



Development and Pilot of a Patient Reported Outcome Measure for Proximal Thoracic Aortic Aneurysms

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by

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Abstract

Background: Disease-specific questionnaires are increasingly being used to evaluate treatment outcomes from the perspective of patients. There are currently no validated questionnaires that measure patient-reported outcomes after proximal thoracic aortic aneurysm surgery.

Objectives: To develop and pilot a newly formulated patient focussed questionnaire that measures the patient's health status and health-related quality of life before and after proximal thoracic aortic aneurysm surgery.

Methods: Based on a literature review, a thematic analysis of audio recorded patient interviews and expert clinical testimony, a pool of items was generated to form a new questionnaire instrument. Suitable patients who were scheduled for elective aortic surgery at Liverpool Heart and Chest Hospital were identified and invited to participate in the pilot study. Patients were asked to complete the questionnaire prior to surgery and then at 6 weeks and 3 months after their operation. The newly developed instrument underwent preliminary testing for its appropriateness, acceptability, feasibility, interpretability, precision, reliability and responsiveness.

Results: Several items from the CROQ (Coronary Revascularisation Outcomes Questionnaire) formed the basis of the instrument, with the addition of 10 items derived from a newly formulated conceptual model of proximal thoracic aortic disease. The items were arranged into four domains (symptoms, physical, psychosocial and cognitive). Initial testing showed that the newly developed instrument performed to acceptable standards. It showed good internal consistency (Cronbach's alpha results for all domains >0.85), and test-retest reliability (intraclass correlation coefficient for all domains >0.85). In paired sample tests, the values in each domain led to statistically significant differences from baseline at either 6 weeks or 3 months ($p < 0.05$), supporting the construct validity and responsiveness of the instrument.

Conclusions: The new instrument demonstrated satisfactory validity as well as good internal reliability and test retest reliability for each item across all four domains. The initial findings suggest that the measure is sensitive and responsive to the effects of surgical treatment for proximal thoracic aortic aneurysms.

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List of Abbreviations

The following abbreviations have been used in this thesis:

AAA	Abdominal Aortic Aneurysm
AAS	Acute Aortic Syndrome
ARSA	Aberrant Right Subclavian Artery
BAV	Bicuspid Aortic Valve
CCS	Canadian Cardiovascular Society
CDC	Centers for Disease Control and Prevention
CDF	Cumulative Distribution Frequency
CoA	Coarctation of the Aorta
COPD	Chronic Obstructive Pulmonary Disease
CPB	Cardiopulmonary Bypass
CROQ	Coronary Revascularisation Outcome Questionnaire
CT	Computerised Tomography
CTT	Classical Test Theory
CU-Q2oL	Chronic Urticaria Quality of Life Questionnaire
DLQI	Dermatology Life Quality Index
DoH	Department of Health
EDS	Ehlers-Danlos Syndrome
EQ-5D	EuroQoL 5 Dimension Questionnaire
EQ-5D-5L	EuroQoL 5 Dimension 5 Level Questionnaire
EQ-VAS	EuroQoL Visual Analogue Scale
EuroSCORE	European System for Cardiac Operative Risk Evaluation
EVAR	Endovascular Aneurysm Repair
FFT	Friends and Family Test
GP	General Practitioner
HLY	Healthy Life Years
HRQoL	Health-Related Quality of Life
HSI	Health Status Index

ICC	Intraclass Correlation Coefficient
ICD-10	International Statistical Classification of Diseases, 10th Revision
IMD	Index of Multiple Deprivation
IQR	Inter-Quartile Range
IRT	Item Response Theory
IT	Information Technology
KD	Kommerell Diverticulum
LHCH	Liverpool Heart and Chest Hospital
LOS	Length of Stay
LVEF	Left Ventricular Ejection Fraction
MCID	Minimal Clinically Important Difference
MIC	Minimal Important Change
MMP	Matrix Metalloproteinase
MRI	Magnetic Resonance Imaging
MYMOP	Measure Yourself Medical Outcomes Profile
NHS	National Health Service
NICE	National Institute for Health and Clinical Excellence
NICOR	National Institute for Cardiovascular Outcomes Research
NYHA	New York Heart Association
PAS	Patient Administration System
PCI	Percutaneous Coronary Intervention
PREM	Patient Reported Experience Measure
PROM	Patient Reported Outcome Measure
PSEQ	Pain Self-Efficacy Questionnaire
Q1	Pre-Operative Questionnaire
Q2	6 Weeks Post-Operative Questionnaire
Q3	3 Months Post-Operative Questionnaire
QALY	Quality-Adjusted Life Year
QoL	Quality of Life
RCT	Randomised Control Trial

ROC	Receiver Operating Characteristic
ROS	Reactive Oxygen Species
SAS	Statistical Analysis System
SCTS	Society for Cardiothoracic Surgery in Great Britain & Ireland
SEM	Standard Error of Measurement
SF	Medical Outcomes Study Short Form
SF-6D	Short Form 6 Dimension
SMaRT	Screening Management and Referrals Tracking
TA	Thematic Analysis
TAA	Thoracic Aortic Aneurysm
TOE	Transoesophageal Echocardiography
TTE	Transthoracic Echocardiography
UAS	Urticaria Activity Score
UCT	Urticaria Control Test
UK	United Kingdom
US FDA	United States Food and Drug Administration
VSMC	Vascular Smooth Muscle Cells
WHO	World Health Organisation

Chapter 1

Introduction

1.1 General introduction

The aim of this thesis is to contribute to the understanding, recording and reporting of health-related quality of life (HRQoL) and health status in patients undergoing an elective surgical repair of a proximal thoracic aortic aneurysm (TAA).

This is addressed, initially, with a literature review that focusses on aortic aneurysms, the measurement of health outcomes and the development and use of questionnaire instruments designed to evaluate patients' HRQoL, these questionnaires are known as Patient Reported Outcome Measures (PROMs).

Subsequently, qualitative research in the form of in-depth interviews was used to develop of a conceptual model of proximal TAA patient's perceptions of their diagnosis, treatment and recovery. The results of this analysis included a set of themes that were used to support the composition of a PROM instrument.

Finally, the proximal TAA PROM itself was piloted and the validity, reliability and sensitivity of the tool were assessed. Before providing details of the aims, objective and structure of this thesis, a brief introduction to thoracic aortic aneurysms, HRQoL and PROMs is given, and the scope of the thesis is clarified.

1.2 Background

1.2.1 Thoracic aortic aneurysms

Diseases of the thoracic aorta are increasing in prevalence worldwide (Benchimol, 2015; NHS National Statistics, 2017). In the United States, The Centers for Disease Control and Prevention reports that aortic aneurysms (both thoracic and abdominal)

are the 15th leading cause of death in individuals aged over 55 years and the 19th leading cause of death overall (CDC, 2007). Although the prevalence of TAAs is likely lower than the reported prevalence of abdominal aortic aneurysms (AAAs), TAAs represent an important component of vascular disease due to their particularly lethal nature (Elefteriades, 2007; Kuzmik, 2012)

In the United Kingdom (UK), between 1999 and 2010, hospital admissions for thoracic aortic dissection increased from 7.2 to 8.8 and for TAA from 4.4 to 9.0 per 100,000 inhabitants (Bridgewater et al, 2009). These diseases have a high mortality; in the UK, mortality rates for thoracic aortic dissection and aneurysm are 3.2 and 7.5 per 100,000 inhabitants, respectively (Bridgewater et al, 2009; Bottle et al, 2017).

Mean annual admission rates for proximal aortic surgery in England and Wales have been reported at over 1440 cases per annum between 2007 and 2013 (National Institute for Cardiovascular Outcomes Research (NICOR) dataset results, 2007-2013), with an associated mortality rate of 9.0% (95% confidence intervals: 8.4% to 9.6%) (NICOR dataset results, 2007-2013). Thus, as the incidence of TAA is increasing, mortality rates are high and methods of detection are improving, further research within this area is justified. This leads to a compelling argument for the development of appropriate outcome measures that allow evaluations of the impact of hospital treatment from the patient's perspective.

Figure 1.1 shows the position of the heart and its major anatomical components, including the origin of the aorta, in a person's chest. A healthy, adult heart is about the size of a clenched fist (Shier, 2015).

