10)	8 (8)	
Illness experience			
Has arthritis	26 (26)	35 (35)	
Knows someone with arthritis	48 (48)	38 (38)	
Has chronic illness	12 (12)	9 (9)	
None	14 (14)	18 (18)	
EQ-5D index (mean ± SD)	0.768 (± 0.349)	0.765 (± 0.328)	
Interviewer questions			
The respondent seemed to understand the VAS well			
Strongly agree	24 (25)	24 (25)	

Agree	38 (39)	39 (41)
Neither agree nor disagree	20 (21)	19 (20)
Disagree	10 (10)	9 (9)
Strongly disagree	5 (5)	5 (5)
The respondent seemed to understand the TTO well		
Strongly agree	17 (18)	23 (24)
Agree	47 (48)	41 (42)
Neither agree nor disagree	21 (22)	22 (23)
Disagree	8 (8)	8 (8)
Strongly disagree	4 (4)	2 (2)
The respondent appeared to lose interest when valuing the latter health states		
Strongly agree	3 (3)	3 (3)
Agree	5 (5)	9 (9)
Neither agree nor disagree	23 (24)	21 (22)
Disagree	38 (39)	29 (30)
Strongly disagree	28 (29)	34 (35)
The respondent gave a lot of thought to the decisions that he/she was asked to make		
Strongly agree	15 (15)	13 (13)
Agree	24 (25)	31 (32)
Neither agree nor disagree	49 (51)	47 (48)
Disagree	7 (7)	5 (5)
Strongly disagree	2 (2)	0 (0)
The respondent was engaged with the patients' message in the recordings		
Strongly agree	20 (21)	19 (20)
Agree	18 (19)	24 (25)
Neither agree nor disagree	39 (40)	32 (33)
Disagree	15 (15)	17 (18)
Strongly disagree	5 (5)	4 (4)

[a] Reported in as a count unless otherwise indicated.
[b] There are some missing responses so count does not necessarily equate to frequency.
[c] UK Statistics Authority (2009). *Age structure of England and Wales* [online]. [Accessed 27 January 2009].

Table 8.3: Sex-age distribution of the three response sets

	Count (%)											
	Visual Analogue Scale						Time Trade Off					
	Initially Uninformed Group			Informed Group			Initially Uninformed Group			Informed Group		
	Complete Response Set	Less Strict Response Set	More Strict Response Set	Complete Response Set	Less Strict Response Set	More Strict Response Set	Complete Response Set	Less Strict Response Set	More Strict Response Set	Complete Response Set	Less Strict Response Set	More Strict Response Set
Males												
< 30	9 (18.8)	7 (17.5)	6 (16.2)	13 (27.1)	12 (27.9)	12 (30.8)	9 (18.8)	7 (16.3)	1 (7.7)	13 (27.1)	10 (23.3)	2 (13.3)
30-39	10 (20.8)	10 (25.0)	10 (27.0)	8 (16.7)	7 (16.3)	7 (17.9)	10 (20.8)	9 (20.9)	3 (23.1)	8 (16.7)	8 (18.6)	2 (13.3)
40-49	9 (18.8)	8 (20.0)	8 (21.6)	9 (18.8)	7 (16.3)	6 (15.4)	9 (18.8)	8 (18.6)	0	9 (18.8)	8 (18.6)	6 (0.4)
50-59	8 (16.7)	6 (15.0)	5 (13.5)	6 (12.5)	6 (14.0)	6 (15.4)	8 (16.7)	7 (16.3)	3 (23.1)	6 (12.5)	5 (11.6)	1 (6.7)
60-69	6 (12.5)	4 (10.0)	3 (8.1)	6 (12.5)	5 (11.6)	4 (10.3)	6 (12.5)	6 (14.0)	4 (30.8)	6 (12.5)	6 (14.0)	0
> 70	6 (12.5)	5 (12.5)	5 (13.5)	6 (12.5)	6 (14.0)	4 (10.3)	6 (12.5)	6 (14.0)	2 (15.4)	6 (12.5)	6 (14.0)	4 (26.7)
Total	48	40	37	48	43	39	48	43	13	48	43	15
Females												
< 30	9 (17.3)	9 (20.5)	8 (19.0)	10 (19.2)	9 (18.8)	9 (23.7)	9 (17.3)	7 (14.9)	3 (13.6)	10 (19.2)	9 (19.6)	2 (13.3)
30-39	9 (17.3)	7 (15.9)	7 (16.7)	9 (17.3)	8 (16.7)	8 (21.1)	9 (17.3)	9 (19.1)	3 (13.6)	9 (17.3)	7 (15.2)	2 (13.3)
40-49	10 (19.2)	10 (22.7)	10 (23.8)	9 (17.3)	8 (16.7)	5 (13.2)	10 (19.2)	8 (17.0)	5 (22.7)	9 (17.3)	7 (15.2)	3 (20.0)
50-59	9 (17.3)	7 (15.9)	6 (14.3)	8 (15.4)	6 (12.5)	4 (10.5)	9 (17.3)	9 (19.1)	4 (18.2)	8 (15.4)	8 (17.4)	3 (20.0)
60-69	9 (17.3)	5 (11.4)	5 (11.9)	9 (17.3)	9 (18.8)	8 (21.1)	9 (17.3)	9 (19.1)	4 (18.2)	9 (17.3)	8 (17.4)	4 (26.7)
>70	6 (11.5)	6 (13.6)	6 (14.3)	7 (13.5)	8 (16.7)	4 (10.5)	6 (11.5)	5 (10.6)	3 (13.6)	7 (13.5)	7 (15.2)	1 (6.7)
Total	52	44	42	52	48	38	52	47	22	52	46	15

Table 8.4: Health state values for all three response sets

Health States	Attempt	Initially Uninformed Group						Informed Group					
		Response Set						Response Set					
		Complete		Less Strict		More Strict		Complete		Less Strict		More Strict	
		Mean (SD)	Change	Mean (SD)	Change	Mean (SD)	Change	Mean (SD)	Change	Mean (SD)	Change	Mean (SD)	Change
Visual Analogue Scale													
Full health	1st	0.95 (0.87)	0.01	0.95 (0.07)	0.02	0.95 (0.65)	0.02	0.94 (0.12)	0.02	0.95 (0.10)	0.01	0.96 (0.09)	0
	2nd	0.97 (0.77)		0.97 (0.05)		0.97 (0.52)		0.96 (0.09)		0.96 (0.09)		0.96 (0.09)	
Your own health	1st	0.76 (0.25)	0.02	0.76 (0.25)	0.02	0.77 (0.25)	0.02	0.82 (0.33)	0.03	0.78 (0.20)	0	0.81 (0.18)	-0.01
	2nd	0.78 (0.22)		0.78 (0.23)		0.79 (0.23)		0.79 (0.20)		0.78 (0.20)		0.80 (0.18)	
Dead	1st	0.01 (0.05)	0	0.01 (0.05)	0	0.01 (0.06)	0	0.01 (0.06)	0	0.01 (0.07)	0	0.01 (0.06)	0
	2nd	0.01 (0.05)		0.01 (0.06)		0.01 (0.06)		0.01 (0.05)		0.01 (0.06)		0.01 (0.06)	
State X	1st	0.57 (0.20)	0.07	0.58 (0.18)	0.02	0.55 (0.18)	0.06	0.62 (0.32)	-0.01	0.58 (0.18)	0.02	0.60 (0.18)	0.01
	2nd	0.64 (0.19)		0.60 (0.18)		0.61 (0.17)		0.61 (0.20)		0.60 (0.18)		0.61 (0.18)	
State Y	1st	0.39 (0.19)	0.07	0.41 (0.18)	0	0.37 (0.19)	0.06	0.46 (0.30)	-0.02	0.41 (0.18)	0	0.41 (0.14)	0
	2nd	0.46 (0.19)		0.41 (0.16)		0.43 (0.17)		0.44 (0.20)		0.41 (0.16)		0.41 (0.16)	
State Z	1st	0.18 (0.23)	0.05	0.19 (0.17)	-0.01	0.14 (0.16)	0.06	0.25 (0.36)	-0.03	0.19 (0.17)	-0.01	0.17 (0.12)	0.01
	2nd	0.23 (0.19)		0.18 (0.15)		0.20 (0.14)		0.22 (0.21)		0.18 (0.15)		0.18 (0.15)	
Time Trade-off													
State X	1st	0.80 (0.25)	0.05	0.81 (0.25)	0.06	0.81 (0.21)	-0.01	0.86 (0.23)	-0.06	0.87 (0.24)	-0.03	0.89 (0.17)	0.01
	2nd	0.85 (0.23)		0.87 (0.22)		0.80 (0.30)		0.80 (0.33)		0.84 (0.28)		0.90 (0.16)	
State Y	1st	0.63 (0.33)	0.06	0.64 (0.32)	0.06	0.52 (0.27)	0.05	0.63 (0.46)	0.06	0.66 (0.42)	0.04	0.64 (0.32)	0.08
	2nd	0.69 (0.35)		0.70 (0.34)		0.57 (0.35)		0.69 (0.40)		0.70 (0.39)		0.72 (0.22)	
State Z	1st	0.25 (0.47)	0.16	0.25 (0.48)	0.17	0.10 (0.39)	0.08	0.33 (0.54)	0.08	0.36 (0.54)	0.06	0.19 (0.46)	0.07
	2nd	0.41 (0.50)		0.42 (0.50)		0.18 (0.45)		0.41 (0.52)		0.42 (0.52)		0.26 (0.42)	

Table 8.5: Effects sizes for the visual analogue scale and time trade-off values for all three response sets

Health States	**Effect Size**	
	Initially Uninformed Group	**Informed Group**
Complete Response Set		
Visual analogue scale		
State X	0.36	-0.07
State Y	0.39	-0.09
State Z	0.24	-0.12
Time Trade-off		
State X	0.21	-0.21
State Y	0.18	0.14
State Z	0.33	0.15
Less Strict Response Set		
Visual analogue scale		
State X	0.41	0.13
State Y	0.35	-0.02
State Z	0.24	-0.02
Time Trade-off		
State X	0.24	-0.12
State Y	0.18	0.10
State Z	0.35	0.11
More Strict Response Set		
Visual analogue scale		
State X	0.34	0.07
State Y	0.35	0.02
State Z	0.35	0.08
Time Trade-off		
State X	-0.04	0.06
State Y	0.16	0.29
State Z	0.19	0.16

Table 8.6: Health state values of the less strict response set

Health States	Valuation	Initially Uninformed Group: Health State Values: Mean (SD)	Difference[a]	Percentiles 5%	Median	95%	States worse than dead (count, %)	Informed Group: Health State Values: Mean (SD)	Difference[a]	Percentiles 5%	Median	95%	States worse than dead (count, %)
Visual Analogue Scale													
Full health	1st	0.95 (0.07)^{d}	0.02	0.80	1.00	1.00	---	0.95 (0.10)	0.01	0.75	1.00	1.00	---
	2nd	0.97 (0.05)^{d}		0.84	1.00	1.00	---	0.96 (0.09)		0.78	1.00	1.00	---
Your own health^{b}	1st	0.76 (0.25)^{c}	0.02	0.29	0.85	1.00	---	0.78 (0.20)	0	0.40	0.84	1.00	---
	2nd	0.78 (0.23)^{c}		0.31	0.87	1.00	---	0.78 (0.20)		0.39	0.83	1.00	---
Dead	1st	0.01 (0.05)	0	0	0	0.10	---	0.01 (0.07)	0	0	0	0.11	---
	2nd	0.01 (0.06)		0	0	0.09	---	0.01 (0.06)		0	0	0.06	---
State X^{b}	1st	0.55 (0.18)^{d}	0.07	0.26	0.55	0.86	1 (1.2)	0.58 (0.18)	0.02	0.15	0.58	0.88	1 (1.1)
	2nd	0.62 (0.17)^{d}		0.30	0.63	0.89	0 (0)	0.60 (0.18)		0.26	0.57	0.90	0 (0)
State Y^{b}	1st	0.37 (0.19)^{d}	0.06	0.03	0.38	0.63	2 (2.4)	0.41 (0.18)	0	0.12	0.41	0.67	1 (1.1)
	2nd	0.43 (0.17)^{d}		0.11	0.45	0.70	0 (0)	0.41 (0.16)		0.14	0.40	0.67	0 (0)
State Z^{b}	1st	0.16 (0.20)^{c}	0.04	0	0.12	0.45	3 (3.6)	0.19 (0.17)	-0.01	0	0.16	0.46	2 (2.2)
	2nd	0.20 (0.15)^{c}		0.01	0.18	0.54	0 (0)	0.18 (0.15)		0	0.16	0.46	3 (3.3)
Time Trade-off													
State X	1st	0.81 (0.25)^{c}	0.06	0.34	0.96	1.00	0 (0)	0.87 (0.24)	-0.03	0.14	0.98	1.00	0 (0)
	2nd	0.87 (0.22)^{c}		0.44	0.96	1.00	0 (0)	0.84 (0.28)		0.06	0.98	1.00	1 (1.1)
State Y	1st	0.64 (0.32)^{c}	0.06	0.02	0.72	0.98	3 (3.6)	0.66 (0.42)	0.04	-0.10	0.82	0.98	7 (7.7)
	2nd	0.70 (0.34)^{c}		0.11	0.80	0.98	3 (3.6)	0.70 (0.39)		-0.04	0.86	0.99	3 (3.3)
State Z	1st	0.25 (0.48)^{d}	0.17	-0.56	0.26	0.92	26 (31.0)	0.36 (0.54)^{d}	0.06	-0.74	0.42	0.98	24 (26.4)
	2nd	0.42 (0.50)^{d}		-0.52	0.54	0.98	20 (23.8)	0.42 (0.52)^{d}		-0.72	0.58	0.98	19 (20.9)

[a] Difference = 1st valuation – 2nd valuation
[b] Standardized health state values
Comparison of mean values (using paired t-tests): $^{c}p \leq 0.05$, $^{d}p \leq 0.01$

Table 8.7: Comparison of the Initially Uninformed and Informed Groups' second valuations relative to the patient values

Health State	Aggregate Health State Values			Frequency of Informed Group's Second Values Relative to Patient Values (count, %)[a]	
	Patients	Initially Uninformed Group	Informed Group	Less than Patient Value	Greater than Patient Value
State X	0.81	0.87	0.84	21 (23.6)	68 (76.4)
State Y	0.73	0.70	0.70	30 (33.7)	59 (66.3)
State Z	0.66	0.42	0.42	51 (57.3)[b]	37 (41.6)[b]

[a] Only the Informed Group was assessed to determine the affect of the patient values presentation on the respondents' health state values. Total sample size is 89 because less strict response set is used.
[b] One respondent provided the same value as the patient values for both attempts.

Table 8.8: Health state values of the Initially Uninformed Group to test the effect of the adaptation exercise (less strict response set)

Health State	**Valuation**	**Health State Values**	
		Mean (SD)	**Difference**[a]
Visual Analogue Scale			
Full health	1st	0.95 (0.07)[d]	0.02
	2nd	0.97 (0.05)[d]	
Your own health[b]	1st	0.76 (0.25)[c]	0.02
	2nd	0.78 (0.23)[c]	
Dead	1st	0.01 (0.05)	0
	2nd	0.01 (0.06)	
State X[b]	1st	0.55 (0.18)[d]	0.07
	2nd	0.62 (0.17)[d]	
State Y[b]	1st	0.37 (0.19)[d]	0.06
	2nd	0.43 (0.17)[d]	
State Z[b]	1st	0.16 (0.20)[c]	0.04
	2nd	0.20 (0.15)[c]	
Time Trade-off			
State X	1st	0.81 (0.25)[c]	0.06
	2nd	0.87 (0.22)[c]	
State Y	1st	0.64 (0.32)[c]	0.06
	2nd	0.70 (0.34)[c]	
State Z	1st	0.25 (0.48)[d]	0.17
	2nd	0.42 (0.50)[d]	

[a] Difference = 1st valuation – 2nd valuation
[b] Standardized health state values
Comparison of mean values (using paired t-tests): [c] $p \leq 0.05$, [d] $p \leq 0.01$

Table 8.9: Health state values of the Informed Group to test the effect of the patient values presentation (less strict response set)

Health State	Valuation	Health State Values	
		Mean (SD)	Difference[a]
Visual Analogue Scale			
Full health	1st 2nd	0.95 (0.10) 0.96 (0.09)	0.01
Your own health[b]	1st 2nd	0.78 (0.20) 0.78 (0.20)	0
Dead	1st 2nd	0.01 (0.07) 0.01 (0.06)	0
State X[b]	1st 2nd	0.58 (0.18) 0.60 (0.18)	0.02
State Y[b]	1st 2nd	0.41 (0.18) 0.41 (0.16)	0
State Z[b]	1st 2nd	0.19 (0.17) 0.18 (0.15)	-0.01
Time Trade-off			
State X	1st 2nd	0.87 (0.24) 0.84 (0.28)	-0.03
State Y	1st 2nd	0.66 (0.42) 0.70 (0.39)	0.04
State Z	1st 2nd	0.36 (0.54)[c] 0.42 (0.52)[c]	0.06

[a] Difference = 1st valuation – 2nd valuation

[b] Standardized health state values

Comparison of mean values (using paired t-tests): [c] $p \leq 0.01$

Table 8.10: The level of significance between the second valuation of the Initially Uninformed Group and the first valuation of the Informed Group to test the interviewer effect (less strict response set)

Health States	Second Valuation of Initially Uninformed Group	First Valuation of Informed Group	Difference[a]	P-Value
Visual Analogue Scale				
State X	0.62	0.58	0.04	0.12
State Y	0.43	0.41	0.02	0.40
State Z	0.20	0.18	0.02	0.25
Time Trade-off				
State X	0.87	0.87	0	0.93
State Y	0.70	0.66	0.04	0.54
State Z	0.42	0.36	0.06	0.47

Table 8.11: Changes in health state values to test the effect of the adaptation exercise *versus* the effect of the patient values presentation

Health States	Initially Uninformed Group	Informed Group	Between-Group Significance (p-value)
	Change in Values[a] Mean (SD)	Change in Values[a] Mean (SD)	
Visual Analogue Scale			
Full health	0.02 (0.05)	0.01 (0.05)	0.17
Your own health[b]	0.03 (0.12)	0 (0.07)	0.06
Dead	0 (0.07)	0 (0.04)	0.80
State X[b]	0.07 (0.17)	0.02 (0.16)	0.07
State Y[b]	0.06 (0.19)	0 (0.14)	0.01
State Z[b]	0.04 (0.18)	0 (0.12)	0.04
Time Trade-off			
State X	0.05 (0.26)	-0.03 (0.16)	0.02
State Y	0.06 (0.26)	0.03 (0.27)	0.37
State Z	0.17 (0.37)	0.06 (0.23)	0.02

[a] Change in values: Difference between the first and second valuation for each group.
[b] Standardized health state values

Table 8.12: Results from the principal components analysis of the Reasons to Change Questionnaire for Initially Uninformed Group [a]

RCQ Items	Principal Components							
	Label	1	2	3	4	5	6	7
POSOUTLOOK	Personality	0.75						
ADAPT		0.73						
FAMTIME		0.65						
NOTWEAK		0.65						
GOODQOL		0.58						
PAINOK		0.49						
FAMCOPE		0.44			0.42			
RECORDING	Information		0.76					
TRADEOFFDIFF			0.74					
UNDARTHDIS			0.72					
COVERUP	Recognition of coping strategies			0.82				
SELFCOPE				0.79				
PATCOPE				0.54				
NOTOLD	Opinions of arthritis				0.69			
UNDARTHPAT					0.61	0.48		
WORSEDIS					0.54			
IMAGINARTH	Empathy					0.69		
UNFAIR						-0.66		
OTHERSHOE						0.65		
FIRSTEXER	Ease with valuation exercises						0.89	
SECDEXER							0.85	
HLTHPRBSELF	Illness experience							0.83
HLTHPRBOTH								0.73

[a] Extraction methods: Principal component analysis. Rotation method: Varimax with Kaiser Normalization. Rotation converged in 7 iterations.

Table 8.13: Tukey's values for change in health state values for the Initially Uninformed Group

Variable	Tukey's Values				
	Mean	Median	Minimum	Maximum	Interquartile Range
Sex					
Male	0.08	0	-1.58	1.44	0.19
Female	0.07	0.04	-0.65	1.24	0.16
Presence of Chronic Condition					
No	0.11	0.04	-0.68	1.44	0.19
Yes	0.02	0	-1.58	1.40	0.11
Arthritis Experience					
No	0.06	0.10	-1.58	0.73	0.24
Yes	0.08	0	-0.65	1.44	0.14
Age					
< 30	0.15	0.08	-0.33	1.44	0.26
30-59	0.08	0.02	-1.08	1.40	0.15
≥ 60	0.03	0	-1.58	1.04	0.12
EQ-5D Index					
> 0.95	0.11	0.04	-0.68	1.12	0.20
0.65 – 0.95	0.06	0.04	-1.58	1.40	0.15
< 0.65	0.02	0	-0.40	1.44	0.08
RA States					
State X	0.06	0	-1.58	0.96	0.14
State Y	0.06	0.02	-1.08	0.92	0.16
State Z	0.11	0.08	-0.65	1.44	0.21
Valuation Method					
VAS	0.06	0.03	-0.65	0.73	0.19
TTO	0.09	0	-1.58	1.44	0.16

Table 8.14: Univariate analyses for model assessing changes in health state values for the Initially Uninformed Group

Explanatory Variables	**Model Fit**		**Individual Estimates**		
	F-test	**p-Value**	**Coefficient**	**Standard Error**	**p-Value**
Health States (referent group = State Z)	1.98	0.14			
State X			-0.048	0.027	0.07
State Y			-0.045	0.027	0.10
Valuation Method (referent group = VAS)	2.20	0.14			
TTO			0.033	0.022	0.14
Sex (referent group = male)	0.07	0.79			
Female			-0.006	0.022	0.79
Age (referent group = less than 30 years)	5.76	< 0.01			
30-59 years			-0.068	0.030	0.02
Over 60 years			-0.115	0.034	< 0.01
Current Health Status (referent group – EQ-5D < 0.65)	5.31	< 0.01			
EQ-5D > 0.95			0.087	0.028	< 0.01
EQ-5D 0.65-0.95			0.037	0.033	0.26
Illness Experience (referent group = no illness experience)	3.41	0.02			
Has arthritis			-0.083	0.034	0.02
Knows someone with arthritis but has no direct illness experience			-0.007	0.024	0.78
Has chronic illness but not arthritis			-0.068	0.032	0.03
Components of the Reasons to Change Questionnaire					
Personality			0.029	0.014	0.04
Information			0.033	0.013	0.01
Recognition of coping strategies			0.018	0.011	0.10
Opinions of arthritis			0.013	0.010	0.20
Empathy			-0.001	0.012	0.90
Ease with valuation exercises			0.027	0.010	< 0.01
Illness experience			-0.017	0.012	0.13

Table 8.15: Main effects model assessing changes in health state values for the Initially Uninformed Group

Variables	Estimates		
	Coefficient	Standard Error	p-Value
Constant	0.106	0.037	< 0.01
Age			
30-59 years	-0.061	0.030	0.05
Over 60 years	-0.102	0.035	< 0.01
Current health status			
EQ-5D > 0.95	0.062	0.029	0.03
EQ-5D 0.65 – 0.95	0.017	0.033	0.62
Recognition of coping strategies	0.022	0.011	0.05
Ease with valuation exercises	0.025	0.010	0.01

$R^2 = 0.05$, $F = 4.75$, $p < 0.01$

Table 8.16: Model consisting of main effects and interaction terms predicting changes in health state values for the Initially Uninformed Group

Variables	Estimates		
	Coefficient	Standard Error	p-Value
Constant	0.091	0.037	0.01
Age			
30-59 years	-0.059	0.030	0.05
Over 60 years	-0.112	0.034	< 0.01
Current health status			
EQ-5D > 0.95	0.062	0.028	0.03
EQ-5D 0.65 – 0.95	0.012	0.033	0.71
Recognition of coping strategies	0.020	0.011	0.05
Ease with valuation exercises	0.026	0.010	0.01
EQ-5D > 0.95 * Opinions of arthritis	0.027	0.012	0.02
State Z * TTO	0.114	0.028	< 0.01

$R^2 = 0.09$, $F = 6.76$, $p < 0.01$

Figure 8.1: Scree plot for the responses to the Reasons to Change Questionnaire for the Initially Uninformed Group

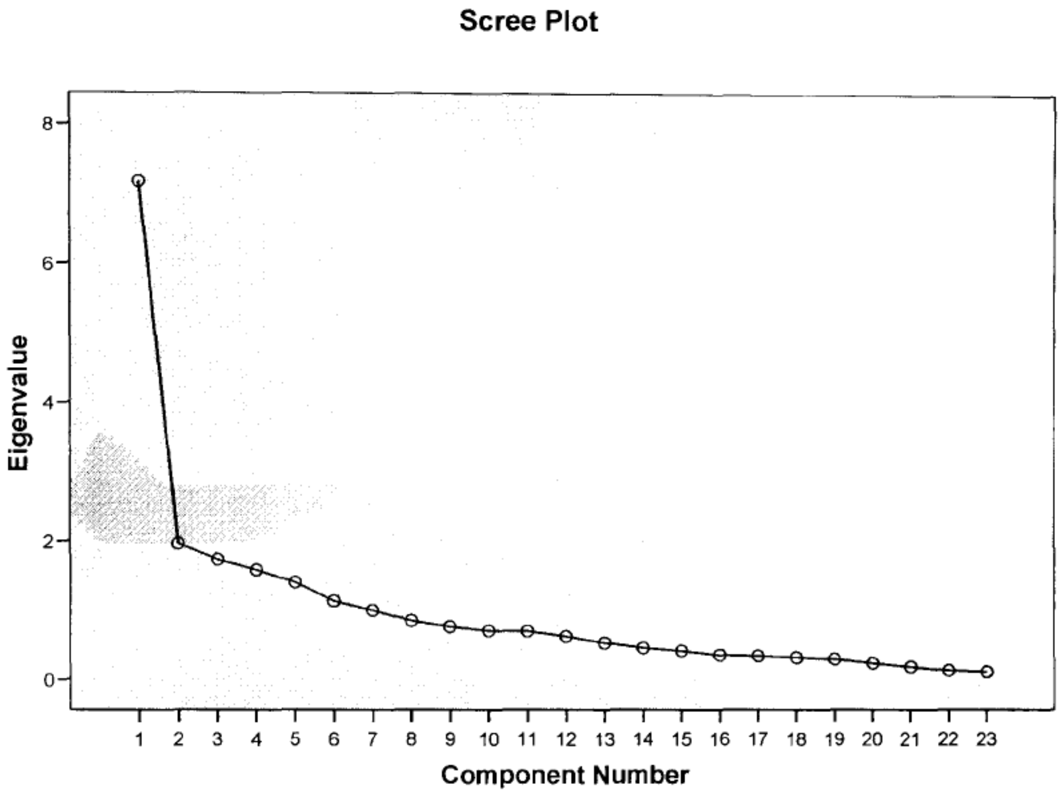


Figure 8.2: Histogram for changes in health state values for the Initially Uninformed Groups

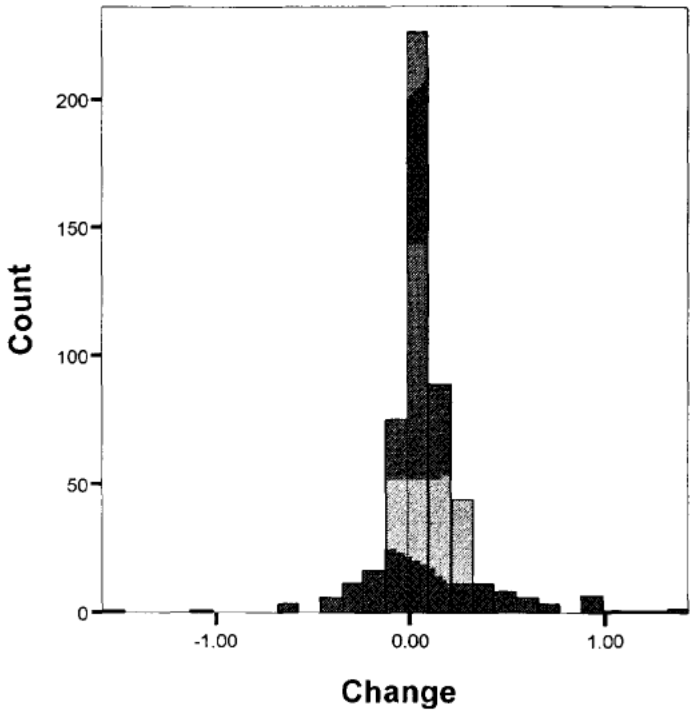
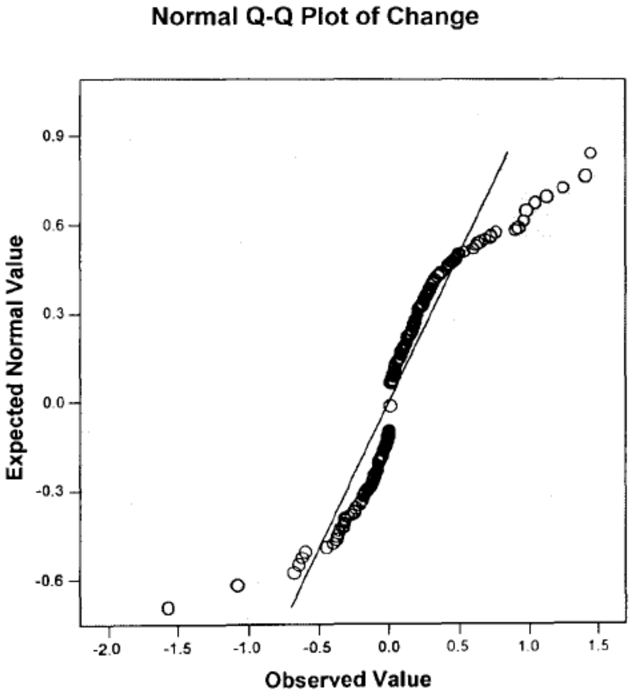


Figure 8.3: Quantile-quantile plot for the changes in health state values for the Initially Uninformed Group



Chapter Nine

General Discussion and Conclusions

The main objective of this thesis was to understand how informing general population respondents about disease adaptation influenced their valuations for rheumatoid arthritis (RA) states. This objective has been addressed through a multi-component sequential mixed-methods study design. The first component involved the development of health states describing RA symptoms based on patient-level data (Chapter Five). The second component used qualitative interviews and the developed health states to explore individuals' perceptions of RA after being provided with disease adaptation information (Chapter Seven). On the basis of the results from this qualitative component, a Reasons to Change Questionnaire (RCQ) was developed for use in the third phase of this investigation. This final component used quantitative methodologies to determine the magnitude of change in health state values after informing respondents about disease adaptation, and to identify the factors contributing to this change (Chapter Eight).

The current chapter offers a general overview of the most significant findings of the research, and provides an interpretation of these results in the context of the construction of informed general population values. The originality and the research contributions of the study findings are discussed. Finally, recommendations for future research to develop an even greater understanding of the construction of informed general population values are presented.

9.1 Summary of Key Results

The results from this thesis provide significant and novel insights into the development of informed general population values for use in a healthcare decision-making context. Specifically, this thesis has generated the following key outcomes:

- Plausible and comprehensible health states were developed using a combination of Rasch and cluster analyses;

- The innovative adaptation exercise was shown to affect members of the general population's perceptions and valuations for RA states; and
- Individuals, who were younger in age and in better health, were found to change their initial health state values by a larger margin after being informed about disease adaptation than those who were older and in poorer health.

Each of the aforementioned outcomes is discussed below.

9.1.1 The Development of Health States

The joint use of Rasch and cluster techniques was successful at producing objective health states using patient responses to a unidimensional condition-specific instrument. Rasch analysis identified the most representative items of the unidimensional Stanford Health Assessment Questionnaire (HAQ) that could be combined with the pain and discomfort dimension of the EuroQol-5D (EQ-5D). By applying *k*-means cluster analysis to the responses to the items pre-selected for the health state description, item levels were grouped to form RA states. In general, the combination of Rasch and cluster analyses is a novel and effective way for researchers to create plausible and comprehensible health states tailored to the needs of valuation studies.

The use of this novel approach afforded three condition-specific states to use in exploring the research question. By combining the HAQ with the pain and discomfort dimension of the EQ-5D, the health states clearly described the physical limitations that are a key part of the impact of RA on patients. When the participants valued the three states developed for this study, they carefully considered each item and traded off between them; this provides further evidence that the respondents found the range of choices meaningful and adequate.

9.1.2 The Influence of the Adaptation Exercise on Health State Values

The adaptation exercise developed for this thesis was demonstrated to both inform the general population about disease adaptation and encourage them to reflect upon this information. First implemented in the qualitative component, the adaptation exercise was shown to influence individual respondents' perceptions of RA. The fact that participants discussed the effects the recorded scenarios had on them in the qualitative interviews provides further evidence of the suitability of the adaptation exercise as a tool for informing respondents about disease adaptation.

The capability of the adaptation exercise to inform the general population respondents about disease adaptation was further confirmed by the results obtained from the large-scale quantitative component of this work. Statistically significant differences were detected between the first and second valuations of the Initially Uninformed Group for all RA states (Table 8.8, page 208); this indicated that the adaptation exercise was effective. Furthermore, conducting a cross-group comparison with the values subjected to the adaptation exercise (i.e., the second value from the Initially Uninformed Group and the first value from the Informed Group) supported the view that respondents were not significantly influenced by the interviewer (Table 8.10, page 210). Thus, it is reasonable to use a comparison of the two values provided by the Initially Uninformed Group to assess the effect of the adaptation exercise.

While the adaptation exercise induced individual respondents to alter their health state values, the patient values presentation had significantly less impact on these values (Table 8.9, page 209). This is, perhaps, due to respondents in the Informed Group having both undergone the adaptation exercise and completed their first valuation before viewing the patient values. As a result, the patient values presentation may not have provided any further, or different, insight into what life is like for a patient with RA. Similarly, the qualitative results demonstrated that the patient values presentation had a minimal effect on the participants' perceptions of RA as it influenced only two of the five identified themes (Figure 7.1, page 163): viewing patient values only affected individual's attitudes toward coping and their desire to live a long life, while the adaptation exercise played a role in all five themes.

The minimal influence of the patient values presentation on individual respondent's health state values was further confirmed by the magnitude of the second values provided by the Informed Group (i.e., values provided after being subjected to the patient values presentation). These results demonstrated that the respondents did not adjust their values for the sake of simply trying to replicate the patient values. Individuals in this group were giving their own values for the hypothetical states, and were not trying to simply please the interviewer or adopt a 'patients know best' perspective by replicating the values presented. The findings in the qualitative component also aligned with those obtained in the large-scale quantitative component. Individuals, after hearing the patients' messages, understood that adaptation was possible but they did not want to match the patient values because they felt that they did not fully understand the implication of living with the condition. This finding suggests that the normative issue of 'whose values should be used in a decision-making context', as highlighted in Chapter Two, might be ameliorated by the use of informed general population values.