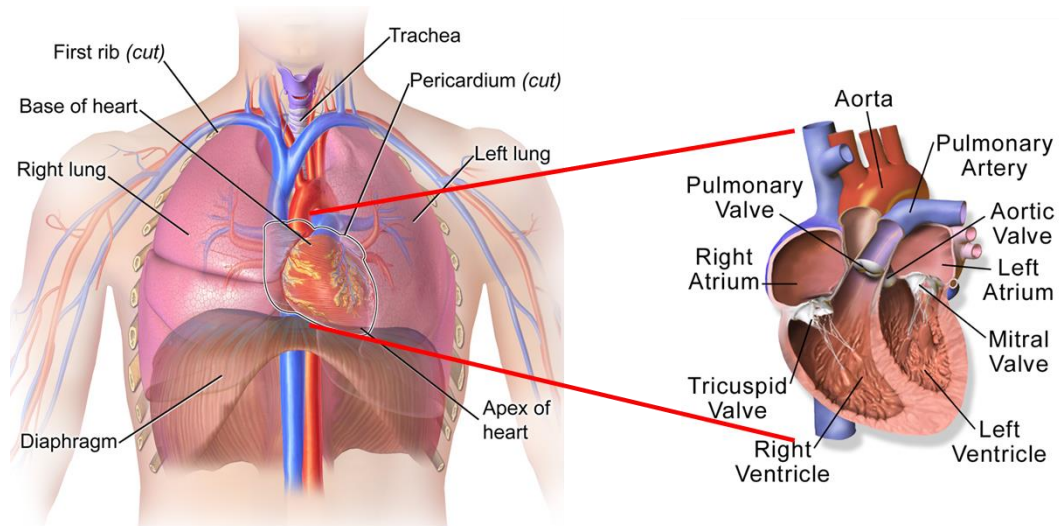


Figure 1.1: The human heart and its major anatomical components (adapted from Blausen, 2014a and 2014b)

Figure 1.2 then illustrates the difference between a normally sized aortic root and an aortic root that has been enlarged, or dilated, with aneurysmal disease. The affected section of the diseased aorta begins to bulge out “like a weak spot in a tire” (sic. as US spelling. Siwek, 1993).

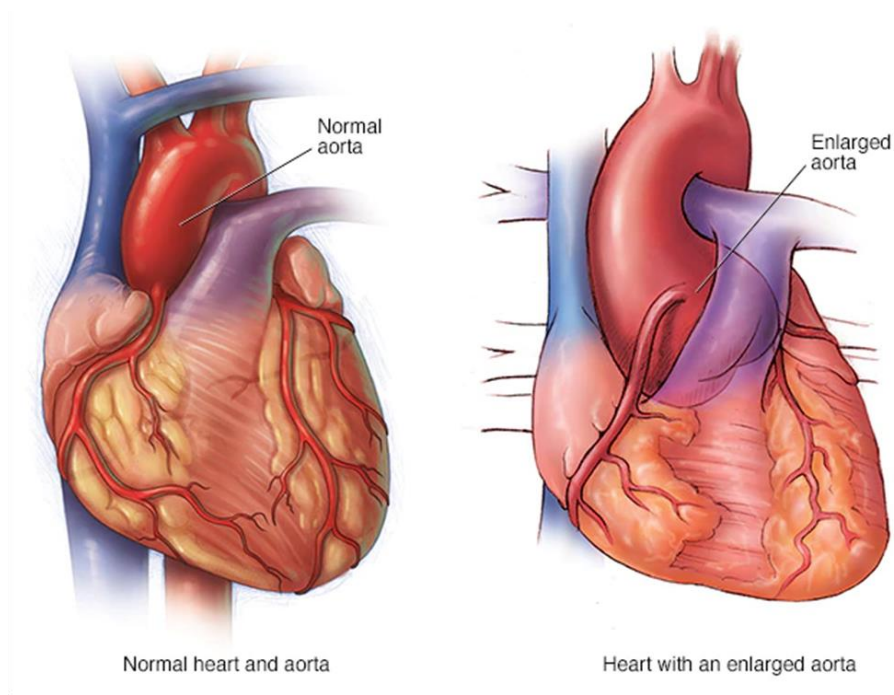


Figure 1.2: A normal heart and aorta, and a heart with an aortic root aneurysm (Mayo Clinic, 2019, used with permission of Mayo Foundation for Medical Education and Research, all rights reserved).

Figure 1.3 shows the basic anatomy of the thoracic aorta. The aorta is divided into two main sections; the proximal and distal. The proximal thoracic aorta consists of the aortic root, the ascending aorta and the aortic arch. The longer descending aorta segment is categorised as the distal thoracic aorta, and the abdominal aorta segment begins further down the vessel, below the abdomen. Proximal TAA surgery therefore includes procedures that address the proximal aorta up to and including the aortic arch via a sternotomy incision, without the need for a more invasive thoracotomy incision (these procedures upon the distal aorta are described in more detail by Safi (1998)).

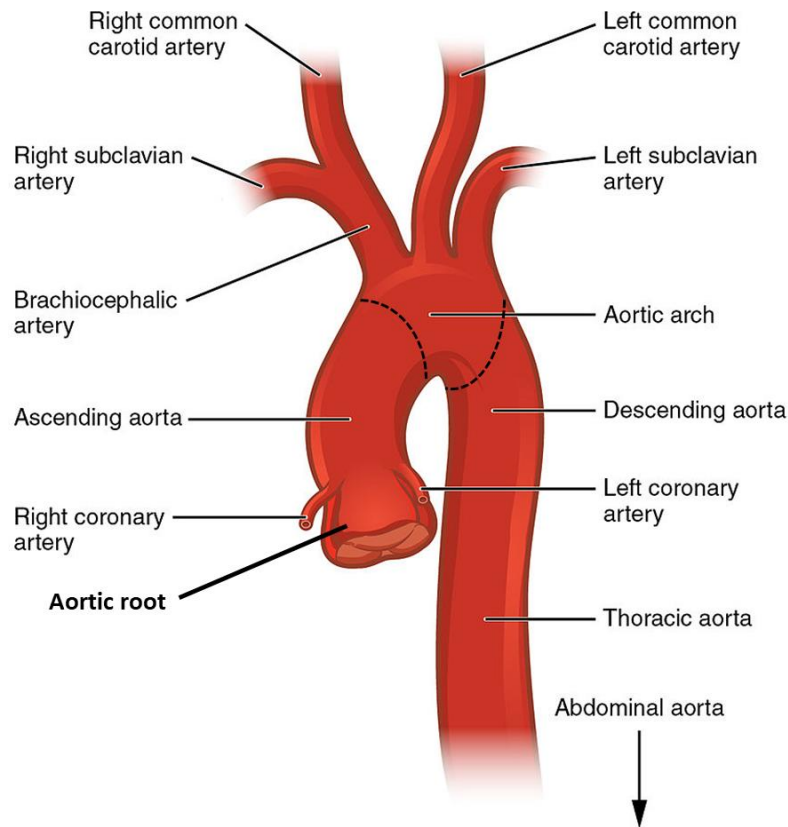


Figure 1.3: Basic anatomy of a normal thoracic aorta, adapted from OpenStax College (2013). (The heart is not shown in this diagram, but would appear below the aortic root).

As an aortic aneurysm increases in size, so the risk that the artery may rupture and cause a catastrophic injury also increases. The average diameter of a normal ascending aorta in a 75 year old woman is approximately 3.6–3.7cm and 4.1–4.2cm in a man of the same age (Hager, 2002). Current guidelines state that a typical patient with a proximal aortic aneurysm measured at or above 5.5cm should be considered a candidate for surgical intervention, although this generalised threshold can be lowered based on each individual patient’s risk profile (Saliba, 2015).

Unruptured TAA’s are found in the International Statistical Classification of Diseases and Related Health Problems 10th Revision (ICD-10), at ‘Chapter IX: Diseases of the circulatory system’, in the block ‘I70-I79: Diseases of arteries, arterioles and capillaries’. The code and description for this

condition is 'I71.2: Thoracic aortic aneurysm, without mention of rupture' (ICD-10, 2019).