9.1.3 Factors Found to Influence Change in Health State Values

The results from the qualitative and quantitative components revealed that an array of factors caused individuals to change their health state values after being informed about disease adaptation. Specifically, the qualitative results suggested that, after being informed about adaptation and reflecting on that information, participants were more likely to consider adaptation if they were able to empathize with the patients' messages in the adaptation exercise; this also resulted in these individuals having more favourable perceptions of the RA states. In addition, the adaptation exercise helped individuals to feel that they could both better understand, and better adapt to, the chronic health condition: it encouraged them to reflect on their experience of RA amongst family members and friends; it showed them that they could draw on others for support if they had RA; and it underlined the benefit of having a positive attitude towards life.

The findings from the qualitative interviews suggested that the recorded scenarios tapped into the respondents' emotions such as empathy regarding health and illness. This raises questions – beyond the scope of this thesis – of whether emotion, or which of its various types, should be considered in the valuation of health states. However, as will be discussed below, when the RCQ principal component results from the qualitative interviews were subjected to quantitative analysis, the 'Empathy' component did not significantly influence individuals to alter their health state values. This suggests that, while analysis of the qualitative interviews revealed that emotion was important, it did not actually contribute significantly in a consistent manner to their decisions regarding the valuations of the health states.

In general, comparing the findings from the qualitative interviews and the large-scale quantitative component showed similarities in terms of the importance of being informed about disease adaptation. Similarities included individual's recognition of coping strategies and an improved opinion of life with arthritis affecting perceptions and valuations. However, some contradictions were uncovered, principally in the variable pertaining to an individual's current health status (i.e., an individual's EQ-5D index) and the aforementioned variable pertaining to the 'Empathy' principal component. This 'inter-method discrepancy' is not unusual as bringing different methods together almost always raises differences in findings and their interpretations (Moffat et al., 2006).

Analysis of the qualitative interviews revealed that individuals who were healthy – without any experience with illness – and individuals who were not empathetic struggled to engage with the recorded scenarios. On this basis, healthy and non-empathetic individuals were hypothesized to be less likely to change the level of their health values, even after being informed about disease adaptation. Contrary to these expectations, the univariate analyses demonstrated that healthy individuals (i.e., EQ-5D index > 0.95) increased their health state values more than those individuals in poorer health; while empathy did not have a significant role in influencing individuals to alter their health state values.

A multivariate linear regression was used to examine the combination of factors that predict change in health state values (Table 8.15, page 215). The results indicated that the individual most likely to increase his/her health state value by the largest margin is someone who is under 30 years of age, who is in good health (i.e., an EQ-5D index > 0.95), who recognizes the need for coping strategies, who found the valuation exercises easy to complete, and who had an improved opinion of RA. This may be a result of individuals with the aforementioned characteristics providing lower health state values for the initial valuation and therefore, they had further to change during the second valuation.

9.2 Methodological Contributions

While the key findings outlined in the previous section met the objectives outlined at the start of this thesis, these findings also contribute to the existing literature in terms of the methodology use in the construction of informed general population values. The methodological contributions are described below.

9.2.1 The Study Design

Two groups of participants were used, primarily to detect the presence of prevarication and interviewer biases. The design of the study was considered to be pivotal in revealing the effectiveness of the adaptation exercise. As discussed in earlier chapters, using only one group of participants would have made it difficult to examine the true effect of the intervention (i.e., adaptation exercise). The results from the quantitative component demonstrated that prevarication and interviewer biases were unlikely and, therefore, the influence of the adaptation exercise on health state values can be effectively assessed using the results from only one participant group.

This is not the first study to have adopted a design with two participant arms. Ubel et al. (2003) used two groups of participants to examine the utilization of an intervention to encourage respondents to defocus from the specifics of the health condition when valuing states relating to paraplegia and below the knee amputation. Ubel et al. (2003) state that the cross-comparison aimed to alleviate any potential anchoring effects (e.g., when providing a second valuation, respondents may attach it to their initial values); their results showed that such effects were unlikely. This rationale is very different from the reasons for using the two groups in the current study which, as mentioned previously, focused on investigating and avoiding the potential for prevarication and interviewer biases.

9.2.2 The Adaptation Exercise

This thesis demonstrated the novel use of condition-specific audio-recordings from patients to describe concepts of disease adaptation. Previous studies have also used adaptation exercises (Ubel et al., 2005; Damschroder et al., 2005 and 2008), but the methods used were fundamentally different from those reported here. The previous studies adopted an introspective approach in encouraging respondents to consider the possibility of adaptation (e.g., think back to a previously difficult time in your life and consider how your feelings toward this event changed over time). The current adaptation exercise adopted a combination of both introspective and extrospective techniques. By using recorded scenarios, respondents were encouraged to consider aspects of adaptation first externally, and then to internally consider this information. After this deliberation process, they either incorporated this information into their own valuations of the RA states or they disregarded it as they preferred.

Providing an adaptation exercise to respondents in the general population prior to their valuing health states ensures that members of the general population are starting from approximately the same initial level of knowledge regarding the disease state of interest. If no information is provided to the respondents, some respondents from the general population will have some knowledge about the health states under investigation, while others will not. As a result, there are varying levels of knowledge used to guide decisions regarding the allocation of healthcare resources. While using the adaptation exercise to inform general population respondents does not ensure that everyone is starting at exactly the same point, the possibility of complete unawareness of the health state should, to all intents and purposes, be eliminated.

The study results indicated that the respondents were much more responsive to the adaptation exercise than to a patient values presentation, when the latter was preceded by the adaptation exercise. This provides an indication that, if the adaptation exercise was to be utilized for future valuation studies, only one valuation would be needed. Respondents would first listen to the recorded scenarios and would then discuss and reflect upon the information, before completing the valuation tasks. Eliminating one of the valuations will decrease both the resources required (e.g., interviewer time and questionnaire printing costs) and the time needed for the respondents to value the health states of interest.

9.2.3 General Contributions

While this study was conceived and conducted in the United Kingdom (UK), the results presented in this thesis may be of significance elsewhere in the world. First, the results may be beneficial to researchers and decision-makers in the United States (US), as the Washington Panel for Cost-effectiveness in Health and Medicine proposes the use of informed general population values, rather than patient values, in the context of healthcare decision-making. In addition to the US, other countries – for example, Canada and Australia – with health technology assessment organizations that adopt similar guidelines to those proposed by the UK's National Institute for Health and Clinical Excellence may also have an interest in the thesis results.

Furthermore, while RA was used as a case study for this study, the methods used to inform the general population, and its results, are meaningful to conditions that have physical disabilities like those of RA (e.g., painful and swollen joints). Such health conditions include, for example, other forms of arthritis, sport injuries, and back pain. It is, however, anticipated that more work is needed to develop a condition-specific adaptation exercise for complex health issues that target multiple domains.

9.3 Recommendations and Future Work

This thesis demonstrated a novel approach to deriving informed general population values. The results, presented in the earlier chapters of this thesis, have the potential to guide future research aimed at optimizing the development of informed general population values. There is, however, a need to address the validity, generalizability, and usability of the adaptation exercise.

9.3.1 Validity of the Adaptation Exercise

It is important that the validity of the adaptation exercise used to inform the general population be more carefully examined. Future research could involve incorporating life domains that remain at the same level or may even be enhanced (e.g., personal and spiritual relationships) in the health state descriptions. Incorporating aspects such as these could provide a more comprehensive description of the health state. Furthermore, it would be interesting to investigate the potential of explicitly incorporating social functioning as an additional item in the health state description and to compare it with the results from this current study. Results from such a study would indicate whether the respondents have indeed gained a better insight into adaptation.

Different forms of adaptation exercises should also be investigated. For example, it would be interesting to compare what differences in values occur if videos or interview transcripts of patients describing how they have adapted to their impaired health states were used in place of the audio-recordings in this study. Audio-recordings may not be suitable to everyone since individuals have different mechanisms to receive new information.

Although the independent effect of the patient values presentation was not an objective of this thesis, another potential area of interest would be to study its influence separately from any other form of adaptation exercise. In the present study, the effect of viewing the patient values was revealed to be negligible, when combined with the adaptation exercise. However, it would be useful to determine if respondents can understand from a simple presentation of numerical patient values that adaptation to a condition is possible. This would, most likely, require some (numerical) demonstration of the adaptation process that patients undergo; such information is lacking from the current literature. Furthermore, solely using a patient values presentation to inform the general population raises the significant concern that respondents may anchor on the numerical health state value rather than incorporating the underlying information into their assessment.

9.3.2 Generalizability of the Adaptation Exercise

Future research will doubtless move beyond this fundamental level to explore a wide range of illnesses. For example, a condition-specific adaptation exercise for use in more complex health issues, such as those that involve significant changes in mental health conditions, would be useful. In this case, it would be very interesting to get members of the general population to value a series of mental health states to determine if a change occurs following an adaptation exercise. (It is possible in this example that a change would occur in the negative direction.)

9.3.3 Usability of the Adaptation Exercise

Before an adaptation exercise may be implemented in practice, there are several issues that need to be addressed. These include informing the general population about 'generic' adaptation, obtaining informed values from the 'appropriate' members of the general population, and incorporating these informed values into a cost-effectiveness analysis. By addressing these issues, the use of adaptation exercises in future valuation studies may be considered.

As mentioned in earlier chapters of this thesis, guidelines proposed by the National Institute for Health and Clinical Excellence (NICE) advise that generic health states (i.e., health states composed of EQ-5D dimensions), rather than condition-specific states, should be used in the economic evaluation of healthcare technologies (National Institute for Health and Clinical Excellence, 2008). Therefore, future efforts should focus on developing methods to inform the general population respondents about adaptation to these generic states. One possible option may be to adopt introspective methods previously reported in the literature (Ubel et al., 2005; Damschroder et al., 2005 and 2008); however, there are drawbacks to using this approach. First, when respondents, who are prompted to think about a previous personal health event, may focus on a condition that is very different from the health state itself; the resulting value may be influenced by a potential labelling effect, as previously discussed in Section 8.6.4. Second, the use of an introspective method may make informing the respondents about the different severities of the generic health state difficult, especially when there is a need to distinguish between abilities to adapt to different generic states. As a result, there is a need to develop, and validate, a generic adaptation exercise to afford values for generic health states. This is significant from a policy perspective as using these generic 'adapted' values in an economic evaluation framework would permit comparison across different diseases.

The results from this study revealed that different types of individuals yield different health state values after being informed about disease adaptation. For example, Section 8.5.5 demonstrated that those who are younger and in better health are more likely to increase their values by a larger margin than those who are older and in poorer health. This provides indication that the general population may provide different adapted values than patients would, in part because they represent a different demographic group. Therefore, further work is needed to address the type of respondents from the general population that *should* be recruited to participate in valuation studies. That is, should these respondents be 'matched' to ensure their personal characteristics (e.g., age, gender) correspond to the proportion of patients with these same characteristics? While this strategy seems reasonable, such a matched general population sample will impair the comparability across different patient groups and healthcare technologies. In an attempt to investigate the impact of respondent type on health state, an examination of aggregate values obtained from both representative and 'matched' samples from the general population is needed.

Finally, there is also a need to incorporate informed general population values into a cost-effectiveness analysis (CEA), work which was beyond the scope and resources of this thesis. Going forward, it is necessary to obtain these figures and to compare them with those obtained using patient and 'uninformed' general population values. However, this may not be a simple exercise of populating existing CEAs with the informed values and examining its impact on the incremental cost-effectiveness ratios. Decisions will need to be made as to how best to assess the impact these values on a CEA. Should the respondent be asked to value health states after being informed about a fully adapted patient? Or, should the respondent be informed about a series of events – onset of disease, during the adaptation process, and after a period of adaptation – and be asked to provide a value for each of these events? In the latter scenario, the theoretical model of the quality-adjusted life year (QALY) will need to be re-considered. By calculating individual QALYs for each of the event (e.g., disease onset, adaptation process, full adaptation) (Equation 2.2), quantity and quality of life can no longer be regarded as utility independent. This could significantly impact the standard practice of using tariffs or valuation sets (e.g., EQ-5D) in economic evaluation of healthcare technologies.

By conducting work in the following areas – informing the general population about 'generic' adaptation, obtaining informed general population values, and incorporating these informed values into a cost-effectiveness analysis – important contributions to the emerging field of developing better informed general population values can be made. This should ultimately lead to more comprehensive information of health state valuations that can be used to provide fairer and more efficient decisions regarding the allocation of healthcare resources.

9.4 Conclusions

This thesis demonstrates that the use of a multi-component mixed-methods study was effective at meeting the research objectives. The study involved three components, developed and implemented in a sequential fashion: (i) Rasch and cluster techniques to form RA health states; (ii) qualitative interviews to determine the effect of the disease adaptation on general population's perception of RA; and (iii) quantitative methods to investigate the magnitude of change in health state values achieved by informing respondents about disease adaptation, and the factors contributing to this change. In combination, these components demonstrate that respondents from the general population do change their initial health state values when informed about disease adaptation.

Overall, the results from this thesis provide a unique contribution to knowledge. First, the use of the two respondent groups in the study design evaluated the presence of prevarication and interviewer biases and found them to be negligible. This led to the conclusion that the observed changes in health state values were a result of the novel adaptation exercise, hence demonstrating that this method was effective at informing members of the general population about disease adaptation. Second, the use of recorded scenarios and deliberation in the adaptation exercise was novel in that it combined introspective and extrospective techniques in advancing concepts of disease adaptation. Third, the effects of informing respondents about adaptation have been shown to result in significant changes in respondent valuations for health states. Finally, the presented results can be applied to other health condition with symptoms similar to that of RA.

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Appendices

Appendix A.1: List of Dissemination from the Results of the Thesis

Research Grants and Awards

- Brazier J.E., Tsuchiya, A., O'Cathain, A., Dieppe, P.A., and **McTaggart-Cowan, H.M.** General population values for rheumatoid arthritis states: Making them better informed. Arthritis Research Campaign. 2008.
  Role: Prepared entire research grant.
- Canadian Institutes for Health Research, Doctoral Research Award, 2007-2010.
- International Society for Quality of Life Research, New Investigator Scholarship, 2009.

Chapter Five: The Development of Rheumatoid Arthritis States Using Rasch and Cluster Analyses

A version of this chapter is:

- Published as **McTaggart-Cowan, H.**, Brazier, J., and Tsuchiya, A. (2008). Combining Rasch and cluster analyses: A novel method to develop rheumatoid arthritis health states. *Health Economics and Decision Sciences Discussion Paper*, 08/15.
- Under review in *Value in Health* as **McTaggart-Cowan, H.M.**, Brazier, J.E., and Tsuchiya, A. (2008). Clustering Rasch results: A novel method to develop rheumatoid arthritis health states.

Chapter Seven: A Qualitative Exploration of how Disease Adaptation Information Affects the General Population's Perceptions of Rheumatoid Arthritis

A version of this chapter has been:

- Presented as **McTaggart-Cowan, H.**, O'Cathain, A., Tsuchiya, A., and Brazier, J. (2008). Understanding why the general population may or may not change their values in hypothetical health state valuation: A qualitative study using rheumatoid arthritis states at the *2008 Joint Meeting of the UK Health Economists' Study Group and the Nordic Health Economists' Study Group meeting* in Aberdeen.
- Published as **McTaggart-Cowan, H.**, O'Cathain, A., Tsuchiya, A., and Brazier, J. (2009). A qualitative study exploring the general population's perception of rheumatoid arthritis after being informed about disease adaptation. *Health Economics and Decision Sciences Discussion Paper*, 09/02.

Chapter Eight: A Quantitative Assessment of the Influence of Disease Adaptation Information on Values for Rheumatoid Arthritis States

A version of this chapter has been:

- Presented as **McTaggart-Cowan, H.M.**, Tsuchiya, A., O'Cathain, A., and Brazier, J.E. (2009). The impact of disease adaptation on general population values at the *International Society for Pharmacoeconomics and Outcome Research* in Orlando, USA.

- Presented as **McTaggart-Cowan, H.,** Tsuchiya, A., O'Cathain, A., and Brazier, J. (2009). To change or not to change: Understanding the factors that influence the general population to alter their rheumatoid arthritis values. *2009 Health Economists' Study Group meeting* in Sheffield.

- Presented as **McTaggart-Cowan, H.M.**, Tsuchiya, A., O'Cathain, A., and Brazier, J.E. (2009). To change or not to change: What influences the general population to alter their values for rheumatoid arthritis states? *2009 International Society for Quality of Life Research meeting abstracts* in New Orleans, USA.

Appendix A.2: The EuroQol-5D

Your own health state today

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 dress or wash 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

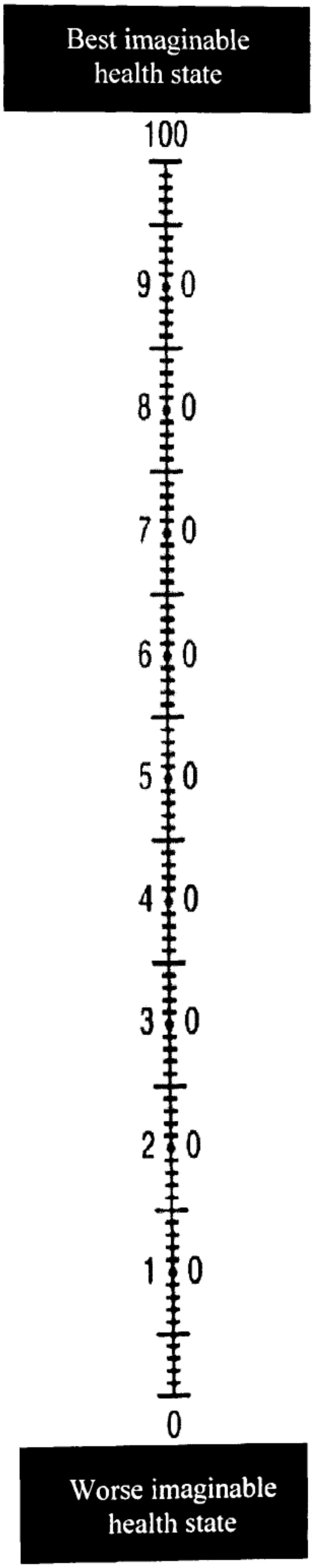
☐ I am not anxious or depressed
☐ I am moderately anxious or depressed
☐ I am extremely anxious or depressed

Your own health state today

To help people say how good their health is, we have drawn a scale 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.

Your own
health state
today



Appendix A.3: The Stanford Health Assessment Questionnaire (abridged)

HEALTH ASSESSMENT QUESTIONNAIRE©
Stanford University School of Medicine
Division of Immunology & Rheumatology

Name ______________________________ **Date** ___________

In this section we are interested in learning how your illness affects your ability to function in daily life. Please feel free to add any comments on the back of this page.

Please check the response which best describes your usual abilities OVER THE PAST WEEK:

	Without ANY difficulty^0	With SOME difficulty^1	With MUCH difficulty^2	UNABLE to do^3
DRESSING & GROOMING				
Are you able to:				
-Dress yourself, including tying shoelaces and doing buttons?	☐	☐	☐	☐
-Shampoo your hair?	☐	☐	☐	☐
ARISING				
Are you able to:				
-Stand up from a straight chair?	☐	☐	☐	☐
-Get in and out of bed?	☐	☐	☐	☐
EATING				
Are you able to:				
-Cut your meat?	☐	☐	☐	☐
-Lift a full cup or glass to your mouth?	☐	☐	☐	☐
-Open a new milk carton?	☐	☐	☐	☐
WALKING				
Are you able to:				
-Walk outdoors on flat ground?	☐	☐	☐	☐
-Climb up five steps?	☐	☐	☐	☐

Please check any AIDS OR DEVICES that you usually use for any of these activities:

☐ Cane
☐ Walker
☐ Crutches
☐ Wheelchair
☐ Devices used for dressing (button hook, zippers, long-handled shoe horn, etc.)
☐ Built up or special utensils
☐ Special or built up chair
☐ Other (Specify: ________________)

Please check any categories for which you usually need HELP FROM ANOTHER PERSON:

- ☐ Dressing and Grooming
- ☐ Arising
- ☐ Eating
- ☐ Walking

Please check the response which best describes your usual abilities **OVER THE PAST WEEK:**

	Without ANY difficulty^0	With SOME difficulty^1	With MUCH difficulty^2	UNABLE to do^3
HYGIENE				
Are you able to:				
-Wash and dry your body?	☐	☐	☐	☐
-Take a tub bath?	☐	☐	☐	☐
- Get on and off the toilet?	☐	☐	☐	☐
REACH				
Are you able to:				
-Reach and get down a 5-pound object (such as a bag of sugar) from just above your head?	☐	☐	☐	☐
-Bend down to pick up clothing from the floor?	☐	☐	☐	☐
GRIP				
Are you able to:				
-Open car doors?	☐	☐	☐	☐
-Open jars which have been previously opened?	☐	☐	☐	☐
-Turn faucets on and off?	☐	☐	☐	☐
ACTIVITIES				
Are you able to:				
-Run errands and shop?	☐	☐	☐	☐
-Get in and out of a car?	☐	☐	☐	☐
-Do chores such as vacuuming or yardwork?	☐	☐	☐	☐

Please check any AIDS OR DEVICES that you usually use for any of these activities:

☐ Raised toilet seat
☐ Bathtub seat
☐ Jar opener (for jars previously opened)
☐ Bathtub bar
☐ Long-handled appliances for reach
☐ Long-handled appliances in bathroom

☐ Other (Specify: ________________)

Please check any categories for which you usually need HELP FROM ANOTHER PERSON:

- ☐ Hygiene
- ☐ Reach
- ☐ Gripping and opening things
- ☐ Errands and chores

Appendix A.4: Health State Valuation Exercise Booklet

Respondent Booklet

Respondent ID ____________________

Interviewer Initials ____________________

Date ____________________

YOUR HEALTH

In this section, we are interested about your health.

Please tick the response which best describes your usual abilities OVER THE PAST WEEK:

Are you able to:	Without ANY difficulty ▼	With SOME difficulty ▼	With MUCH difficulty ▼	UNABLE to do ▼
1. Bend down to pick up clothing from the floor?	☐	☐	☐	☐
2. Climb up 5 steps?	☐	☐	☐	☐
3. Lift a full cup or glass to your mouth?	☐	☐	☐	☐
4. Stand up from a straight and armless chair?	☐	☐	☐	☐

Please answer the following question, which pertain to your health today:

5. **Pain/Discomfort**

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

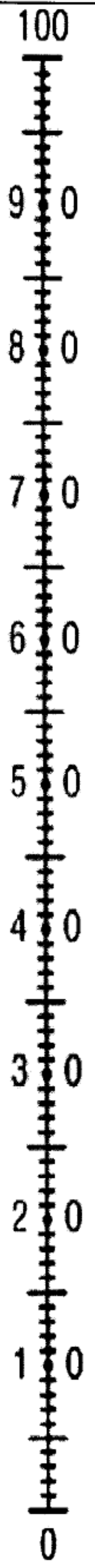
RATING EXERCISE

To help people say how good their health is, we have drawn a scale 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 the following health states are: full health, your health today, State X, State Y, State Z, and dead.

Please do this by drawing lines from the boxes below to whatever point on the scale (lines can cross). It is possible that more than one health state share the same point on the scale.

Best imaginable health state



Worst imaginable health state

Full health

Dead

Your health today

State X

State Y

State Z

VALUING HEALTH STATES
PRACTICE QUESTION: A

You are going to be asked to make a choice between two choices: Choice A and Choice B.

For Choice A, we would like you to imagine that you will live for 25 years in the health state described in the left-hand box and then you will die.

For Choice B, we would like you to imagine that you will live in the health state described in the right-hand box and then you will die. The years in Choice B will vary.

We would like you to indicate how many years in Choice B would be the same as 25 years in Choice A.

Please use the table at the bottom of the page overleaf.

CHOICE A

Health State A

You have **some difficulty** bending down to pick up clothes from the floor.

You have **much difficulty** climbing up 5 steps.

You have **some difficulty** lifting a full cup or glass to your mouth.

You have **some difficulty** standing up from a straight and armless chair.

You have **moderate** pain or discomfort.

CHOICE B

Full Health

Please put an “A” against all cases where you are CONFIDENT that you would choose Choice A.

Please put a “B” against all cases where you are CONFIDENT that you would choose Choice B.

Please put an “=” against all cases where you cannot choose between Choice A and Choice B.

Choice A: Health State A	Your Choice	Choice B: Full Health
25 years		0 years
25 years		1 years
25 years		2 years
25 years		3 years
25 years		4 years
25 years		5 years
25 years		6 years
25 years		7 years
25 years		8 years
25 years		9 years
25 years		10 years
25 years		11 years
25 years		12 years
25 years		13 years
25 years		14 years
25 years		15 years
25 years		16 years
25 years		17 years
25 years		18 years
25 years		19 years
25 years		20 years
25 years		21 years
25 years		22 years
25 years		23 years
25 years		24 years
25 years		25 years

If you answered B for the first row, please turn to page 5

If you did not answer B for the first row, please turn to page 7

PRACTICE QUESTION: B

CHOICE A

Health State A

You have **some difficulty** bending down to pick up clothes from the floor.

You have **much difficulty** climbing up 5 steps.

You have **some difficulty** lifting a full cup or glass to your mouth.

You have **some difficulty** standing up from a straight and armless chair.

You have **moderate** pain or discomfort.

Followed by

Full Health

CHOICE B

Dead

Please put an “A” against all cases where you are CONFIDENT that you would choose Choice A.

Please put a “B” against all cases where you are CONFIDENT that you would choose Choice B.

Please put an “=” against all cases where you cannot choose between Choice A and Choice B.

Choice A		**Your Choice**	**Choice B**
Health State A	**Full Health**		
25 years	0 years		Dead
24 years	1 years		Dead
23 years	2 years		Dead
22 years	3 years		Dead
21 years	4 years		Dead
20 years	5 years		Dead
19 years	6 years		Dead
18 years	7 years		Dead
17 years	8 years		Dead
16 years	9 years		Dead
15 years	10 years		Dead
14 years	11 years		Dead
13 years	12 years		Dead
12 years	13 years		Dead
11 years	14 years		Dead
10 years	15 years		Dead
9 years	16 years		Dead
8 years	17 years		Dead
7 years	18 years		Dead
6 years	19 years		Dead
5 years	20 years		Dead
4 years	21 years		Dead
3 years	22 years		Dead
2 years	23 years		Dead
1 years	24 years		Dead
0 years	25 years		Dead

VALUING HEALTH STATES

QUESTION 1

You are going to be asked to make a choice between two choices: Choice A and Choice B.

For Choice A, we would like you to imagine that you will live for 25 years in the health state described in the left-hand box and then you will die.

For Choice B, we would like you to imagine that you will live in the health state described in the right-hand box and then you will die. The years in Choice B will vary.

We would like you to indicate how many years in Choice B would be the same as 25 years in Choice A.

Please use the table at the bottom of the page overleaf.

CHOICE A	CHOICE B
Health State X You have **some** difficulty bending down to pick up clothes from the floor. You have **some** difficulty climbing up 5 steps. You have **no** difficulty lifting a full cup or glass to your mouth. You have **some** difficulty standing up from a straight and armless chair. You have **mild** pain or discomfort.	**Full Health**

Please put an "A" against all cases where you are CONFIDENT that you would choose Choice A.

Please put a "B" against all cases where you are CONFIDENT that you would choose Choice B.

Please put an "=" against all cases where you cannot choose between Choice A and Choice B.

Choice A: Health State X	Your Choice	Choice B: Full Health
25 years		0 years
25 years		1 years
25 years		2 years
25 years		3 years
25 years		4 years
25 years		5 years
25 years		6 years
25 years		7 years
25 years		8 years
25 years		9 years
25 years		10 years
25 years		11 years
25 years		12 years
25 years		13 years
25 years		14 years
25 years		15 years
25 years		16 years
25 years		17 years
25 years		18 years
25 years		19 years
25 years		20 years
25 years		21 years
25 years		22 years
25 years		23 years
25 years		24 years
25 years		25 years

If you answered B for the first row, please turn to page 9

If you did not answer B for the first row, please turn to page 11

QUESTION 1: B

CHOICE A

Health State X

You have **some** difficulty bending down to pick up clothes from the floor.

You have **some** difficulty climbing up 5 steps.

You have **no** difficulty lifting a full cup or glass to your mouth.

You have **some** difficulty standing up from a straight and armless chair.

You have **mild** pain or discomfort.

Followed by

Full Health

CHOICE B

Dead

Please put an "A" against all cases where you are CONFIDENT that you would choose Choice A.

Please put a "B" against all cases where you are CONFIDENT that you would choose Choice B.

Please put an "=" against all cases where you cannot choose between Choice A and Choice B.

Choice A		**Your Choice**	**Choice B**
Health State X	**Full Health**		
25 years	0 years		Dead
24 years	1 years		Dead
23 years	2 years		Dead
22 years	3 years		Dead
21 years	4 years		Dead
20 years	5 years		Dead
19 years	6 years		Dead
18 years	7 years		Dead
17 years	8 years		Dead
16 years	9 years		Dead
15 years	10 years		Dead
14 years	11 years		Dead
13 years	12 years		Dead
12 years	13 years		Dead
11 years	14 years		Dead
10 years	15 years		Dead
9 years	16 years		Dead
8 years	17 years		Dead
7 years	18 years		Dead
6 years	19 years		Dead
5 years	20 years		Dead
4 years	21 years		Dead
3 years	22 years		Dead
2 years	23 years		Dead
1 years	24 years		Dead
0 years	25 years		Dead

QUESTION 2

You are going to be asked to make a choice between two choices: Choice A and Choice B.

For Choice A, we would like you to imagine that you will live for 25 years in the health state described in the left-hand box and then you will die.

For Choice B, we would like you to imagine that you will live in the health state described in the right-hand box and then you will die. The years in Choice B will vary.

We would like you to indicate how many years in Choice B would be the same as 25 years in Choice A.

Please use the table at the bottom of the page overleaf.

CHOICE A	CHOICE B
Health State Y You have **some difficulty** bending down to pick up clothes from the floor. You have **much** difficulty climbing up 5 steps. You have **some** difficulty lifting a full cup or glass to your mouth. You have **much difficulty** standing up from a straight and armless chair. You have **moderate** pain or discomfort.	**Full Health**

Please put an "A" against all cases where you are CONFIDENT that you would choose Choice A.

Please put a "B" against all cases where you are CONFIDENT that you would choose Choice B.

Please put an "=" against all cases where you cannot choose between Choice A and Choice B.

Choice A: Health State Y	Your Choice	Choice B: Full Health
25 years		0 years
25 years		1 years
25 years		2 years
25 years		3 years
25 years		4 years
25 years		5 years
25 years		6 years
25 years		7 years
25 years		8 years
25 years		9 years
25 years		10 years
25 years		11 years
25 years		12 years
25 years		13 years
25 years		14 years
25 years		15 years
25 years		16 years
25 years		17 years
25 years		18 years
25 years		19 years
25 years		20 years
25 years		21 years
25 years		22 years
25 years		23 years
25 years		24 years
25 years		25 years

If you answered B for the first row, please turn to page 13

If you did not answer B for the first row, please turn to page 15

QUESTION 2: B

CHOICE A

Health State Y

You have **some** difficulty bending down to pick up clothes from the floor.

You have **much** difficulty climbing up 5 steps.

You have **some** difficulty lifting a full cup or glass to your mouth.

You have **much** difficulty standing up from a straight and armless chair.

You have **moderate** pain or discomfort.

Followed by

Full Health

CHOICE B

Dead

Please put an "A" against all cases where you are CONFIDENT that you would choose Choice A.

Please put a "B" against all cases where you are CONFIDENT that you would choose Choice B.

Please put an "=" against all cases where you cannot choose between Choice A and Choice B.

Choice A		Your Choice	Choice B
Health State Y	Full Health		
25 years	0 years		Dead
24 years	1 years		Dead
23 years	2 years		Dead
22 years	3 years		Dead
21 years	4 years		Dead
20 years	5 years		Dead
19 years	6 years		Dead
18 years	7 years		Dead
17 years	8 years		Dead
16 years	9 years		Dead
15 years	10 years		Dead
14 years	11 years		Dead
13 years	12 years		Dead
12 years	13 years		Dead
11 years	14 years		Dead
10 years	15 years		Dead
9 years	16 years		Dead
8 years	17 years		Dead
7 years	18 years		Dead
6 years	19 years		Dead
5 years	20 years		Dead
4 years	21 years		Dead
3 years	22 years		Dead
2 years	23 years		Dead
1 years	24 years		Dead
0 years	25 years		Dead

QUESTION 3

You are going to be asked to make a choice between two choices: Choice A and Choice B.

For Choice A, we would like you to imagine that you will live for 25 years in the health state described in the left-hand box and then you will die.

For Choice B, we would like you to imagine that you will live in the health state described in the right-hand box and then you will die. The years in Choice B will vary.

We would like you to indicate how many years in Choice B would be the same as 25 years in Choice A.

Please use the table at the bottom of the page overleaf.

CHOICE A	CHOICE B
Health State Z You have **much** difficulty bending down to pick up clothes from the floor. You are **unable** to climb up 5 steps. You have **much** difficulty lifting a full cup or glass to your mouth. You have **much** difficulty standing up from a straight and armless chair. You have **extreme** pain or discomfort.	**Full Health**

Please put an “A” against all cases where you are CONFIDENT that you would choose Choice A.

Please put a “B” against all cases where you are CONFIDENT that you would choose Choice B.

Please put an “=” against all cases where you cannot choose between Choice A and Choice B.

Choice A: Health State Z	Your Choice	Choice B: Full Health
25 years		0 years
25 years		1 years
25 years		2 years
25 years		3 years
25 years		4 years
25 years		5 years
25 years		6 years
25 years		7 years
25 years		8 years
25 years		9 years
25 years		10 years
25 years		11 years
25 years		12 years
25 years		13 years
25 years		14 years
25 years		15 years
25 years		16 years
25 years		17 years
25 years		18 years
25 years		19 years
25 years		20 years
25 years		21 years
25 years		22 years
25 years		23 years
25 years		24 years
25 years		25 years

If you answered B for the first row, please turn to page 17

If you did not answer B for the first row, please turn to page 19

QUESTION 3: B

CHOICE A

Health State Z

You have **much** difficulty bending down to pick up clothes from the floor.

You are **unable** to climb up 5 steps.

You have **much** difficulty lifting a full cup or glass to your mouth.

You have **much** difficulty standing up from a straight and armless chair.

You have **extreme** pain or discomfort.

Followed by

Full Health

CHOICE B

Dead

Please put an "A" against all cases where you are CONFIDENT that you would choose Choice A.

Please put a "B" against all cases where you are CONFIDENT that you would choose Choice B.

Please put an "=" against all cases where you cannot choose between Choice A and Choice B.