1.2.2 Health-related quality of life

The World Health Organisation (WHO) define quality of life (QoL) as “an individual’s perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns” (The WHOQoL Group, 1995).

The ultimate goal of health care is to maintain or improve a patient’s QoL. Health is an important determinant of a person’s QoL although, as the WHO definition above makes clear, it is not the only one. QoL is comprised of a range of perceptions related to a patient’s well-being, based on their subjective experiences (Revicki, 2000). Many variables, both objective and subjective, interact to define QoL (Higginson and Carr, 2003), but it is dependent upon individual patient experiences, states, and perceptions of their illness. QoL can vary as a result of life events or changes in functional health status, with each area of QoL impacting the others (Revicki, 2000).

Factors such as culture, religion, environment, education, social status and income also affect QoL but they are often neglected in the context of health care interventions. Patient HRQoL is one of the main concerns of health care professionals and over recent decades has become an important health outcome indicator (Greenfield and Nelson, 1992; Wilson and Cleary, 1995; Lam, 1997). One possible reason for this is that advances in medical science and technology have resulted in an increasing number of people living with chronic diseases and disabilities. This change in our population’s morbidity pattern has called for a paradigm shift in how we should evaluate outcomes of illness and care. This raises questions around decision making such as: Is it worthwhile to keep a comatose person alive on a respirator? Is renal transplant a better treatment than haemodialysis for patients with renal failure? Is one particular health care delivery system better for patients with

chronic diseases than another? Traditional methods that focus on outcomes such as mortality rates, stroke rates and objective clinical parameters are no longer viewed as adequate to answer these questions (Tian-hui, 2005). There has been a concurrent change in public health discourse away from life expectancy and towards Healthy Life Years (HLY), with health outcomes becoming less focused on the quantity of years lived and more directed towards the quantity and quality of life (Hyder, 1998; Murray, 2012).

Although the term is used regularly in relation to patient care, there is no single, established definition of HRQoL. There is, however, general agreement that HRQoL focuses on the functional outcome associated with a medical condition and/or any subsequent treatment upon a patient (Cella, 1995; Schipper et al, 1996). This definition reflects a broad view of well-being encompassing the person's satisfaction, HRQoL is therefore subjective and multidimensional, comprising not only the basic property of disease 'symptoms' but also physical and occupational performance, psychological wellbeing, depth of social interaction and cognitive functioning (Schipper et al, 1996).

The measurement of HRQoL allows clinicians and researchers to further their understanding of the impact of diseases such as TAAs on the lives of patients (Guyatt, 1993). The results of these studies also allow patients to have a better grasp of their clinical situation. Patients who are familiar with relevant HRQoL results may have an increased ability to manage their own expectations of disease and the impact of possible treatments. In the case of patients with proximal TAA disease, comorbidities and complications such as concomitant cardiovascular or valvular disease, peripheral vascular disease, diabetes, renal insufficiencies or contributing congenital malformations could all play a part in influencing HRQoL. Equally, as TAA disease is often asymptomatic, the diagnosis of aneurysmal disease and the knowledge that this unwanted, potentially life-threatening abnormality is present may mean that the patient feels under increased psychological pressure, which could also in turn influence HRQoL.

Several different views exist on how to use PROMs to measure HRQoL, in large part due the subjective nature of HRQoL, but also due to the lack of distinction between independent and dependent variables, as well as mediating variables (Higginson and Carr, 2003). Other barriers identified that could limit the routine use of PROMs to gather HRQoL data are cost, feasibility and clinical relevance. For a measure to have clinical usefulness it must not only be valid, appropriate, reliable, responsive, and able to be interpreted, but it must also be simple, quick to complete, easy to score, and provide useful clinical data (Tian-hui, 2005). Introducing a PROM into general practice is not necessarily a straightforward undertaking, especially in healthcare systems which have limited resources. By way of an introduction to how a PROM programme may be carried out, Higginson and Carr (2001) provide a simple step-by-step method of firstly: Questions to ask when choosing a PROM (Table 1.1) and secondly: Stages to follow when integrating a PROM into regular clinical procedures (Table 1.2):

Table 1.1: Step 1. Questions to ask when choosing a PROM to measure HRQoL for use in clinical practice (from Higginson and Carr, 2001).

Order	Questions to ask when choosing a PROM
1	Are the domains covered relevant?
2	In what population and setting was it developed and tested, and are these similar to those situations in which it is planned to be used?
3	Is the measure valid, reliable, responsive, and appropriate?
4	What were the assumptions of the assessors when determining validity?
5	Are there floor and ceiling effect—that is, does the measure fail to identify deterioration in patients who already have a poor QoL or improvement in patients who already have a good QoL?
6	Will it measure differences between patients or over time and to what extent?
7	Who completes the measure: patients, their family, or a professional? What effect will this have—that is, will they complete it?
8	How long does the measure take to complete?
9	Do staff and patients find it easy to use?
10	Who will need to be trained and informed about the measure?

Table 1.2: Step 2. Introducing a PROM to measure HRQoL into clinical practice (from Higginson and Carr, 2001).

Order	Stages of PROM implementation
1	Review who is using which measures internally and externally
2	Choose a measure
3	Decide whether other outcomes also need to be monitored
4	Involve staff and patients
5	Adapt the measure for local use and requirements
6	Identify a leader of the project
7	Assign responsibilities (decide who will be doing what)
8	Agree on a timetable
9	Test when and where the measure will be completed
10	Prepare and test paperwork
11	Plan and begin training in both the use of the measure and associated clinical skills
12	Agree on start date and review period
13	Begin using the measure
14	Review its use in the first week and month and then at regular intervals
15	Review individual patients' results and group results to improve care
16	Modify measure as patients and staff feel appropriate for improving the use of the measure or make other changes

These checklists were particularly useful for the current study, as they provided a structured reference for timeline planning and involvement of key stakeholders during the PROM development and pilot.

1.2.3 Patient reported outcome measures

PROMs are a means of collecting information on the effectiveness of care delivered to patients, as perceived by the patients themselves (NHS Digital, 2017). Clinical interventions no longer aim only to treat specific medical problems in physical terms but to improve QoL as well, these instruments are designed to evaluate aspects of a patient's QoL. The intention is for PROM data to supplement and enrich any routinely collected clinical data for a particular disease and treatment, thus producing a more detailed and balanced information set regarding the quality of clinical interventions, and the success of the treatment from the patient's point of view.

Over recent years, clinicians and researchers have given greater recognition and academic attention to these subjective patient experiences. This increase in attention aims at a more holistic understanding of the patient and how hospital procedures which aim to treat specific diseases affect physical and psychosocial functioning in everyday life (Baiardini, 2010).

The term 'PROM' covers a group of outcomes used to measure a wide variety of aspects of care including: HRQoL, patient illness perceptions, treatment satisfaction and adherence. PROMs can be distinguished from other outcomes such as laboratory results and clinician or caregiver ratings because the data collected is from the patient's perspective, usually without interpretation by another individual (Patrick, 2007). Furthermore, PROMs are designed to focus on specific disease-related dimensions, such as the degree of chest pain caused by a TAA. Researchers are more frequently including PROMs in randomised control trials (RCTs) as they help demonstrate benefit, patient feelings regarding treatments and even survival (insofar as patients who respond can be assumed to still be alive) (Patrick, 2007). However, PROMs tend to be evaluated as a secondary measure, and are rarely the primary outcome of an RCT (Baiardini, 2010).

In the UK National Health Service (NHS), providers of four key elective interventions (unilateral hip and knee replacements, groin hernia and

varicose vein surgeries) were required to collect and report PROMs from 1st April 2009, under the terms of the Standard NHS Contract for Acute Services. In practice, this meant that all NHS hospitals were expected to invite patients undergoing one of these four relevant NHS-funded procedures to complete a specified pre-operative PROM questionnaire in accordance with the relevant guidance (Department of Health, 2008). Post-operative questionnaires were then sent to patients at a specified point in time after their operation.