Choice A		**Your Choice**	**Choice B**
Health State Z	**Full Health**		
25 years	0 years		Dead
24 years	1 years		Dead
23 years	2 years		Dead
22 years	3 years		Dead
21 years	4 years		Dead
20 years	5 years		Dead
19 years	6 years		Dead
18 years	7 years		Dead
17 years	8 years		Dead
16 years	9 years		Dead
15 years	10 years		Dead
14 years	11 years		Dead
13 years	12 years		Dead
12 years	13 years		Dead
11 years	14 years		Dead
10 years	15 years		Dead
9 years	16 years		Dead
8 years	17 years		Dead
7 years	18 years		Dead
6 years	19 years		Dead
5 years	20 years		Dead
4 years	21 years		Dead
3 years	22 years		Dead
2 years	23 years		Dead
1 years	24 years		Dead
0 years	25 years		Dead

PERSONAL INFORMATION

(please tick the appropriate box in each group)

Sex:

☐ Male	☐ Female

Age:

☐ Under 19	☐ 50-59
☐ 20-29	☐ 60-69
☐ 30-39	☐ Over 70
☐ 40-49	☐ I prefer not to answer

Marital Status:

☐ Single	☐ Divorced
☐ Married/Civil partnership	☐ I prefer not to answer
☐ Widowed	

Highest Education Level Attained:

☐ Primary School (left at age 11)	☐ Other ____________________
☐ Secondary School (left at age 16)	*please specify*
☐ 'A' level (left at age 18)	☐ I prefer not to answer
☐ University	

Current Employment Situation:

☐ Self-employed	☐ Full time student / at school
☐ Paid employment (full or part-time)	☐ Long term sick or disabled
☐ Unemployed	☐ Other ____________________
	please specify
☐ Retired	☐ I prefer not to answer
☐ Looking after family or home	

Do you have a chronic health condition?	☐ Yes	☐ No
Do you have arthritis?	☐ Yes	☐ No

If yes, what type of arthritis do you have?

____

Do you know someone with arthritis?	☐ Yes	☐ No

If yes, what type of arthritis does he/she have?

____

If yes, what is your relationship to him/her?

____

Please answer the following 5 questions, which pertain to your health today:

1. **Mobility**
   - ☐ I have no problems in walking about
   - ☐ I have some problems in walking about
   - ☐ I am confined to bed

2. **Self-care**
   - ☐ I have no problems with self-care
   - ☐ I have some problems washing or dressing myself
   - ☐ I am unable to dress or wash myself

3. **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

4. **Pain/Discomfort**
   - ☐ I have no pain or discomfort
   - ☐ I have moderate pain or discomfort
   - ☐ I have extreme pain or discomfort

5. **Anxiety/Depression**
   - ☐ I am not anxious or depressed
   - ☐ I am moderately anxious or depressed
   - ☐ I am extremely anxious or depressed

How did you find the entire study?

- ☐ Very easy
- ☐ Easy
- ☐ Neither easy nor difficult
- ☐ Difficult
- ☐ Very difficult

Appendix A.5: Topic Guide for the Qualitative Component

PLAN OF ACTION FOR THE INITIALLY UNINFORMED GROUP:

VALUATION I -> ADAPTATION EXERCISE -> VALUATION II -> QUALITATIVE INTERVIEW

1. Introduction

Thank you for taking part in this study. I hope you had a chance to look over the information sheet that my colleague passed out to you. Do you have any questions regarding this information? If you don't have any questions pertaining to this study, please look over this consent form. This form states that you have looked over the information sheet and that you are willing to participate in this study. Please initial the boxes and sign and date the bottom of the form.

[*Participant to look over the consent form*]

Once again, thank you for taking part in this study. The results you give to us today will help inform us about how members of the general public think about living in a chronic health condition.

The study today has four parts. The first part will consist of this exercise booklet [*pass booklet out, along with a pen*]. Let's look through this together. The first page gives a description of the four types of different health you'll be looking at; please have a more detail look at this afterwards. Please rate the states along this scale, which 100 represents full health and 0 represents death. The next exercise looks at the three health states a bit more closely. This exercise asks you to imagine living in the one of the health states in a certain amount of time and to compare it to varying lengths of time in your current health. Please follow the instructions at the top of the page to select which option you prefer better.

For the second part of the study, I will present information to you about patients living in these health states and I will then ask you a few questions as to what you think about these patients' experiences.

For the third part, you will complete another exercise booklet similar to the booklet which you did at the start of the study.

Before the final part of the study, we'll take a short break and you can help yourself to the refreshments that I have provided. After that, we'll conclude with a brief discussion. I anticipate the whole session will take over an hour.

2. Valuation I

This is the first questionnaire booklet. Please read over the instructions and fill in your responses. If you have any questions, please let me know.

3. Adaptation Exercise

For this part of the study, I'll be using the audio recorder to record what you are saying. Please don't feel intimated by the machine; it's really for me to remember all the interesting things that you'll be saying.

What are the common symptoms of arthritis?

Answers:

- An illness that can cause pain and swelling in the joints (places were two bones meet, such as your elbow or knee)
- Lead to joint weakness, instability and visible deformities that, depending on the location of joint involvement, can interfere with the most basic daily tasks such as walking, climbing stairs, using a computer keyboard, cutting your food or brushing your teeth
- Inflammation of the joints from arthritis is characterized by joint stiffness, swelling, redness, and warmth. Tenderness of the inflamed joint may be present

Do you know anyone that has arthritis? How does it affect them?

A. Being diagnosed with and living with a chronic disease, such as arthritis, can be difficult to come to terms with and adjust to.

Now we'll hear from a patient with arthritis. Her name is Lisa. She's 30 years old and has only been diagnosed with arthritis for 1 year. Let's listen to what Lisa had to say about the impacts of arthritis on her everyday life:

> I didn't let anyone know how bad it was. You put a front on. It wasn't until I got indoors that I'd do the little weeping and the wailing kind of thing [laughs]. So yeah, I don't, I don't think they really knew, like, as I say, my mum didn't know until we'd gone to [the] Zoo, how bad I was. And she was really, really shocked. 'Cos I just didn't tell, you know, I'd just got on with it. Struggled, I didn't, you know, I didn't cope with it, I struggled. But as far as everyone else was aware it wasn't as bad as, you know, obviously for [daughter's name] and my husband, they didn't really know how bad it was. So I did cope with, I could go to Hollywood, couldn't I? I could be in Hollywood. But no, I did, I did really, yeah, yeah, I did cover it.
>
> I think one instance we'd gone to, we'd gone out with my brother-in-law and all our families and I was, just sat down normally. I was sat in a club kind of thing, you know, sat down having a drink and it was just like, 'I've got to go to the toilet' and it took me about 5 minutes, to get up, to get up and get out of the chair. And you know people were going, "We didn't realise you were that bad". 'Cos I just couldn't get my body to do anything.

What are the key aspects from what you just heard?

Have you experienced something similar to what Lisa has gone through? [If not, do you know a family member or a friend that has gone through/is going through to what Lisa has gone through? Do you know a social figure that has gone through/is going through to what Lisa has gone through? Can you imagine acting in the way Lisa is?]

- Have you felt the pain Lisa has described?
- Have you ever put on a strong face for someone despite what you're truly feeling?

Those are all very interesting points. This is what I think is happening in Lisa's quotation: She is obviously hiding her disease from her family but that frustration is getting to her, as she has to cry in private.

- Any other thoughts about Lisa's quotation?

It is possible that people take a while to come to terms with having the disease. For example, another patient, a 28-year old woman, diagnosed at the age of 14, still didn't really accept that she was a disabled woman. Some people did not visit the doctor or played down their symptoms because they didn't want to admit that something was wrong.

- Why do you think people do that?

B. Research has shown that individuals being diagnosed with a chronic, life-time condition, such as arthritis need to make changes to their everyday life. For example, an individual who would run professionally may need to modify their activities, such that they could run for a moderate amount or take up swimming so it won't be too hard on their joints. Other individuals have reported using gadgets have helped them out in the kitchen and in the bathroom. These gadgets include jar openers, reachers to help get things from high shelves, bathtub bar, raised toilet seats.

Now the next patient we'll hear from is Ann. She's in her early 50's and has been diagnosed with arthritis for 4 years. Let's listen to what Ann had to say about making changes due to her condition:

> But, and then I think it was about two years ago now I started swimming and that has just been fantastic. Because that is something I can do and I do it five days a week, every morning. I started off it, doing, it was this time of year, October, I got into the pool and I could do 35 lengths and I thought by Christmas I want to swim a mile and at Christmas I did. I was doing my 64 lengths in the hour.
>
> And now there's a new pool opened, and the same group of people go, and we all sort of, I mean they're not all sufferers, some just go because they enjoy going but we all sort of support each other, if you like, and I haven't been for two days this week so I'm already in trouble.
>
> But I can swim now for about an, well I could swim for 2 hours if I wanted to but I don't because I have other things to do, but I, I have found that that has helped and my consultant, you know, just sees me, says, 'ah my swimmer'. You know, he's, he's really impressed that of the you know, the way I've sort of dealt with it. I didn't think, "Ah, my life has ended, I'm never going to be able to do anything". I just thought "Well OK, this is what it is and I'm not going to let it beat me, you know. So I don't, I try to do everything as I did before, but in

> moderation and that seems to have worked quite well so far. I do still have bad days and sometimes the medicine upsets me.

What are the key aspects of what you just heard?
Have you experienced something similar to what Ann has gone through? [If not, do you know a family member or a friend that has gone through/is going through to what Ann has gone through? Do you know a social figure that has gone through/is going through to what Ann has gone through? Can you imagine yourself acting in the way Ann is?]

- o Have you ever had to change the way you've done something like Ann has described?
- o Can you provide some examples?

This is what I think is happening in Ann's quotation: Ann started swimming after being diagnosed with arthritis and has excelled at that activity. She enjoys both the physical and social aspects of it. She talks a bit of doing things in moderation.

- o Do you have other thoughts about Ann?

C. People suggested that taking control and learning how much they could do without overdoing it was the answer. Others with arthritis said, "Don't give in to it". Many felt lucky because they had lived many years able to do activities they enjoyed without the disease; they felt better now their disease had been diagnosed and was being treated; that they had arthritis and not something they considered worse.

Now the next patient we'll hear from is Patricia. She's in her late 70's and has been diagnosed with arthritis for over 40 years. Let's listen to what Patricia had to say:

> As I said earlier on, there are three ways you can deal with arthritis and I've found this out personally when I first started this. You can be very angry and fight it. That only lasts for a certain time because the only one that's getting hurt is you. 'Cos the more of a temper and, and that you get in the more you create, "Ooh that hurts", sort of thing.
>
> The other thing is you can give in right from the beginning and you can say, "I can't do that". And let everybody else do it for you and give no thought to the fact that they've got their lives to live and they shouldn't be feeling that way that they've got to do it for you. And the third thing is to come terms with it and don't live against it, live with it. And when you get a bad pain just sit, whatever suits you. If you get a bad pain and painting the wall gives you relief, go and paint the wall. If you find, like me myself, the only way to get over it is to just sit quietly and rest and it will go.

1. What do are the key points of what you just heard?

2. Have you experienced something similar to what Patricia has gone through? [If not, do you know a family member or a friend that has gone through/is going through to what Patricia has gone through? Do you know a social figure that has gone through/is going through to what Patricia has gone through? Can you imagine acting in the way Patricia is?]

- Have you felt the feelings Patricia has described?
- What were the circumstances?

3. This is what I think is happening in Patricia's quotation: Patricia is describing the natural course of a disease. You may be very negative and upset about it and maybe think "Why me?" Then you might want to give up and get everyone to help you with everything things. The final stage is that you accept the disease, and as Patricia says, "don't live against it, live with it". She talks about things she does to help her get over the pain.

- Do you have any other thoughts about Patricia?

Did you find this information interesting and helpful? Would this information have been helpful before the previous questionnaire?

3. Valuation II & Refreshment

Now please complete the second questionnaire booklet. Once you're finished, please hand in your completed answers to me and help yourself to the refreshments on the table.

[*HMC to calculate TTO and VAS values during this time*]

4. Qualitative Interviews

Based on your answers from the questionnaire you did today, this is how you valued the health states. [*Present them a table of their scores*]. The first scores are what you did at the start of the session and the second scores are after listening to what patients had to say about living with arthritis.

Using a scale, where one represents full health and zero represents death, you valued health state X at **xx**, which implies that you are willing to trade **xx** years off of your life to not live in this state. For health state Y, your value was **xx**, which implies that you are willing to trade **xx** years off of your life, and for health state Z, your value was **xx**, which implies that you are willing to trade **xx** years off of your life.

[If the first and second set of values changed] The two sets of values changed quite a bit. Why do you think this is so?

- Do you think that the information that I provided about the patients' experiences with arthritis influenced the second set of values?

[If the first and second set of values did not change] The two sets of values didn't change. Why do you think this is so?

- Do you think that the information that I provided about the patients' experiences had any affect on the second set of values?

PLAN OF ACTION FOR THE INFORMED GROUP:

ADAPTATION EXERCISE -> VALUATION I -> VALUES PRESENTATION & VALUATION II -> QUALITATIVE INTERVIEW

1. Introduction

Thank you for taking part in this study. I hope you had a chance to look over the information sheet that my colleague passed out to you. Do you have any questions regarding this information? If you don't have any questions pertaining to this study, please look over this consent form. This form states that you have looked over the information sheet and that you are willing to participate in this study. Please initial the boxes and sign and date the bottom of the form.
[*Participant to look over the consent form*]

Once again, thank you for taking part in this study. The results you give to us today will help inform us about how members of the general public think about living in a chronic health condition.

The study today has four parts. For the first part, I will present information to you about patients living in a chronic health condition and I will then ask you a few questions as to what you think about these patients' experiences.

The second part will consist of this exercise booklet [*pass booklet out, along with a pen*]. Let's look through this together. The first page gives a description of the three health states you'll be looking at; please have a more detail look at this afterwards. Please rate the states along this scale, which 100 represents full health and 0 represents death. The next exercise looks at the three health states a bit more closely. This exercise asks you to imagine living in the one of the health states in a certain amount of time and to compare it to varying lengths of time in your current health. Please follow the instructions at the top of the page, to select which option you prefer better.

For the third part, I will present some more information about patients living with the chronic condition and you'll once again some another exercise booklet similar to the booklet which you did at the start of the study.

Before the final part of the study, we'll take a short break and you can help yourself to the refreshments that I have provided. After that, we'll conclude with a brief discussion. I anticipate the whole session will take over an hour.

2. Adaptation Exercise

For this part of the study, I'll be using the audio recorder to record what you are saying. Please don't feel intimated by the machine; it's really for me to remember all the interesting things that you'll be saying.

What are the common symptoms of arthritis?
Answers:

- An illness that can cause pain and swelling in the joints (places were two bones meet, such as your elbow or knee)
- Lead to joint weakness, instability and visible deformities that, depending on the location of joint involvement, can interfere with the most basic daily tasks such as walking, climbing stairs, using a computer keyboard, cutting your food or brushing your teeth

- Inflammation of the joints from arthritis is characterized by joint stiffness, swelling, redness, and warmth. Tenderness of the inflamed joint may be present

Do you know anyone that has arthritis? How does it affect them?

A. Being diagnosed with and living with a chronic disease, such as arthritis, can be difficult to come to terms with and adjust to.

Now we'll hear from a patient with arthritis. Her name is Lisa. She's 30 years old and has only been diagnosed with arthritis for 1 year. Let's listen to what Lisa had to say about the impacts of arthritis on her everyday life:

> I didn't let anyone know how bad it was. You put a front on. It wasn't until I got indoors that I'd do the little weeping and the wailing kind of thing [laughs]. So yeah, I don't, I don't think they really knew, like, as I say, my mum didn't know until we'd gone to [the] Zoo, how bad I was. And she was really, really shocked. 'Cos I just didn't tell, you know, I'd just got on with it. Struggled, I didn't, you know, I didn't cope with it, I struggled. But as far as everyone else was aware it wasn't as bad as, you know, obviously for [daughter's name] and my husband, they didn't really know how bad it was. So I did cope with, I could go to Hollywood, couldn't I? I could be in Hollywood. But no, I did, I did really, yeah, yeah, I did cover it.
>
> I think one instance we'd gone to, we'd gone out with my brother-in-law and all our families and I was, just sat down normally. I was sat in a club kind of thing, you know, sat down having a drink and it was just like, 'I've got to go to the toilet' and it took me about 5 minutes, to get up, to get up and get out of the chair. And you know people were going, "We didn't realise you were that bad". 'Cos I just couldn't get my body to do anything.

What are the key aspects from what you just heard?

Have you experienced something similar to what Lisa has gone through? [If not, do you know a family member or a friend that has gone through/is going through to what Lisa has gone through? Do you know a social figure that has gone through/is going through to what Lisa has gone through? Can you imagine acting in the way Lisa is?]

- Have you felt the pain Lisa has described?
- Have you ever put on a strong face for someone despite what you're truly feeling?

Those are all very interesting points. This is what I think is happening in Lisa's quotation: She is obviously hiding her disease from her family but that frustration is getting to her, as she has to cry in private.

- Any other thoughts about Lisa's quotation?

It is possible that people take a while to come to terms with having the disease. For example, another patient, a 28-year old woman, diagnosed at the age of 14, still didn't really accept that she was a disabled woman. Some people did not visit the doctor or played down their symptoms because they didn't want to admit that something was wrong.

- Why do you think people do that?

B. Research has shown that individuals being diagnosed with a chronic, life-time condition, such as arthritis need to make changes to their everyday life. For example, an individual who would run professionally may need to modify their activities, such that they could run for a moderate amount or take up swimming so it won't be too hard on their joints. Other individuals have reported using gadgets have helped them out in the kitchen and in the bathroom. These gadgets include jar openers, reachers to help get things from high shelves, bathtub bar, raised toilet seats.

Now the next patient we'll hear from is Ann. She's in her early 50's and has been diagnosed with arthritis for 4 years. Let's listen to what Ann had to say about making changes due to her condition:

> But, and then I think it was about two years ago now I started swimming and that has just been fantastic. Because that is something I can do and I do it five days a week, every morning. I started off it, doing, it was this time of year, October, I got into the pool and I could do 35 lengths and I thought by Christmas I want to swim a mile and at Christmas I did. I was doing my 64 lengths in the hour.
>
> And now there's a new pool opened, and the same group of people go, and we all sort of, I mean they're not all sufferers, some just go because they enjoy going but we all sort of support each other, if you like, and I haven't been for two days this week so I'm already in trouble.
>
> But I can swim now for about an, well I could swim for 2 hours if I wanted to but I don't because I have other things to do, but I, I have found that that has helped and my consultant, you know, just sees me, says, 'ah my swimmer'. You know, he's, he's really impressed that of the you know, the way I've sort of dealt with it. I didn't think, "Ah, my life has ended, I'm never going to be able to do anything". I just thought "Well OK, this is what it is and I'm not going to let it beat me, you know. So I don't, I try to do everything as I did before, but in moderation and that seems to have worked quite well so far. I do still have bad days and sometimes the medicine upsets me.

What are the key aspects of what you just heard?

Have you experienced something similar to what Ann has gone through? [If not, do you know a family member or a friend that has gone through/is going through to what Ann has gone through? Do you know a social figure that has gone through/is going through to what Ann has gone through? Can you imagine yourself acting in the way Ann is?]

- Have you ever had to change the way you've done something like Ann has described?
- Can you provide some examples?

This is what I think is happening in Ann's quotation: Ann started swimming after being diagnosed with arthritis and has excelled at that activity. She enjoys both the physical and social aspects of it. She talks a bit of doing things in moderation.

- Do you have other thoughts about Ann?

C. People suggested that taking control and learning how much they could do without overdoing it was the answer. Others with arthritis said, "Don't give in to it". Many felt lucky because they had lived many years able to do activities they enjoyed without the disease; they felt better now their disease had been diagnosed and was being treated; that they had arthritis and not something they considered worse.

Now the next patient we'll hear from is Patricia. She's in her late 70's and has been diagnosed with arthritis for over 40 years. Let's listen to what Patricia had to say:

> As I said earlier on, there are three ways you can deal with arthritis and I've found this out personally when I first started this. You can be very angry and fight it. That only lasts for a certain time because the only one that's getting hurt is you. 'Cos the more of a temper and, and that you get in the more you create, "Ooh that hurts", sort of thing.
>
> The other thing is you can give in right from the beginning and you can say, "I can't do that". And let everybody else do it for you and give no thought to the fact that they've got their lives to live and they shouldn't be feeling that way that they've got to do it for you. And the third thing is to come terms with it and don't live against it, live with it. And when you get a bad pain just sit, whatever suits you. If you get a bad pain and painting the wall gives you relief, go and paint the wall. If you find, like me myself, the only way to get over it is to just sit quietly and rest and it will go.

What do are the key points of what you just heard?

Have you experienced something similar to what Patricia has gone through? [If not, do you know a family member or a friend that has gone through/is going through to what Patricia has gone through? Do you know a social figure that has gone through/is going through to what Patricia has gone through? Can you imagine acting in the way Patricia is?]

- Have you felt the feelings Patricia has described?
- What were the circumstances?

This is what I think is happening in Patricia's quotation: Patricia is describing the natural course of a disease. You may be very negative and upset about it and maybe think "Why me?" Then you might want to give up and get everyone to help you with everything things. The final stage is that you accept the disease, and as Patricia says, "don't live against it, live with it". She talks about things she does to help her get over the pain.

- Do you have any other thoughts about Patricia?

3. Valuation I (10 min)

This is the first questionnaire booklet. Please read over the instructions and fill in your responses. If you have any questions, please let me know.

[*HMC will need to calculate TTO and VAS values once participant hands in booklet*]

4. Values Presentation, Valuation II, and Refreshment Break (20 min)

Based on your answers you provided, this is how you valued the health states. [*Present them a table of their scores*].

Using a scale, where one represents full health and zero represents death, you valued health state X at **xx**, which implies that you are willing to trade **xx** years off of your life to not live in this state. For health state Y, your value was **xx**, which implies that you are willing to trade **xx** years off of your life, and for health state Z, your value was **xx**, which implies that you are willing to trade **xx** years off of your life.

Now, a study has shown that patients have valued these states as **xx**, **yy**, and **zz** [*fill these values on the same table*].

Are you willing to change your values to match those of the patients'? [*HMC to record the responses of the participants*]

Now please complete the second questionnaire booklet. Once you're finished, please hand in your completed answers to me and help yourself to the refreshments on the table.

[*HMC to calculate TTO and VAS values during this time*]

5. **Qualitative Interviews**

For this part of the study, I'll be using the audio recorder to record what you are saying.

Based on your answers from the questionnaire you did today, this is how you valued the health states. [*Present them a table of their scores*]. The first scores are what you did at the start of the session and the second scores are after listening to what patients had to say about living with arthritis.

Using a scale, where one represents full health and zero represents death, you valued health state X at **xx**, which implies that you are willing to trade **xx** years off of your life to not live in this state. For health state Y, your value was **xx**, which implies that you are willing to trade **xx** years off of your life, and for health state Z, your value was **xx**, which implies that you are willing to trade **xx** years off of your life.

[If the first and second set of values changed] The two sets of values changed quite a bit. Why do you think this is so?

- Do you think that the information that I provided about the patients' experiences with arthritis influenced the second set of values?

[If the first and second set of values did not change] The two sets of values didn't change. Why do you think this is so?

- Do you think that the information that I provided about the patients' experiences had any affect on the second set of values?

Now, a study has shown that patients have valued these states as 0.87, 0.74, and 0.67 [*fill these values on the same table*].

- What do you think about these values?
- Why do you think these values differ/do not differ from your values?
- Are you willing to change your values to match that of the patients'?
    - Why or why not?

Appendix A.6: Topic Guide for the Quantitative Component

Initially Uninformed Group

Respondent ID ____________________

Interviewer Initials ____________________

Date ____________________

Start time ____________________

End time ____________________

Introduction

Thanks for agreeing to take part in this survey. As explained in the letter you received, this is a survey for the School of Health and Related Research at the University of Sheffield. And it is about different ways people value health and illness.

All your responses will be treated as confidential, and all analysis will be carried out anonymously.

We are interested in people's views, and there are no right or wrong answers. Please tell us what you think.

Interviewer helpline:

Helen – 0114 222 0722 or 07950 346 550

[A] Self-reported health

FILL IN "START TIME" ON FRONT PAGE.

To start off, I would like you to answer a few questions about your own health.

Here are some statements about different aspects of your health. For the first four questions, could you tick just one statement that best describes your own health over the past week? For the last question, could you tick the statement that best describes your health today?

HAND RESPONDENT SELF-COMPLETION BOOKLET OPEN AT PAGE **1**.

AFTER THESE PAGES HAVE BEEN COMPLETED TAKE THE SELF-COMPLETION BOOKLET FROM RESPONDENT. MAKE SURE RESPONDENT HAS ONLY TICKED ONE BOX IN EACH GROUP.

GO TO THE RATING EXERCISE [B].

[B] Rating of 3 hypothetical arthritis health states plus full health, own health, and dead

Now, here is a set of 6 cards. Each of them has a description of a health state written on it. You will see that some description mention the aspects of health you have just looked at, but indifferent combinations. Each card has a different health state description on it.

I would like to ask you to indicate how good or bad these health states are. In this booklet, there is a scale like a thermometer. The top end of the scale is for "best imaginable health", and the bottom end of the scale is for "worst imaginable health".

HAND THE RESPONDENT THE SELF-COMPLETION BOOKLET OPEN AT PAGE **3**.

Please indicate how good or bad your health is by drawing a line from the box to the scale.

CHECK THAT THE RESPONDENT DRAWS ONE LINE FROM THE BOX TO THE SCALE.

NOW GO TO [C] TTO EXERCISE

[C] Interviewer script for TTO exercise

C1. TURN TO PAGE 4.
Now we're going to look at a different type of question. You are going to ask to make a choice between 2 options: Choice A and Choice B. For Choice A [POINT TO CHOICE A], we would like you to imagine that you will live for 25 years in the health state described in the left-hand box and then you will die. For Choice B [POINT TO CHOICE B], we would like you to imagine that you will live in the health state described in the right-hand box and then you will die. The years in Choice B will change. We would like you to indicate how many years in Choice B would be the same as 25 years in Choice A. Remember, I want you to imagine that <u>you</u> are in these states.

This here is a practice question. Please have a read at the two health states we will be looking at. [GIVE TIME SO PARTICIPANT CAN READ OVER THE STATES]

Using the table at the bottom of the page overleaf, I would like you to put an "A" against all cases where you are confident that you would choose Choice A, and a "B" against all cases where you are confident that you would choose Choice B. Please put an "=" against all cases where you cannot choose between Choice A and Choice B.

Let's work through this question together.

Looking at the first row, the question is would you rather live 25 years in Choice A or 0 years in Choice B [POINT TO FIRST ROW]?

a) IF INDIVIDUAL INDICATES THAT THEY WOULD RATHER LIVE 25 YEARS IN CHOICE A, PLACE "A" IN THE MIDDLE COLUMN. GO TO THE BOTTOM OF THE TABLE.
(If individual would rather live 0 years in Choice A, go to 'b'.)

Let's now work from the bottom of the table. Would you rather live 25 years in Choice A or 25 years in Choice B [POINT TO THE BOTTOM ROW]? [WHEN RESPONDENT INDICATES HIS/HER PREFERENCE, PLACE "A", "B", OR "=" IN THE MIDDLE COLUMN].

Let's go up one row. Would you rather live 25 years in Choice A or 24 years in Choice B [POINT TO THE SECOND TO BOTTOM ROW]? [WHEN RESPONDENT INDICATES HIS/HER PREFERENCE, PLACE "A", "B", OR "=" IN THE MIDDLE COLUMN].

PROCEED IN THIS FASHION, UNTIL ALL THE ROWS HAVE BEEN FILLED IN. IF THE INDIVIDUAL IS STRUGGLING WITH A PARTICULAR ROW, SUGGEST TO THE INDIVIDUAL TO START FROM THE TOP AND WORK DOWNWARDS UNTIL ALL THE ROWS HAVE BEEN COMPLETED.

b) IF INDIVIDUAL INDICATES THAT THEY WOULD RATHER LIVE 0 YEARS IN CHOICE B, PLACE "B" IN THE MIDDLE COLUMN. FLIP THE PAGE OVER TO CONTINUE

Look over these two new choices. [GIVE TIME FOR INDIVIDUAL TO LOOK OVER]. You can see there is a slight change from the previous page. Choice A has you spending some time in Health State A and then some time in Full Health. Choice B is being in a dead state.

Let's start at the bottom of the table. This question is asking if you would prefer to live in State A for 0 years followed by 25 years in Full Health or would you prefer to die immediately [POINT TO THE LAST ROW]. [WHEN RESPONDENT INDICATES HIS/HER PREFERENCE, PLACE "A", "B", OR "=" IN THE MIDDLE COLUMN].

PROCEED IN THIS FASHION, UNTIL ALL THE ROWS HAVE BEEN FILLED IN.

Do you have any questions?

Now, I would like to look at three health states.

FLIP TO PAGE 6.

INTERVIEWER NOTES:
WHEN PARTICIPANT COMPLETES A QUESTION, MAKE SURE TWO PAGES ARE FLIPPED. A NEW QUESTION STARTS ON PAGE 8 AND 12.

WHEN PARTICIPANT COMPLETES HEALTH STATE Z, IT IS POSSIBLE THAT HE/SHE WILL NEED TO COMPLETE THE "STATES WORSE THAN DEAD QUESTION" (E.G. ANSWERS CHOICE B FOR THE FIRST ROW). FLIP ONE PAGE OVER FOR HIM/HER. YOU MAY NEED TO WALK THROUGH EACH ROW OF THE TABLE FOR HIM/HER IN A SIMILAR FASHION TO THE PRACTICE QUESTION.

A PARTICIPANT WILL NOT NEED TO PUT "=", AS LONG AS THERE IS A SEQUENCE OF A'S AND A SEQUENCE OF B'S.

IF A PARTICIPANT WANTS TO PUT MORE THAN ONE "=", THAT'S FINE AS WELL.

STATES BETTER THAN DEATH CALCULATION:
IF INDIVIDUAL FILLED IN THE STATES WORSE THAN DEAD QUESTIONS, THEIR HEALTH STATE VALUE IS LESS THAN ZERO AND THEY WANT TO GIVE UP MORE THAN 25 YEARS TO AVOID LIVING IN THIS STATE.

[D] Adaptation exercise

Now we're going to listen to some recordings from patients and have a bit of a discussion about them. But first...

D1. Are you familiar with a condition known as arthritis? [WAIT FOR ANSWER]
Do you know what the common symptoms of arthritis are? [WAIT FOR ANSWER]

[PROVIDE ANSWER] Arthritis is an illness that leads to inflammation in the joints; a joint is where two bones meet, such as your elbow or knee. This inflammation can lead to stiffness, swelling, redness, tenderness, and pain. Arthritis can lead to joint weakness and visible deformities.

** IF SOMEONE ASKS FOR A DISTINCTION BETWEEN RHEUMATOID ARTHRITIS OR OSTEOARTHRITIS (DON'T PROVIDE OTHERWISE):
Osteoarthritis is a condition of wear-and-tear associated with aging or injury. Joints have become worn down from excessive use. Rheumatoid arthritis is when your own immune system mistakenly attacks healthy tissue, causing swelling which damages your joints.

D2. Do you know anyone that has arthritis? [WAIT FOR ANSWER]

ASK THE FOLLOWING QUESTION IF PARTICIPANTS KNOWS SOMEONE WITH ARTHRITIS:
How does it affect him/her?

[PROVIDE ANSWER] Arthritis can affect most of your basic daily tasks such as walking, climbing stairs, using a computer keyboard, cutting your food or brushing your teeth.

D3. So being diagnosed with and living with a chronic disease, such as arthritis, can be difficult to come to terms with and adjust to. First, we'll hear from a patient with arthritis. Her name is Lisa; she's 30 years old and has only been diagnosed with arthritis for 1 year. Let's listen to what Lisa had to say about the impacts of arthritis on her everyday life. [PLAY RECORDING]

- Can you summarize the information we just heard?
- Have you experienced something similar to what Lisa has gone through, for example put on a brave face for someone else despite what you're truly feeling inside? Please briefly explain.

ASK THE FOLLOWING QUESTIONS IF THE PARTICIPANT HASN'T EXPERIENCED SOMETHING SIMILAR TO LISA:

- If not, do you know a family member or a friend that has gone through/is going through to what Lisa is going through? Please briefly explain.

- If not, can you imagine acting in the way Lisa is if you were diagnosed with arthritis? Please briefly explain.

IF RESPONDENT TALKS ABOUT VULNERABILITY/COVERING UP/DOING ANYTHING NEGATIVE TO HIDE SYMPTOMS FROM OTHERS, ASK WHY?

This is what I think is happening in Lisa's quotation: Lisa is newly diagnosed with arthritis and, for some reason, she feels the need to hide her health condition from her family but the frustration is getting to her, as there was a point in the audio clip that she says she has to cry in private.

- Do you have any other comments to add?

D4. Research has shown that individuals being diagnosed with a chronic, life-time condition, such as arthritis, need to make changes to their everyday life. For example, an individual who would run professionally may need to modify their activities, such that they could run for a moderate amount or take up swimming so it won't be too hard on their joints or do an entirely new activity, such as learning a new musical instrument or doing crafts. Other individuals have reported that using gadgets have helped them out in the kitchen and in the bathroom. These gadgets include jar openers to help out in the kitchens; bathtub bars and raised toilet seats in the bathrooms; and even shoes with velcros can help with putting shoes on.

Now the next patient we'll hear from is Ann. She's in her early 50's and has been diagnosed with arthritis for 4 years. Let's listen to what Ann had to say about making changes due to her condition. [PLAY RECORDING]

- Can you summarize the information we just heard?
- Have you experienced something similar to what Ann has gone through by making changes when faced with a challenge in your life? Please briefly explain.

ASK THE FOLLOWING QUESTIONS IF THE PARTICIPANT HASN'T EXPERIENCED SOMETHING SIMILAR TO ANN:
- If not, do you know a family member or a friend that has gone through/is going through to what Ann has gone through? Please briefly explain.

- If not, can you imagine yourself acting in the way Ann is if you were diagnosed with arthritis? Please briefly explain.

DID THE RESPONDENT MENTION THE FOLLOWING WORDS?
☐ Coping ☐ Adapting ☐ Accommodating ☐ 'Similar' words

This is what I think is happening in Ann's quotation: Ann started swimming after being diagnosed with arthritis but it's not clear if she took up this sport because of her doctor's suggestion or just because she wanted to. But she has totally excelled at that activity. She enjoys both the physical and social aspects of it. She talks a bit of doing things in moderation.

- Do you have any other comments to add?

D5. People suggested that taking control and learning how much they could do without overdoing it was the answer. Others with arthritis said, "Don't give in to it". Many felt lucky because they had lived many years able to do activities they enjoyed without the disease; they felt better now their disease had been diagnosed and was being treated; that they had arthritis and not something they considered worse.

Now the next patient we'll hear from is Patricia. She's in her late 70's and has been diagnosed with arthritis for over 40 years. Let's listen to what Patricia had to say. [PLAY RECORDING]

- Can you please summarize the information we just heard?
- Have you experienced something similar to what Patricia has gone through? Please briefly explain.

Ask the last two questions if the first question didn't elicit any response:

- If not, do you know a family member or a friend that has gone through/is going through to what Patricia has gone through? Please briefly explain.

- If not, can you imagine acting in the way Patricia if you were diagnosed with arthritis? Please briefly explain.

This is what I think is happening in Patricia's quotation: Patricia is describing the natural course of a disease. At first, when you're told that you have arthritis, you may start off being very negative and upset about it and maybe think "Why me?". You might want to give up and get everyone to help you with everything things. The final stage is that you accept the disease, and as Patricia says, "don't live against it, live with it". She also talks about things she does to help her get over the pain.

- Do you have any other comments to add?

D6. How did you find this information interesting and helpful?