The UK Government White Paper, 'Equity and excellence: Liberating the NHS', envisaged an increase in the scope and coverage of PROMs in future, starting from April 2011:

“Information generated by patients themselves will be critical to this process, and will include much wider use of effective tools like Patient Reported Outcome Measures (PROMs), patient experience data, and real-time feedback. At present, PROMs, other outcome measures, patient experience surveys and national clinical audit are not used widely enough. We will expand their validity, collection and use. The Department will extend national clinical audit to support clinicians across a much wider range of treatments and conditions, and it will extend PROMs across the NHS wherever practicable.” (paragraph 2.7)

In early 2016 the NHS performed a consultation on the national PROM programme, asking patients, clinicians, healthcare managers and academics about how they used PROM data. The results were subsequently published by the NHS England Insight & Feedback Team (2017). This report found a range of views towards PROMs, with many potential uses being identified for patients, clinicians and commissioners. The two main purposes of the NHS PROM programme were identified as:

- “1. Demonstrating the effectiveness of treatments; and
2. Providing information to reduce variation in care”

The consultation report found that where engagement from care providers, commissioners and patient representation groups was high, the utility gained from PROM results had value. However, the report also notes that in many cases the potential of PROMs was not being realised, with the financial and staffing costs of administering the programme being most keenly felt in these areas (the annual cost of collecting the four nationally mandated PROMs was estimated to be £825,000). The response of NHS England to the report findings was to discontinue the mandatory groin hernia and varicose vein PROM collections, as the results from these were found to be limited in scope. NHS England further resolved to explore digital collection of PROM data (whilst stressing the importance of appropriate models of patient consent), with the intention of increasing the timeliness of useful results and reducing the burden of data collection on front-line staff. They also indicated their intention to open up the supply of PROMs data to companies and other healthcare organisations in an effort to encourage innovation and new ideas.

In addition, the consultation report found that there was an appetite among respondents for PROM collection in other clinical areas, the most frequent being Cancer care and Long-Term Conditions. The need for robust, substantiated, dedicated tools for different disease types is also emphasised, as only PROMs with a high level of relevance will provide the necessary insights for patients, clinicians and other stakeholders (Insight & Feedback Team, 2017).

As focussed, patient-centred healthcare grows in importance, clinicians and researchers need a way to make health care decisions that meet the needs of patients. It is a key objective to ensure that treatment decisions meet patient and societal values, and to recognise that perceptions of treatment success may vary between the patient and clinicians (Revicki, 2000; Milne, 2012). Furthermore, investigations into public perceptions of PROM data suggest that areas judged important by the general public have not been included in some commonly used measurement tools (Higginson and Carr, 2003).

Collecting well designed and validated PROMs allows clinicians and researchers to take into account a wider array of information that cannot be obtained through laboratory or physical measures, and permit a subjective description of functioning alongside objective findings (McDowell, 2006). Through the collection of patient perceptions of interventions used to treat proximal TAAs, health care providers will be able to have a better understanding of which aspects of health patients value most highly and therefore what types of treatment may provide the greatest benefit from their perspectives.

The tools used for evaluating PROMs can vary significantly. Evaluation tools may be as simple as a single question asking the patient to state their QoL; however, they are more likely to take the form of a questionnaire with multiple items, which investigate several different domains that are related to HRQoL (Guyatt, 1993). The common thread that exists among measurement tools is that they attempt to summarise the judgments patients make about their health and illness experiences (Higginson and Carr, 2003). PROM tools can be placed into two broad categories: instruments that assess general health and instruments that are disease-specific.

Generic PROM tools investigate all important aspects of HRQoL and allow broad comparisons, but they do not necessarily investigate a specific aspect of a disease. Typically generic tools include questions relating to the four main domains of HRQoL: physical, functional, social and psychological health (de Boer et al, 2004). These tools may be less responsive to change as they provide an overall summary score of HRQoL, rather than a score on a specific area of health (Guyatt, 1993; Milne, 2012).

Specific HRQoL instruments are designed to target a disease, population, or an outcome. Where generic tools allow broad comparisons, specific tools may be more responsive to HRQoL changes in the specific patient population under investigation (Guyatt, 1993).

1.3 Scope of this thesis

This research focusses on people who have developed aneurysmal disease positioned somewhere in the proximal segments of their aorta, namely the aortic root, ascending aorta or the aortic arch. Patients who have aneurysmal disease in more distal areas (the descending or thoracoabdominal aorta) are not included within the scope of this work. Aneurysmal disease in these more distal aortic areas has a different natural history, aetiology and pathology to their proximal counterparts, along with considerably divergent treatment options. Proximal TAA patients are more likely to have concomitant coronary heart disease or heart valve insufficiencies. This could be either as a consequence of their aneurysmal disease, or a contributing factor towards it.

The cohorts of patients who took part in the qualitative interviews and the PROM pilot study were all elective attendees at a single tertiary NHS institution located in the North-West of England – Liverpool Heart and Chest Hospital (LHCH).

All emergency presentations were excluded, along with any patients who had diagnoses of chronic or acute aortic dissection, or indeed any other acute aortic syndrome (AAS) such as an aortic ulcer or an intramural haematoma (Vilacosta, 2001). The reason for the exclusion of acute presentations was the impossibility of delivering a pre-operative PROM, in order to have a baseline measure to compare post-operative PROMs against. Chronic dissection patients are sometimes well enough to be placed on the surgical waiting list and present electively, however they were considered to have a significantly dissimilar pathology to proximal TAA patients, hence their exclusion.

1.4 Overview of the thesis

1.4.1 Objectives

The main objectives for this thesis are to develop an item suite of key themes which describe the lived experience of proximal TAA patients, and from this develop a PROM tool to measure HRQoL that is suitable for use with future patients who receive an elective surgical procedure to treat proximal TAA disease.

This study aims to achieve this goal by considering a number of aspects related to proximal TAAs, HRQoL and PROMs:

1. To present a clear overview of the natural history of TAA's, the risks and consequences of aneurysmal disease (along with the likelihood of concomitant heart problems) and the current treatment options that are available. Also to review the history, philosophy and importance of monitoring HRQoL as an outcome of clinical interventions and to describe the adoption of PROMs as the main facilitating tool for collecting and analysing HRQoL information.
2. To explore the key themes of HRQoL within the proximal TAA patient population via detailed analysis of patient testimony.
3. To design and pilot a PROM tool based on the themes emerging from the lived experiences of proximal TAA patients, which can then be used within the prospective elective patient population.
4. To establish the feasibility of using a PROM tool in the proximal TAA patient population.
5. To analyse and present the results of the proximal TAA PROM pilot study, and to make recommendations for future development and study.

The outlined work creates an in-depth understanding of how patients perceive their own lived experiences of proximal TAA, and helps to generate important PROM information for this growing cohort of patients. This PROM information may then be useful to inform and advise both patients and clinicians when designing treatment plans and hospital services to improve outcomes in the future.

1.4.2 Chapter outline

The thesis contains six chapters. Following on from this introduction, Chapter 2 provides the literature review, this is followed by the methodology (Chapter 3), qualitative study of patients' experiences of living with proximal TAA (Chapter 4), PROM tool development and pilot study (Chapter 5) and an overall Discussion and Conclusion (Chapter 6). The content of each of these chapters is discussed in more detail in the following paragraphs.

Chapter 2 begins by describing and discussing the aorta and aneurysmal disease, including anatomy, diagnosis, causes of disease, symptoms, treatment options and current published outcomes, including HRQoL. The concept of HRQoL is then covered in detail before reviewing and presenting current proposed best practice with regards to developing a PROM tool designed for regular use. The chapter reviews reports from current PROM programmes to investigate how tools were built, how data was reported and any lessons learned. This systematic critical evaluation of the literature establishes current practice with regards to PROM design and reporting, which is applied to the proximal TAA PROM tool.

Chapter 3 critiques the relevant methodology and underlying assumptions related to the development of PROM tools. The underlying pragmatic philosophy of the study is also introduced and discussed, along with a rationale, description and definition of the mixed methods research approach that was used to execute the project.

Chapter 4 outlines the initial qualitative research study, covering the exploration of key HRQoL issues for patients who have a lived experience of proximal TAA disease. In-depth interview data is interrogated and presented in the form of a thematic analysis. A final conceptual model based on the data is presented and described. The methods and approach to focus group and the role of clinician involvement are discussed. Design considerations and a review of options for PROM construction are also included.