☐ Yes ☐ No ☐ No comment

Did you feel like you learned something?

☐ Yes ☐ No ☐ No comment

Or was it mostly things you already knew?

☐ Yes ☐ No ☐ No comment

Do you think this information would have been helpful before the previous questionnaire?

☐ Yes ☐ No ☐ No comment

[E] Presentation of scores from TTO exercises

THE RESPONDENT'S ANSWERS SHOULD BE FILLED IN ON THIS TABLE AS YOU COMPLETE EACH HEALTH STATE AS YOU COMPLETE THE TTO. YOU WILL NEED TO REFER TO THE RESULTS TABLE TO FILL IN THE REMAINING SPACES.

This table shows your results from the exercises where you had to evaluate the length of time you were willing to live with arthritis.

HAVE THE HEALTH STATES READY SO THE RESPONDENT CAN REFER BACK TO THEM.

The scores you see are evaluated on a scale from zero to one, where zero represents your preference to live in a dead state and one represents your preference to live in full health. So the higher the score you see, the more you prefer to live in that health state. Alternatively, the lower the score you see, it indicates how much you don't want to live in that health state. For each state, you'll see a score, which is between zero and one, and below that you see the number of years that you are willing to give up from your life to not live in that health state. So, for example, State Y you gave it a score of __________, which meant that you were willing to give up ___________ out of 20 years of your life to not live in this year state.

IF THERE IS A NEGATIVE SCORE:
For State ____ you gave a negative score which meant that you felt that this state was so horrible that you would rather die immediately than live in that health state.

Are you surprised with how you valued the three health states?

☐ Yes ☐ No ☐ No comment

Or are your scores what you expected?

☐ Yes ☐ No ☐ No comment

[F] Rating of 3 hypothetical arthritis health states plus full health, own health, and death

Now, here is a set of 6 cards. Each of them has a description of a health state written on it. You will see that some description mention the aspects of health you have just looked at, but indifferent combinations. Each card has a <u>different</u> health state description on it.

Once again, I would like to ask you to indicate how good or bad these health states are. In this booklet, there is a scale like a thermometer. The top end of the scale is for “best imaginable health”, and the bottom end of the scale is for “worst imaginable health”.

HAND THE RESPONDENT THE SELF-COMPLETION BOOKLET OPEN AT PAGE **X**.

Please indicate how good or bad your health is by drawing a line from the box to the scale.

CHECK THAT THE RESPONDENT DRAWS ONE LINE FROM THE BOX TO THE SCALE.

NOW GO TO [G] TTO EXERCISE

[G] Interviewer script for TTO exercise

Once again, please value these three health states in the same format as earlier. If you have questions, I would be happy to help you out.

[H] Background characteristics

In order to help us understand your responses, please fill in the background questions on the last page of the booklet.

INDICATE **PAGE 6** OF SELF-COMPLETION BOOKLET.

[I] Feedback Questions

We would like to know how you found the exercise you have just completed.

INDICATE **PAGE 5** OF SELF-COMPLETION BOOKLET.

Please circle one of the numbers which best reflects your opinion of each statement.

FILL IN “END TIME” ON FRONT PAGE OF INTERVIEWER SCRIPT.

Thank you very much for helping us today.

If you have any comments to make about the whole interview, please feel free to use the last page of the booklet.

Informed Group

Respondent ID ____________________

Interviewer Initials ____________________

Date ____________________

Start time ____________________

End time ____________________

Introduction

Thanks for agreeing to take part in this survey. As explained in the letter you received, this is a survey for the School of Health and Related Research at the University of Sheffield. And it is about different ways people value health and illness.

All your responses will be treated as confidential, and all analysis will be carried out anonymously.

We are interested in people's views, and there are no right or wrong answers. Please tell us what you think.

Interviewer helpline:

Helen – 0114 222 0722 or 07950 346 550

[A] Adaptation exercise

First we're going to listen to some recordings from patients and have a bit of a discussion about them. But first...

A1. Are you familiar with a condition known as arthritis? [WAIT FOR ANSWER]
Do you know what the common symptoms of arthritis are? [WAIT FOR ANSWER]

[PROVIDE ANSWER] Arthritis is an illness that leads to inflammation in the joints; a joint is where two bones meet, such as your elbow or knee. This inflammation can lead to stiffness, swelling, redness, tenderness, and pain. Arthritis can lead to joint weakness and visible deformities.

** IF SOMEONE ASKS FOR A DISTINCTION BETWEEN RHEUMATOID ARTHRITIS OR OSTEOARTHRITIS (DON'T PROVIDE OTHERWISE):
Osteoarthritis is a condition of wear-and-tear associated with aging or injury. Joints have become worn down from excessive use. Rheumatoid arthritis is when your own immune system mistakenly attacks healthy tissue, causing swelling which damages your joints.

A2. Do you know anyone that has arthritis? [WAIT FOR ANSWER]

ASK THE FOLLOWING QUESTION IF PARTICIPANT KNOWS SOMEONE WITH ARTHRITIS:
How does it affect him/her?

[PROVIDE ANSWER] Arthritis can affect most of your basic daily tasks such as walking, climbing stairs, using a computer keyboard, cutting your food or brushing your teeth.

A3. So being diagnosed with and living with a chronic disease, such as arthritis, can be difficult to come to terms with and adjust to. First, we'll hear from a patient with arthritis. Her name is Lisa; she's 30 years old and has only been diagnosed with arthritis for 1 year. Let's listen to what Lisa had to say about the impacts of arthritis on her everyday life. [PLAY RECORDING]

- Can you summarize the information we just heard?

- Have you experienced something similar to what Lisa has gone through, for example put on a brave face for someone else despite what you're truly feeling inside? Please briefly explain.

ASK THE FOLLOWING QUESTIONS IF THE PARTICIPANT HASN'T EXPERIENCED SOMETHING SIMILAR TO LISA:

- If not, do you know a family member or a friend that has gone through/is going through to what Lisa is going through? Please briefly explain.

- If not, can you imagine acting in the way Lisa is if you were diagnosed with arthritis? Please briefly explain.

IF RESPONDENT TALKS ABOUT VULNERABILITY/COVERING UP/DOING ANYTHING NEGATIVE TO HIDE SYMPTOMS FROM OTHERS, ASK WHY?

This is what I think is happening in Lisa's quotation: Lisa is newly diagnosed with arthritis and, for some reason, she feels the need to hide her health condition from her family but the frustration is getting to her, as there was a point in the audio clip that she says she has to cry in private.

- Do you have any other comments to add?

A4. Research has shown that individuals being diagnosed with a chronic, life-time condition, such as arthritis, need to make changes to their everyday life. For example, an individual who would run professionally may need to modify their activities, such that they could run for a moderate amount or take up swimming so it won't be too hard on their joints or do an entirely new activity, such as learning a new musical instrument or doing crafts. Other individuals have reported that using gadgets have helped them out in the kitchen and in the bathroom. These gadgets include jar openers to help out in the kitchens; bathtub bars and raised toilet seats in the bathrooms; and even shoes with velcros can help with putting shoes on.

Now the next patient we'll hear from is Ann. She's in her early 50's and has been diagnosed with arthritis for 4 years. Let's listen to what Ann had to say about making changes due to her condition. [PLAY RECORDING]

- Can you summarize the information we just heard?
- Have you experienced something similar to what Ann has gone through by making changes when faced with a challenge in your life? Please briefly explain.

ASK THE FOLLOWING QUESTIONS IF THE PARTICIPANT HASN'T EXPERIENCED SOMETHING SIMILAR TO ANN:

- If not, do you know a family member or a friend that has gone through/is going through to what Ann has gone through? Please briefly explain.

- If not, can you imagine yourself acting in the way Ann is if you were diagnosed with arthritis? Please briefly explain.

DID THE RESPONDENT MENTION THE FOLLOWING WORDS?

☐ Coping ☐ Adapting ☐ Accommodating ☐ 'Similar' words

This is what I think is happening in Ann's quotation: Ann started swimming after being diagnosed with arthritis but it's not clear if she took up this sport because of her doctor's suggestion or just because she wanted to. But she has totally excelled at that activity. She enjoys both the physical and social aspects of it. She talks a bit of doing things in moderation.

- Do you have any other comments to add?

A5. People suggested that taking control and learning how much they could do without overdoing it was the answer. Others with arthritis said, “Don't give in to it”. Many felt lucky because they had lived many years able to do activities they enjoyed without the disease; they felt better now their disease had been diagnosed and was being treated; that they had arthritis and not something they considered worse.

Now the next patient we'll hear from is Patricia. She's in her late 70's and has been diagnosed with arthritis for over 40 years. Let's listen to what Patricia had to say. [PLAY RECORDING]

- Can you please summarize the information we just heard?
- Have you experienced something similar to what Patricia has gone through? Please briefly explain.

Ask the last two questions if the first question didn't elicit any response:

- If not, do you know a family member or a friend that has gone through/is going through to what Patricia has gone through? Please briefly explain.

- If not, can you imagine acting in the way Patricia if you were diagnosed with arthritis? Please briefly explain.

This is what I think is happening in Patricia's quotation: Patricia is describing the natural course of a disease. At first, when you're told that you have arthritis, you may start off being very negative and upset about it and maybe think “Why me?”. You might want to give up and get everyone to help you with everything things. The final stage is that you accept the disease, and as Patricia says, “don't live against it, live with it”. She also talks about things she does to help her get over the pain.

- Do you have any other comments to add?

A6. How did you find this information interesting and helpful?

☐ Yes ☐ No ☐ No comment

Did you feel like you learned something?

☐ Yes ☐ No ☐ No comment

Or was it mostly things you already knew?

☐ Yes ☐ No ☐ No comment

[B] Self-reported health

FILL IN "START TIME" ON FRONT PAGE.

To start off, I would like you to answer a few questions about your own health.

Here are some statements about different aspects of your health. For the first four questions, could you tick just one statement that best describes your own health over the past week? For the last question, could you tick the statement that best describes your health today?

HAND RESPONDENT SELF-COMPLETION BOOKLET OPEN AT PAGE **1**.

AFTER THESE PAGES HAVE BEEN COMPLETED TAKE THE SELF-COMPLETION BOOKLET FROM RESPONDENT. MAKE SURE RESPONDENT HAS ONLY TICKED ONE BOX IN EACH GROUP.

GO TO THE RATING EXERCISE [C].

[C] Rating of 3 hypothetical arthritis health states plus full health, own health, and dead

Now, here is a set of 6 cards. Each of them has a description of a health state written on it. You will see that some description mention the aspects of health you have just looked at, but indifferent combinations. Each card has a different health state description on it.

I would like to ask you to indicate how good or bad these health states are. In this booklet, there is a scale like a thermometer. The top end of the scale is for "best imaginable health", and the bottom end of the scale is for "worst imaginable health".

HAND THE RESPONDENT THE SELF-COMPLETION BOOKLET OPEN AT PAGE **3**.

Please indicate how good or bad your health is by drawing a line from the box to the scale.

CHECK THAT THE RESPONDENT DRAWS ONE LINE FROM THE BOX TO THE SCALE.

NOW GO TO [D] TTO EXERCISE

[D] Interviewer script for TTO exercise

D1. TURN TO PAGE 4.
Now we're going to look at a different type of question. You are going to ask to make a choice between 2 options: Choice A and Choice B. For Choice A, we would like you to imagine that you will live for 25 years in the health state described in the left-hand box and then you will die. For Choice B, we would like you to imagine that you will live in the health state described in the right-hand box and then you will die. The years in Choice B will change. We would like you to indicate how many years in Choice B would be the same as 25 years in Choice A. Remember, I want you to imagine that you are in these states.

This here is a practice question. Please have a read at the two health states we will be looking at. [GIVE TIME SO PARTICIPANT CAN READ OVER THE STATES]

Using the table at the bottom of the page overleaf, I would like you to put an "A" against all cases where you are confident that you would choose Choice A, and a "B" against all cases where you are confident that you would choose Choice B. Please put an "=" against all cases where you cannot choose between Choice A and Choice B.

Let's work through this question together.

Looking at the first row, the question is would you rather live 25 years in Choice A or 0 years in Choice B [POINT TO FIRST ROW]?

a) IF INDIVIDUAL INDICATES THAT THEY WOULD RATHER LIVE 25 YEARS IN CHOICE A, PLACE "A" IN THE MIDDLE COLUMN. GO TO THE BOTTOM OF THE TABLE.
(If individual would rather live 0 years in Choice A, go to 'b'.)

Let's now work from the bottom of the table. Would you rather live 25 years in Choice A or 25 years in Choice B [POINT TO THE BOTTOM ROW]? [WHEN RESPONDENT INDICATES HIS/HER PREFERENCE, PLACE "A", "B", OR "=" IN THE MIDDLE COLUMN].

Let's go up one row. Would you rather live 25 years in Choice A or 24 years in Choice B [POINT TO THE SECOND TO BOTTOM ROW]? [WHEN RESPONDENT INDICATES HIS/HER PREFERENCE, PLACE "A", "B", OR "=" IN THE MIDDLE COLUMN].

PROCEED IN THIS FASHION, UNTIL ALL THE ROWS HAVE BEEN FILLED IN. IF THE INDIVIDUAL IS STRUGGLING WITH A PARTICULAR ROW, SUGGEST TO THE INDIVIDUAL TO START FROM THE TOP AND WORK DOWNWARDS UNTIL ALL THE ROWS HAVE BEEN COMPLETED.

b) IF INDIVIDUAL INDICATES THAT THEY WOULD RATHER LIVE 0 YEARS IN CHOICE B, PLACE "B" IN THE MIDDLE COLUMN. FLIP THE PAGE OVER TO CONTINUE

Look over these two new choices. [GIVE TIME FOR INDIVIDUAL TO LOOK OVER]. You can see there is a slight change from the previous page. Choice A has you spending some time in Health State A and then some time in Full Health. Choice B is being in a dead state.

Let's start at the bottom of the table. This question is asking if you would prefer to live in State A for 0 years followed by 25 years in Full Health or would you prefer to die immediately [POINT TO THE LAST ROW]. [WHEN RESPONDENT INDICATES HIS/HER PREFERENCE, PLACE "A", "B", OR "=" IN THE MIDDLE COLUMN].

PROCEED IN THIS FASHION, UNTIL ALL THE ROWS HAVE BEEN FILLED IN.

Now, I would like to look at three health states.

FLIP TO PAGE 6.

INTERVIEWER NOTES:
WHEN PARTICIPANT COMPLETES A QUESTION, MAKE SURE TWO PAGES ARE FLIPPED. A NEW QUESTION STARTS ON PAGE 8 AND 12.

WHEN PARTICIPANT COMPLETES HEALTH STATE Z, IT IS POSSIBLE THAT HE/SHE WILL NEED TO COMPLETE THE "STATES WORSE THAN DEAD QUESTION" (E.G. ANSWERS CHOICE B FOR THE FIRST ROW). FLIP ONE PAGE OVER FOR HIM/HER. YOU MAY NEED TO WALK THROUGH EACH ROW OF THE TABLE FOR HIM/HER IN A SIMILAR FASHION TO THE PRACTICE QUESTION.

A PARTICIPANT WILL NOT NEED TO PUT "=", AS LONG AS THERE IS A SEQUENCE OF A'S AND A SEQUENCE OF B'S.

IF A PARTICIPANT WANTS TO PUT MORE THAN ONE "=", THAT'S FINE AS WELL.

STATES BETTER THAN DEATH CALCULATION:
IF INDIVIDUAL FILLED IN THE STATES WORSE THAN DEAD QUESTIONS, THEIR HEALTH STATE VALUE IS LESS THAN ZERO AND THEY WANT TO GIVE UP MORE THAN 25 YEARS TO AVOID LIVING IN THIS STATE.

[E] Presentation of scores from TTO exercises

THE RESPONDENT'S ANSWERS SHOULD BE FILLED IN ON THIS TABLE AS YOU COMPLETE EACH HEALTH STATE AS YOU COMPLETE THE TTO. YOU WILL NEED TO REFER TO THE RESULTS TABLE TO FILL IN THE REMAINING SPACES.

This table shows your results from the exercises where you had to evaluate the length of time you were willing to live with arthritis.

HAVE THE HEALTH STATES READY SO THE RESPONDENT CAN REFER BACK TO THEM.

The scores you see are evaluated on a scale from zero to one, where zero represents your preference to live in a dead state and one represents your preference to live in full health. So the higher the score you see, the more you prefer to live in that health state. Alternatively, the lower the score you see, it indicates how much you don't want to live in that health state. For each state, you'll see a score, which is between zero and one, and below that you see the number of years that you are willing to give up from your life to not live in that health state. So, for example, State Z you gave it a score of __________, which meant that you were willing to give up __________ out of 20 years of your life to not live in this year state.

IF THERE IS A NEGATIVE SCORE:
For State ____ you gave a negative score which meant that you felt that this state was so horrible that you would rather die immediately than live in that health state.

Are you surprised with how you valued the three health states?

☐ Yes ☐ No ☐ No comment

Or are your scores what you expected?

☐ Yes ☐ No ☐ No comment

Now the second part of this table shows how patients, living with arthritis, valued the same 3 RA states. Have a look at their scores and compare it your own.

Patients gave State X a score of 0.81, meaning that they were willing to give up 4.8 years of their lives to not live with State X. Patients gave State Y a score of 0.73, meaning that they were willing to give up 6.8 years of their lives to not live with State Y. And, patients gave State Z a score of 0.66, meaning that they were willing to give up 8.5 years of their lives to live with State Z.

Are you surprised with how the patients valued the three health states?

☐ Yes ☐ No ☐ No comment

What do you think of your scores now?

☐ Yes ☐ No ☐ No comment

[F] Rating of 3 hypothetical arthritis health states plus full health, own health, and death

Now, here is a set of 6 cards. Each of them has a description of a health state written on it. You will see that some description mention the aspects of health you have just looked at, but indifferent combinations. Each card has a different health state description on it.