Chapter 5 explores the results from the pilot of the newly designed proximal TAA PROM. This includes detail on patient selection, the patient population (such as sociodemographic characteristics, co-morbidities and treatment history), how the tool was finally structured and delivered to the patients, along with evaluation of internal consistency and test-retest reliability. Appropriate scoring methods are given and the longitudinal comparison results of pre- and post-HRQoL domains are presented and discussed.

A systematic critical discussion of the research presented in this thesis, as well as relevant conclusions is provided in Chapter 6. This discussion also evaluates the strength and limitations of the research presented, and identifies pathways for continued future validation of the proximal TAA PROM, along with other further areas of research.

1.5 Contribution to knowledge

The purpose of this thesis is to contribute to two main areas of knowledge. Firstly, to identify and present the key domains of HRQoL for patients with proximal TAA disease. This is done via in-depth one-to-one interviews with patients who are either under hospital surveillance for proximal TAA disease (awaiting potential surgery with regular, scheduled scans to monitor any changes in the size of their aneurysm), or patients who have already undergone a previous surgery for the treatment of a proximal TAA. The construction of a conceptual model of patients' perceptions and experiences of proximal TAA disease, and the issues surrounding their care and treatment

contributes to existing literature as well as identifying areas where future research may be carried out.

Secondly, the design, refinement and piloting of a specific PROM tool for elective, surgical proximal TAA patients contributes to the practical medical literature. Previous research has been done on monitoring HRQoL in similar patient populations, but this was done using generic tools rather than specifically designed questionnaires based on patient testimony and expert clinical agreement. One further contribution that the design of this pilot study makes is the delivery of not one but two post-operative PROMs. The study protocol specified that follow-up PROMs should be sent at both six-weeks and at three months after the patient had their initial operation. The details of this approach are expanded upon in Chapter 4, but the data gathered from the double delivery of post-operative PROM questionnaires will allow more refined conclusions and recommendations to be made regarding the effects of treatment on proximal TAA patient HRQoL during the initial weeks and months of the post-operative phase.

Chapter 2

On thoracic aortic aneurysms, health-related quality of life and patient reported outcome measures – a review of the literature

2.1 Introduction

In addition to the traditional clinical indicators associated with patients, the practice of regularly measuring health outcomes from the perspective of the patient is now widespread (Dawson, 2010; Black, 2013). This chapter investigates in more detail the natural history, aetiology and pathological presentation of aortic aneurysmal disease and the concept of HRQoL. The literature related to patient HRQoL data via PROMs will be reviewed and current practice will be compared with and contrasted to prevailing methodologies and guidance.

In order to sufficiently understand the position of the patient suffering from an aortic aneurysm, Section 2.3 includes a brief history of medical treatment for the condition, this is followed by an examination of current understanding on the causes, symptoms and consequences of the disease. In Section 2.4, an overview of the literature on the topic of HRQoL is presented. Section 2.5 then evaluates the concept of PROMs, and provides the rationale for the creation of a disease specific tool for the collection, analysing and reporting of patient data.

2.2 Objectives for this chapter

This chapter aims to critically evaluate the current literature to:

- Create a comprehensive overview of aortic aneurysms, HRQoL and PROMs, thus updating and adding to previous reviews

- Provide a rationale for the development of a new PROM tool that will allow HRQoL to be collected, analysed and reported in patients with proximal aortic aneurysmal disease

2.3 Thoracic aortic aneurysms

2.3.1 History

In the Merriam Webster Dictionary (1999) the word aorta is said to derive from the ancient Greek word **αόρτη** (aorte), meaning “to lift” or “to raise”, demonstrating the anatomical nature of the aorta raising blood flow away from the heart. It is recorded as the term applied by Aristotle to the great artery of the heart, and was used earlier by Hippocrates to name the branches of the windpipe. Accurately pinpointing the origin of words is not easy, however, and there is some discussion about how the word may have first been coined (Albinali, 2007).

Evidence gathered from the close examination of Egyptian mummies dating from 3500 years ago, revealed that atherosclerosis (see Ross, 1986) and arterial calcification were relatively common during those times. Also one of the earliest known medical writings from the Egyptian Ebers Papyrus, thought to have been composed circa 1550 BC, contains a passage where the writer seems to identify arterial aneurysms, and recommends the following treatment: “Treat it with a knife and burn it with a fire so that it bleeds not too much.” (Barker, 1992).

Thompson (1986) provides a concise history for the medical treatment of aneurysms, which includes the work of the Greek writer Oribasius, who lived in the 4th century AD. According to Oribasius, a Greek surgeon named Antyllus in the 2nd century AD, left an early record of his advised therapy for aneurysms at that time, although these original writings have been destroyed. Oribasius noted Antyllus’s recommendations as follows, “We decline exceptionally big aneurysms, but we will operate as follows on aneurysms in the extremities, the limbs and the head.” Antyllus applied

ligatures to the arteries that entered and left the aneurysm and then cut into the aneurysm sac, removed the contents, and packed the cavity. Antyllus did not resect the aneurysm sac. He offered the following useful instruction, “Those who tie the artery, as I advise, at each extremity, but amputate the intervening dilated part, perform a dangerous operation. The violent tension of the arterial pneuma often displaces the ligatures.” (Crowe, 1957).

The next noteworthy advance in aneurysmal treatment came from Ambroise Paré (1510-1590), who advocated the application of proximal ligature to aneurysms but did not believe the sac should be opened because of the danger of fatal bleeding. Paré also described a ruptured aneurysm of the thoracic aorta and wrote, “The aneurysms which happen in the internal parts are incurable.” (Slaney in Greenhalgh, 1990; Barker, 1992). Andreas Vesalius (1514-1564) was a friend of Paré and was the first to describe thoracic and abdominal aortic aneurysms (Garrison, 1921).

John Hunter (1728-1793) studied the development of collateral circulation of occluded main arteries, which led to his method of treating aneurysms. He gained success by ligating the superficial femoral artery high in the thigh, in the area now known as Hunter’s canal. This case represented the first major innovation in the treatment of aneurysms since the Antyllus operation of the 2nd century (Perry, 1993).

Astley Cooper (1768-1841) made contributions in many fields of surgery, but his name is linked foremost to advances regarding the vascular system. In 1817, he provided the first recorded case of ligation of the aorta to treat a leaking iliac aneurysm (Brock, 1952).

Rudolph Matas (1860-1957) introduced endoaneurysmorrhaphy by obtaining proximal and distal control, incising and removing the aneurysmal clot, oversewing collaterals and preserving a lumen of blood flow. This technique successfully reduced the occurrences of gangrene and amputation that usually followed aneurysmal surgery and is a principle still used today (Matas, 1888; Elkin, 1940).

In between Matas' experience and the mid-20th century several methods of treating aortic aneurysms were employed, such as needling, wiring, proximal banding, ligation, cellophane wrapping and electrothermic coagulation. These techniques had persistent advocates, but ultimately fell out of favour (Osler, 1909; Nunn, 1995; Blakemore, 1952).

There was an advancement of diagnostic testing throughout the 20th century, including aortography via clinical angiography (see Wilms, 1995). Treatments also improved and in 1945 teams in Sweden and Boston (Crafoord, 1945; Gross & Hufnagel, 1945) reporting successful end-to-end anastomosis of the aorta to treat coarctation (or aortic narrowing, see Section 2.3.6.3 and Rao, 2005). This progressed in the 1950's with great steps forward in successful aortic aneurysm resection. Between 1951 and 1953, several landmark operations took place in quick succession from international teams (Schafer, 1951; Dubost, 1952; Julian, 1953; Brock 1953; DeBakey 1953a; Bahnson 1953).

The legitimacy of using a homograft replacement for aneurysmal treatment was a considerable step forward, but obtaining suitable arteries and veins for this type of procedure was a significant limitation. Consequently, the development and use of arterial prostheses began in the early 1950's with tubes of Vinyon-N being implanted into animal subjects (Voorhees Jr, 1952). The materials used in these prostheses quickly advanced through Nylon to implants constructed from Teflon and Dacron. Surgeons including DeBakey (1958) and Szilagyi (1966) worked closely with textile engineers to produce these elasticised, woven implants. Refined versions of these man-made devices are still in use today (Spadaccio, 2016)

Aneurysms of the thoracic aorta have long presented a challenge to surgeons. Aneurysms located in the proximal aortic segments can be saccular, fusiform or associated with an aortic coarctation (see Figure 2.3 for more detail on aneurysm morphology). Surgical techniques to tackle these life-threatening conditions have developed from Alexander (1944) simply resecting the aneurysm with the coarctation and sewing the ends without incorporating an anastomosis or graft. Swan (1950) wrote the first report of

surgical removal of an aneurysm associated with a coarctation and use of a homograft replacement. Then in 1951, Gross reported five cases of aneurysm associated with coarctation that he treated by means of resection and homograft. By the early 1950's, Bahnson (1953) and Cooley (1952) had resected saccular aneurysms and repaired the arterial walls using lateral stitching. DeBakey and Cooley (1953b) reported the first case performed with a successful resection and graft of a fusiform thoracic aneurysm. Since that time, all sections of the proximal and distal thoracic aorta from the root to the diaphragm have been operated on successfully and replaced using a variety of custom made prostheses.

2.3.2 The aorta

The aorta is the largest artery in the human body. It is the blood vessel attached to the heart that carries oxygenated blood to all parts of the body through systemic circulation. The aorta is divided anatomically into the thoracic and abdominal components.

The thoracic aorta, which is located above the diaphragm, is further divided into the proximal segments (aortic root, ascending aorta and aortic arch) and the distal descending aorta (see Figure 2.1).

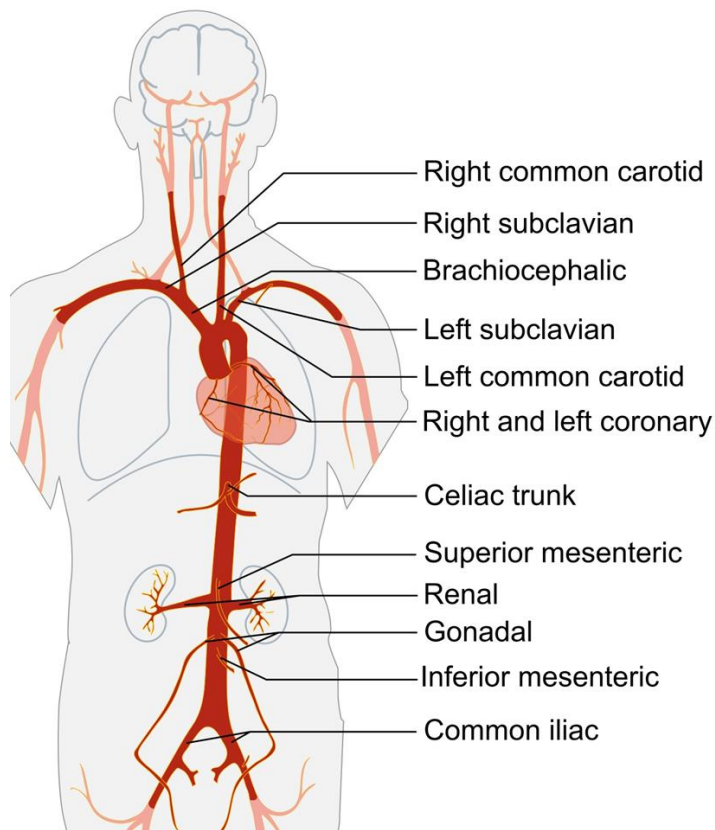


Figure 2.1: The aorta and its major branches (Häggröm, 2017)

In a normal aorta, the aortic root supports the bases of the aortic valve leaflets. The ascending aorta, located between the aortic valve and the innominate artery, is approximately 5 cm long. The three sinuses of Valsalva are the pockets of tissue which bulge outwards at the aortic root. These allow the aortic valve leaflets to manoeuvre during normal heart contractions. Typically, there are three aortic valve leaflets which span the aortic orifice, during a heart contraction the valve leaflets are pushed open to allow blood to flow through the aorta, they then close to seal and prevent any backflow. The left and right coronary arteries both emerge from the sinuses of Valsalva and supply blood to the heart itself. The innominate, or brachiocephalic, artery emerges from the proximal arch and splits into the right subclavian and right carotid arteries which supply blood to the right arm and the right side of the neck and head, respectively. Then the left carotid artery (which supplies blood to the left side of the neck and head) and the left subclavian (supplying the left arm), both subsequently emerge from the aortic arch.

The point where the aortic arch joins the descending aorta just distal to the left subclavian artery is called the aortic isthmus, the aortic arch ends at the level of the T4 vertebra or around the second rib. The thoracic aorta is relatively mobile and tears may occur at points of anatomical fixation, the aortic isthmus is where the aorta is attached to the chest wall by the ligamentum arteriosum and is particularly vulnerable to trauma. The descending aorta is situated distally from the left subclavian artery and descends to the diaphragm.

The walls of the aorta consist of three layers (see Figure 2.2): the tunica (layer, or covering) intima, the tunica media, and the tunica adventitia. The intima is the thin, delicate inner layer that is directly in contact with the blood. It is lined by slender, plate-like endothelium cells which can be easily damaged, the production of reactive oxygen species (ROS) by these endothelium cells may be a contributing factor of aneurysm formation (Miller, 2002).

The middle, or medial, layer is composed of intertwined sheets of elastic tissue such as elastin, collagen type I/III, proteoglycans and vascular smooth muscle cells (VSMCs) (Wolinsky, 1967). These elements are arranged in a spiral manner that provides tensile strength and elasticity; this allows the vessel to comfortably respond to changes in blood pressure. Aneurysmal formation will, over time, significantly reduce the integrity of these load-bearing cells via the degradation of the elastin and the deposition of collagen, resulting in a stiffer, less mobile aorta.

The adventitia is the thin outer layer containing the structural protein collagen, nerve fibres, fibroblasts, and vasa vasorum. It is also affected when aneurysms form.

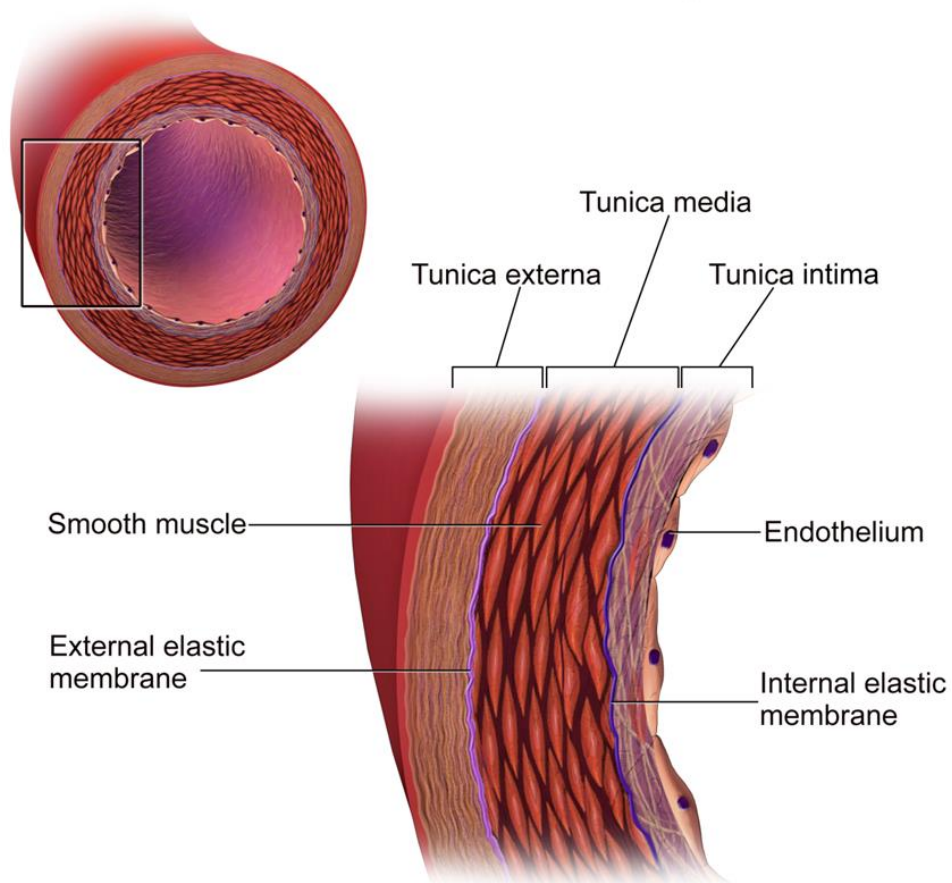


Figure 2.2: Cross section of an arterial wall, showing the three layers of composition (Blausen, 2014c)