Once again, I would like to ask you to indicate how good or bad these health states are. In this booklet, there is a scale like a thermometer. The top end of the scale is for “best imaginable health”, and the bottom end of the scale is for “worst imaginable health”.

HAND THE RESPONDENT THE SELF-COMPLETION BOOKLET OPEN AT PAGE **X**.

Please indicate how good or bad your health is by drawing a line from the box to the scale.

CHECK THAT THE RESPONDENT DRAWS ONE LINE FROM THE BOX TO THE SCALE.

NOW GO TO [G] TTO EXERCISE

[G] Interviewer script for TTO exercise

Once again, please value these three health states in the same format as earlier. If you have questions, I would be happy to help you out.

[H] Background characteristics

In order to help us understand your responses, please fill in the background questions on the last page of the booklet.

INDICATE **PAGE 6** OF SELF-COMPLETION BOOKLET.

[I] Feedback Questions

We would like to know how you found the exercise you have just completed.

INDICATE **PAGE 5** OF SELF-COMPLETION BOOKLET.

Please circle one of the numbers which best reflects your opinion of each statement.

FILL IN "END TIME" ON FRONT PAGE OF INTERVIEWER SCRIPT.

Thank you very much for helping us today.

If you have any comments to make about the whole interview, please feel free to use the last page of the booklet.

Appendix A.7: Table Used to Present Values From Time Trade-off Exercises to the Informed Group

	HEALTH STATES		
	X	Y	Z
Your score			
The number of years you are willing to give up to have full health			
Patients' score	0.81	0.73	0.66
The number of years they are willing to give up to have full health	4.8	6.8	8.5

Appendix A.8: Copies of Approved Ethics Certificate

The
University
Of
Sheffield.

Cheryl Oliver
Ethics Committee Administrator
Regent Court
30 Regent Street
Sheffield S1 4DA

Telephone: +44 (0) 114 2220871
Fax: +44 (0) 114 272 4095 (non confidential)
Email: c.a.oliver@sheffield.ac.uk

Our ref: /CAO

25 September 2007

Helen McTaggart-Cowan
HEDS
ScHARR

Dear Helen

The Influence of Disease Adaptation on Resource Allocation: A Case Study Valuing Rheumatoid Arthritis Health States

Thank you for submitting the above research project for approval by the ScHARR Research Ethics Committee. On behalf of the University ethics reviewers who reviewed your project, I am pleased to inform you that the project was approved.

If during the course of the project you need to deviate significantly from the documents you submitted for review, please inform me since written approval will be required.

Yours sincerely

[redacted]

Cheryl Oliver

Ethics Committee Administrator

The
University
Of
Sheffield.

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Email: c.a.oliver@sheffield.ac.uk

Our ref: /CAO

30 June 08

Helen McTaggart-Cowan
HEDS
ScHARR

Dear Helen

The Influence of Disease Adaptation on Resource Allocation: A Case Study Valuing Rheumatoid Arthritis Health States

Thank you for submitting the above revised research project for approval by the ScHARR Research Ethics Committee. On behalf of the ScHARR chair of ethics, I am pleased to inform you that the project was approved.

If during the course of the project you need to deviate significantly from the documents you submitted for review, please inform me since written approval will be required.

Yours sincerely

[redacted]

Cheryl Oliver
Ethics Committee Administrator

Appendix A.9: Information Sheet and Consent Form for the Qualitative Component

School of Health and Related Research
University of Sheffield
Regent Court
30 Regent Street
Sheffield S1 4DA

7 February 2008

Dear Sir or Madam:

Re: Invitation to Participate in a Research Study

Researchers from the School of Health and Related Research (ScHARR) at the University of Sheffield are inviting you to take part in this research study, which is being undertaken as part of a Ph.D. degree. Your participation is entirely voluntary but, if you chose to participate, the answers you provide for this study will enable policy makers to make informed decisions in regards to allocating healthcare resources within the United Kingdom. This study has been approved by the University of Sheffield.

Before you decide on whether or not to participate in this study, it is important for you to understand what the research involves. The following information sheet will tell you about the study, why the research is being done, and what you will do during the study. Please read the following pages carefully. If you wish to participate in the study, please complete the enclosed Consent Form and the Availability Form; return the forms in the provided freepost envelope. The Study Coordinator will only contact you if you return the consent form.

If you have any questions or desire further information with respect to this study, you may contact the Study Coordinator, Ms. Helen McTaggart-Cowan, at 0114 222 0863 or at <h.m.cowan@sheffield.ac.uk>.

Again, your participation is voluntary and you are under no obligation to take part in this study. If you do not wish to participate, you do not have to provide any reasons for your decision nor will you lose the benefit of medical care to which you are entitled or are presently receiving.

Thank you for your time.

Yours sincerely,

Helen McTaggart-Cowan
Ph.D. Student

INFORMATION SHEET

VALUING HEALTH STATES IN THE GENERAL POPULATION

What is the purpose of this study?
This study is designed to evaluate members of the general public's views for treating chronic health conditions. The results from this study will allow us to aid policy makers about how to improve healthcare resource allocation and to make informed decisions to improve healthcare to meet societal and patient needs.

What does the study involve?
If you agree to participate in this study, you will be asked to meet the Study Coordinator for a one-to-one session at the University of Sheffield. You will be handed questionnaires to complete, which will contain various exercises asking you to value health states. The Study Coordinator will be present during the exercises and she will explain all the tasks thoroughly before you are asked to complete them. Upon completion of the exercises, you will be asked to discuss your results.

To help us with the analysis of the study results, the meeting will be tape recorded and transcribed word-for-word. You will be given £10 towards your transportation costs. Refreshments will be provided during the session.

What if I do not wish to take part?
This will in no way affect your present or future medical treatment. However, if you do decide to take part in this study, you will be given a copy of this information sheet and signed consent form to keep when you arrive for your scheduled session.

What if I change my mind during the study?
You are free to withdraw from the study at any time. This will not affect your medical treatment in any way.

What will happen to the information from the study?
All information will be anonymized and entirely confidential. Data from the study will be kept in a secure locked filing cabinet and on a password-protected computer.

Who is funding the study?
This project is currently funded by Astra Zeneca.

What if I have further questions?
You should contact Helen McTaggart-Cowan at either 0114 222 0863 or h.m.cowan@sheffield.ac.uk

What if I have complaints?
You should contact the University of Sheffield Research Services at 0114 222 1469.

CONSENT FORM

Project title: Valuing health states in the general population

Name of researcher: Helen McTaggart-Cowan

☐ 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 given any reason, without my medical care or legal rights being affected.

☐ I understand that my responses will be anonymized before analysis. I give permission for members of the research team to have access to my anonymized responses.

☐ I understand that the Study Coordinator will contact me by either phone or email upon receipt of the signed consent form. My contact information is:

__________________________________ (day time phone number)

__________________________________ (email address)

☐ I agree to take part in the above study.

Name of Participant (please print)	Date	Signature
Name of Researcher	Date	Signature

A copy of the information sheet and consent form will be given to you when you meet with the researcher. The original copy will be kept in a secured cabinet for our research records.

Appendix A.10: Thematic Framework

1.0 ATTITUDES TOWARD COPING

1.1 Ability to hide vulnerability

- 1.1.1 Act in a perceived easy way
- 1.1.2 British trait
- 1.1.3 Denial
- 1.1.4 Don't want to burden/trouble
- 1.1.5 Ego
- 1.1.6 Hide pain and symptoms
- 1.1.7 Human nature to do so
- 1.1.8 Mask appearance
- 1.1.9 Need to be strong
- 1.1.10 Not able to cope
- 1.1.11 To protect others
- 1.1.12 Scared of the outcome
- 1.1.13 Self-preservation

1.2 Making lifestyle changes

- 1.2.1 Accept illness
  - 1.2.1.1 Acceptance takes a while
  - 1.2.1.2 Acceptance fluctuates
  - 1.2.1.3 Can't let illness ruin life
  - 1.2.1.4 Come to terms with illness
  - 1.2.1.5 Do not dwell on problem
  - 1.2.1.6 For your sanity
  - 1.2.1.7 Mental attitude change
  - 1.2.1.8 A necessity
- 1.2.2 Adapt to/Cope with illness
  - 1.2.2.1 Adaptation is quick
  - 1.2.2.2 Adaptation takes a while
  - 1.2.2.3 Alleviate pain
  - 1.2.2.4 Educate yourself about illness
  - 1.2.2.5 Make changes
  - 1.2.2.6 Manage illness

1.3 Available support

- 1.3.1 Family support
- 1.3.2 Need a wide circle of friends
- 1.3.3 Spirituality and faith

2.0 VIEWS ON LIFE WITH ARTHRITIS

2.1 Experience with arthritis

- 2.1.1 Has arthritis experience
  - 2.1.1.1 It is a mild condition
  - 2.1.1.2 There are worse illnesses to have
  - 2.1.1.3 Own experience with arthritis
  - 2.1.1.4 Experience with arthritis through family and friends
  - 2.1.1.5 Knows no one with arthritis
- 2.1.2 Has no arthritis experience

        - 2.1.2.1 It is frustrating
        - 2.1.2.2 It is distressing
        - 2.1.2.3 It is life changing
        - 2.1.2.4 It is a progressive condition
        - 2.1.2.5 It is a severe health condition
        - 2.1.2.6 It is a sign that your body is failing
        - 2.1.2.7 You have no control
        - 2.1.28 It is a lifetime condition

2.2 An older person's disease
- 2.2.1 Not a young person's disease
    - 2.2.1.1 Older people can cope better
    - 2.2.1.2 Age stops you from doing certain things

3.0 PREVIOUS LIFE EVENTS

3.1 Experience with illness
- 3.1.1 Illness experience
- 3.1.2 Illness experience through family and friends
- 3.1.3 No illness experience

2.3 Not as painful as first perceived
- 2.3.1 Pain can be managed
- 2.3.2 Constant pain
- 2.3.3 Pain is distressing
- 2.3.4 Pain flares
- 2.3.5 Pain is hard to accept
- 2.3.6 Horrible pain

4.0 PERSONALITIES

4.2 Attitudes toward a change in life
- 4.2.1 Attain perceived control
- 4.2.2 Can't imagine having to make changes
- 4.2.3 Can't imagine living with arthritis
- 4.2.4 Can't moderate
- 4.2.5 Do things by yourself
- 4.2.6 Fight any presented changes
- 4.2.7 Hard to cope with changes
- 4.2.8 Look for options
- 4.2.9 Make best of the situation
- 4.2.10 Why me?

4.3 Empathetic people
- 4.3.1 Empathetic
- 4.3.2 Non-empathetic

5.0 DESIRE TO LIFE A LONG LIFE

5.1 Achieve life milestones
- 5.1.1 Watch children grow up
- 5.1.2 Experiences with family

Appendix A.11: Reasons to Change Questionnaire

Please tick the appropriate box to each of the following statements:

Q.1 TAKING PART IN TODAY'S SESSION HAS HELPED ME TO...

	Strongly Agree ▼	Slightly Agree ▼	Neither agree nor disagree ▼	Slightly **DIS**agree ▼	Strongly **DIS**agree ▼
a. Understand more about the disease of arthritis.	☐	☐	☐	☐	☐
b. Understand more about what it is like to live with arthritis.	☐	☐	☐	☐	☐
c. Realise that I now know as much as patients do about what it is like to live with arthritis.	☐	☐	☐	☐	☐
d. Realise that you still could have a good quality of life when living with arthritis.	☐	☐	☐	☐	☐
e. Realise that there are worse diseases to have than arthritis.	☐	☐	☐	☐	☐
f. Realise that arthritis is not "just a part of getting old".	☐	☐	☐	☐	☐
g. Realise that living with pain is not always a horrible thing.	☐	☐	☐	☐	☐
h. Realise that I would rather live longer with arthritis so I can spend more time with my family and friends.	☐	☐	☐	☐	☐
i. Realise that having arthritis does not have to make me look vulnerable or weak.	☐	☐	☐	☐	☐
j. Realise that I can cover up the signs of arthritis to appear normal.	☐	☐	☐	☐	☐
k. Realise that people can cope with having arthritis by themselves.	☐	☐	☐	☐	☐
l. Realise that family and friends can help people cope with arthritis.	☐	☐	☐	☐	☐
m. Realise that I could cope with arthritis because patients cope with it.	☐	☐	☐	☐	☐

Q.2 MY OPINIONS ABOUT ARTHRITIS CHANGED AFTER...

	Strongly Agree ▼	Slightly Agree ▼	Neither agree ▼	Slightly **DIS**agree ▼	Strongly **DIS**agree nor disagree ▼
a. Hearing the recordings.	☐	☐	☐	☐	☐
b. Talking to the interviewer.	☐	☐	☐	☐	☐
c. Seeing the patient scores[a]	☐	☐	☐	☐	☐

Q.3 IN GENERAL, I FEEL THAT...

	Strongly Agree ▼	Slightly Agree ▼	Neither agree ▼	Slightly **DIS**agree ▼	Strongly **DIS**agree nor disagree ▼
a. If I had to, I think that I can imagine living with arthritis for the rest of my life.	☐	☐	☐	☐	☐
b. I can "put myself in other people's shoes" and see things from their point of view.	☐	☐	☐	☐	☐
c. It's unfair for me to value a patient's life because I don't really know what it's like to live with arthritis.	☐	☐	☐	☐	☐
d. I have a positive outlook on life.	☐	☐	☐	☐	☐
e. I am the type of person that can adapt to change.	☐	☐	☐	☐	☐
f. I personally know what it is like to have a health problem.	☐	☐	☐	☐	☐
g. I know what it is like to have a health problem through a family member or a close friend.	☐	☐	☐	☐	☐
h. I understood the first valuation exercise.	☐	☐	☐	☐	☐
i. I understood the second valuation exercise.	☐	☐	☐	☐	☐
j. I had a difficult time deciding how long I wanted to live with arthritis on the valuation exercises.	☐	☐	☐	☐	☐

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[a] This item was only for those in the Informed Group

Appendix A.12: Invitation Letter to Participate in the Research Study (Quantitative Component)

The
University
Of
Sheffield.

Autumn 2008

Dear Resident,

We are writing to ask for your help with an important piece of research being carried out on behalf of the School of Health and Related Research at the University of Sheffield (ScHARR). This study is being done to understand the different ways people value health and illness, and is done as a part of a PhD degree programme.

The information you provide is very important. It will inform future health policies. ScHARR have asked the Centre for Health and Social Care Research at Sheffield Hallam University to undertake the interviewing for this research.

Addresses in your area have been randomly selected. An interviewer may call at your address. If you are at home when we call, we will ask for your help and provide further information on the research. You will be under no obligation to take part in this research. Enclosed is an information sheet that will tell you about the study, why the research is being done, and what you will do during the study. Any information you provide will be treated in the strictest confidence.

If you have any queries about the interview, please contact Rachel Ibbotson on 0114 225 4657. If you would like any further information about the research, please contact Helen McTaggart-Cowan on 0114 222 0722.

We would like to thank you in advance for your co-operation and participation in this important project.

Yours sincerely,

[redacted]

Rachel Ibbotson
Research Fellow
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Appendix A.13: Information Sheet for the Quantitative Component

VALUING HEALTH STATES IN THE GENERAL POPULATION

What is the purpose of this study?

This study is designed to evaluate members of the general public's views for treating chronic health conditions. The results from this study will allow us to aid policy makers about how to improve healthcare resource allocation and to make informed decisions to improve healthcare to meet societal and patient needs.

What does the study involve?

If you agree to participate in this study, you will be asked to complete several questionnaires, which will contain various exercises asking you to value different health states. The interviewer will be present during the exercises and will explain all the tasks thoroughly before you are asked to complete them. Upon completion of the exercises, you will be asked to discuss your results. Depending on the answers you give, the study will take between 60 and 90 minutes.

You will be given £10 for your participation.

What if I do not wish to take part?

This will in no way affect your present or future medical treatment. However, if you do decide to take part in this study, you will be given a copy of this information sheet and signed consent form to keep.

What if I change my mind during the study?

You are free to withdraw from the study at any time. This will not affect your medical treatment in any way.

What will happen to the information from the study?

All information will be anonymised and entirely confidential. Data from the study will be kept in a secure locked filing cabinet and on a password-protected computer.

Who is funding the study?

This project is currently funded by the Arthritis Research Campaign and Astra Zeneca.

What if I have further questions?

You should contact Helen McTaggart-Cowan at either **0114 222 0722** or h.m.cowan@sheffield.ac.uk

What if I have complaints?

You should contact the University of Sheffield Research Services at 0114 222 1469.

Appendix A.14: Consent Form for the Quantitative Component

Project title: Valuing health states in the general population

Name of researcher: Helen McTaggart-Cowan

(*Please tick the boxes*)

☐ 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 given any reason, without my medical care or legal rights being affected.

☐ I understand that my responses will be anonymised before analysis. I give permission for members of the research team to have access to my anonymised responses.

☐ I agree to take part in the above study.

Name of Participant (print)	Date	Signature
Name of Researcher	Date	Signature

Appendix A.15: Interviewer Questions

Please read each of the following statements, and circle the number from 1 to 5 that best represents your views. (Circle ONE number)

1	The respondent seemed to **understand** the VAS 'health thermometer' well	Agree Disagree 1 2 3 4 5
2	The respondent seemed to **understand** the TTO well	Agree Disagree 1 2 3 4 5
3	The respondent appeared to **lose interest** when valuing the latter health states	Agree Disagree 1 2 3 4 5
4	The respondent gave a **lot of thought** to the decisions that he/she was asked to make	Agree Disagree 1 2 3 4 5
5	The respondent was **engaged** with the patients' message in the recordings.	Agree Disagree 1 2 3 4 5

Please use this space to note any additional comments you would like to make.