2.3.3 Defining and describing aneurysms

The word ‘aneurysm’ is derived from the Greek **ἀνεύρυσμα** (aneurysma), which means “a widening/an opening” (Antoniou, 2011; Suy, 2006). Slezac in Magill’s Medical Guide (2018) defines an aneurysm as “A localised dilatation of a blood vessel, particularly an artery, that results from a focal weakness and distension of the arterial wall”. This arterial weakness can be brought about by atherosclerotic plaque formation which over time erodes the vessel wall, or by the loss of the structural fibres elastin and collagen within the vessel itself potentially caused by inflammatory diseases or genetic disorders. The enlargement of the blood vessel typically occurs gradually, and the weakness of the arterial wall becomes more pronounced over time

as the vessel shape, and consequently the flow of blood, alter (Klein, 2005). Congenital abnormalities, trauma and infections can all lead to the formation of aneurysms.

If an aneurysm expands beyond the point where the vessel wall remains intact, and the artery ruptures and splits open, this is known as 'aortic dissection' and has the potential to cause massive haemorrhage and death (Munson, 2005).

Aneurysms can take several different forms (see Figure 2.3).

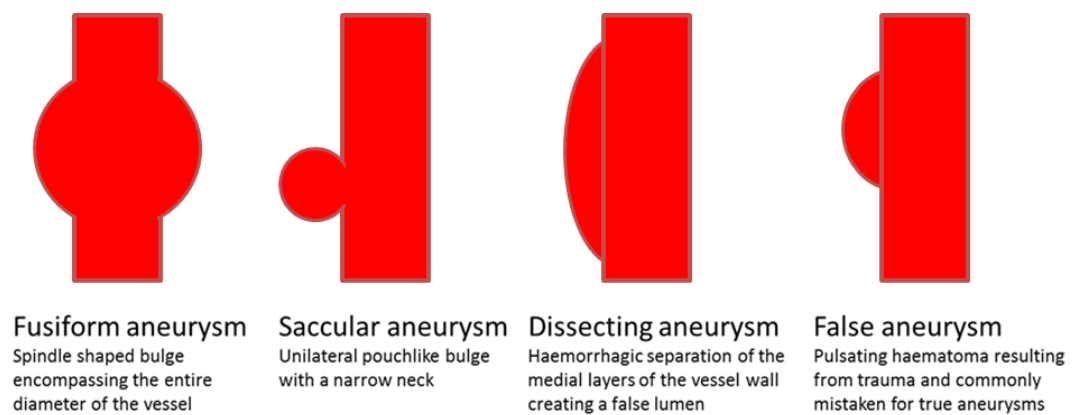


Figure 2.3: Types of aortic aneurysms (text from Munson, 2005)

2.3.3.1 Aneurysm morphology

A fusiform aneurysm develops when the arterial wall weakens around the whole of its circumference, this creates a symmetrical swelling along an extended section creating an aneurysm with the appearance of a spindle.

Saccular aneurysm formations occur when increased pressure in the artery pushes out a pouch on one side of the artery, creating a unilateral bulge. These types of aneurysm are less common than fusiform ones, and are perceived as carrying a greater risk of rupture than their fusiform counterparts (Shang, 2013; Szilagyi, 1966).

A dissecting aneurysm occurs when blood is forced between the layers of the arterial wall, causing them to separate and creating a false lumen.

A false aneurysm (also known as pseudoaneurysm) develops when there is a break in all layers of the arterial wall and blood leaks out, but is contained by surrounding structures. This creates a haematoma, or blood clot, which pushes the arterial wall outwards. These types of aneurysm usually occur after trauma.

Common aneurysmal locations include:

Thoracic aortic aneurysm – an abnormal progressive dilation of the normal aortic lumen involving all three layers of the vessel wall (the intima, media and adventitia). Occurring in one or more of the root, ascending, arch, or descending segments of the aorta.

Abdominal aortic aneurysm – an abnormal progressive dilation in the arterial wall, generally occurring in the aorta between the renal arteries and iliac branches. The aorta appears to be more susceptible to aneurysms than other blood vessels. The reason for this is thought to be because the aorta is the first artery to receive blood from the heart, and it is therefore placed under uniquely high levels of pressure. If there are any deficiencies in the wall of the aorta, it is more likely that aneurysms will occur (Fogoros, 2018)

Aneurysms can also occur in other areas of the body, for example a cerebral aneurysm is a localised dilation of a cerebral artery that may arise at an arterial junction in the circle of Willis, the circular anastomosis forming the major cerebral arteries at the base of the brain.

Femoral and popliteal aneurysms (sometimes called peripheral arterial aneurysms) are the end result of progressive atherosclerotic plaque growth changes occurring in the arterial walls of these major peripheral arteries.

2.3.4 Personal risk factors associated with aortic aneurysms

Personal risk factors for developing aortic aneurysms differ based on the age of the individual. In older people, atherosclerosis is likely to be a contributor to aneurysmal formation. Hypertension (high blood pressure), smoking and chronic obstructive pulmonary disease (COPD, a group of lung conditions that cause breathing difficulties) are all also significantly associated with an increased risk (Hiratzka, 2010; Fogoros, 2018). In younger patients, genetic or physiological variations are more common. However, there have been proposals dating back decades that the sole underlying cause of aortic aneurysms may be genetic anomalies. Kuivaniemi (1991) states that:

“reports from several groups [have] established that aortic aneurysms are familial, and, therefore, strongly suggested that they are caused by genetic defects...The family studies did not contradict the general impression that the development of aneurysms is accelerated by atherosclerosis, hypertension, and other factors. In fact, the higher incidence among brothers than sisters of patients strongly suggested a secondary component such as atherosclerosis contributes to the disease. The results, however, strongly suggested that a genetic defect unrelated to any genetic defect causing atherosclerosis or hypertension is the underlying cause of most aortic aneurysms.”

Studies continue to hypothesise on the fundamentally genetic nature of aortic aneurysms. Humphrey (2015) observes:

“An emerging concept is that altered cell–matrix connections...play important roles in TAADs. Given that such connections are fundamental determinants of cell phenotype and cell survival, this hypothesis is intuitive. Based on our review of the mechanics and mechanobiology...we submit further that many of the identified genetic mutations in [aortic

aneurysms]...directly affect the structural integrity of the aortic wall.”

Genetic syndromes with a predisposition for thoracic aortic aneurysms have been identified and are covered in more detail later in this chapter.

Thoracic aortic aneurysms are also associated with bicuspid aortic valve and other congenital cardiovascular anomalies and inflammatory diseases. Also, as Kuivaniemi alludes to, some thoracic aortic aneurysms are due to an inheritance of a predisposition for the disease, this has been termed ‘familial thoracic aortic aneurysm syndrome’. Still others have an unknown origin (Hiratzka, 2010).

2.3.4.1 Atherosclerosis

The role of atherosclerosis, or the build-up of plaque inside an artery (see Figure 2.4), highlights the differences in aetiology, pathology and natural history of aneurysmal development in different segments of the aorta.

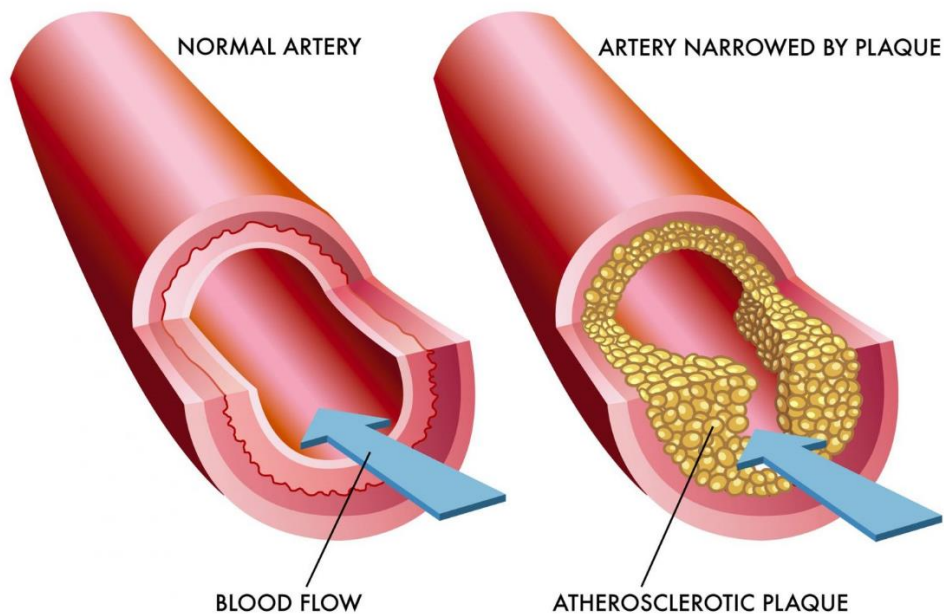


Figure 2.4: Normal artery and artery narrowed by atherosclerotic plaque (Dreamstime, 2019)

Atherosclerosis tends to be accepted as a key risk factor for the formation of aneurysms in the descending and abdominal aorta. However, the effect that atherosclerosis has on aneurysmal formation in the thoracic aorta is disputed. The European Society of Cardiology Task Force Report on 'Diagnosis and management of aortic dissection' (Erbal, 2001) makes the following generalised statement: "Atherosclerosis is the main cause of aortic aneurysms". This conclusion is based solely on the findings of two autopsy studies. There is however, some subsequent evidence that suggests patients who suffer with thoracic aneurysms actually develop less systemic atherosclerosis than age and sex-matched controls (Achneck, 2005). This finding has been supported by laboratory testing that has investigated enzymes linked with ascending aortic aneurysms and dissections, these enzymes are known as matrix metalloproteinase (MMP). It has been hypothesised that an increased level of MMPs could encourage the growth of aneurysms at the same time as disintegrating any formations of atherosclerotic plaques (Elefteriades, 2010; Silence, 2002).

Isselbacher (2005) also maintains that atherosclerosis is "an infrequent cause of ascending thoracic aortic aneurysms", and that plaque build-up as a contributing factor is more associated with descending and abdominal aortic aneurysms.

Currently, it seems that the relationship between atherosclerosis and aneurysmal development is not fully understood. Further detailed investigation is required to reveal the extent of contribution towards aneurysmal disease, based on both the position of the aortic aneurysm in the vessel itself and the presence of other important factors such as MMP enzymes.

2.3.4.2 Degenerative changes

The underlying cause of the tissue abnormality that results in thoracic aortic aneurysms is the degeneration of the central (medial) layer of the blood

vessel. This degeneration presents as a loss of the smooth muscle cells, a breaking up and reduction in the numbers of the elastic tissue fibres, and an increase in the number of proteoglycans (Guo, 2006). Proteoglycans are connective tissue proteins which are produced by smooth muscle cells. When the tissue is subjected to low levels of mechanical strain these proteins are created in greater numbers. It has been suggested that genetic mutations could lead to a decrease in smooth muscle cell contraction, leading to increased tissue strain and resulting in the cells increasing production of both proteoglycans and MMPs (Milewicz, 2008).

This fragmentation of the normal cellular construction of the aorta most often results in a cyst-like formation in the medial layer of the vessel wall (see Figure 2.5). This cystic medial degeneration, also known as cystic medial necrosis, weakens the wall of the aorta and encourages the formation of an aneurysm. In a person with cystic medial degeneration, high blood pressure greatly accelerates the condition and makes rapidly-developing aneurysms much more likely (O'Rourke, 2004). As noted above, proximal thoracic aortic aneurysms are most often the result of these non-atherosclerotic degenerative changes, in contrast to descending and abdominal aortic aneurysms which tend to be associated with atherosclerosis.

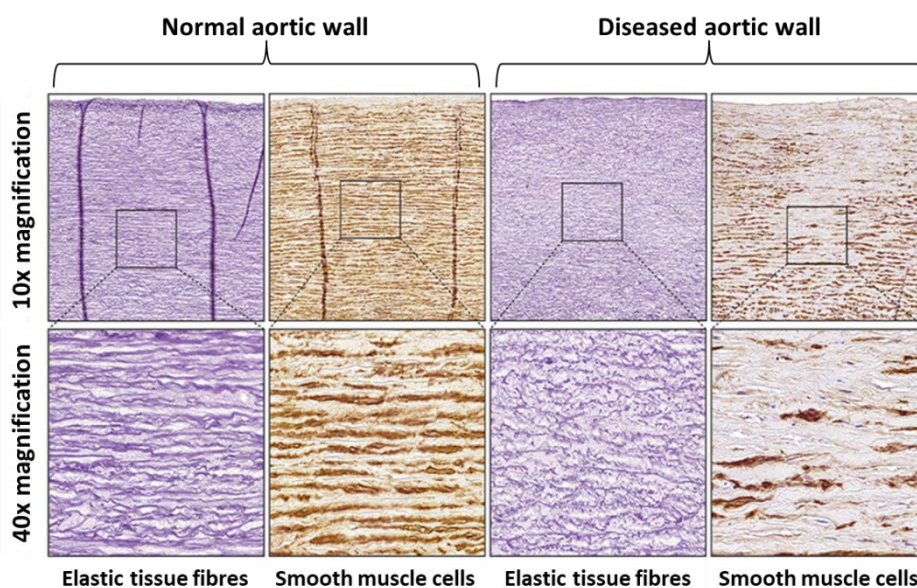


Figure 2.5: Structure of a normal and a diseased aortic wall, showing both elastic tissue and smooth muscle cells (adapted from van de Pol, 2017).

Cystic medial degeneration is usually associated with aging, but it also can be seen in some younger people, especially individuals who have a genetic disorder (Rubin, 1999, p.522; Underwood, 2000, p.278–279). The different types of genetic disorder which can have an effect on aneurysmal formation are discussed in greater detail over the following pages.

2.3.5 Genetic disorders associated with aortic aneurysms

The gene mutations that can lead to the formation of aortic aneurysms are in the early stages of being identified. To date, 30 genes have been shown to have an association with the development of thoracic aortic aneurysms or dissection (Brownstein, 2018). Genes that have been recognised as being important include: *FBN1*, *TGFBR1*, *TGFBR2*, *COL3A1*, *ACTA2* and *MYH11*. Mutations of these genes, and the syndromes associated with these mutations, can cause a variety of clinical features that may contribute to aneurysmal development and growth. These associated genetic features may have consequences for patient treatment, especially if they are cardiovascular in nature. Or if the patient has other coexisting clinical issues to consider then their recovery or care may have to be adapted to take them into account. Some examples of these features are:

- Bicuspid aortic valve (BAV). A normally developed aortic heart valve has three leaflets (known as tricuspid), which open and close when the heart beats. In BAV's two of these leaflets are fused together when the heart is growing in the womb. This results in a disrupted blood flow into the aorta (McKellar, 2007). More detail on BAV is provided in Section 2.3.6.1.
- Livedo reticularis, which presents as a mottled, purplish discolouration of the skin, where small blood clots have obstructed capillary vessels (Sneddon, 1965).

- Iris floccule, where tiny cysts in the eye have a constant progression of breaking down and building up again. This presents as wrinkled blobs on the margin between the iris and the pupil. Patients do not usually report any problems with vision (Shields, 2016).
- Patent ductus arteriosus, which is a condition seen after birth when the ductus arteriosus fails to close. The ductus arteriosus is a blood vessel that connects a foetus's main pulmonary artery to the proximal descending aorta, allowing oxygen-filled blood to bypass the lungs. In a typical birth this closes off and becomes the ligamentum arteriosum (see Section 2.3.2 and Zhu, 2006).
- Thin, translucent skin
- Arterial tortuosity, this rare disorder is characterised by elongation, stenosis and tortuosity (or an excessively frequent twisting) of a person's arteries. This arterial dysfunction is caused by genetic mutation of the elastic fibres in the medial layer of the artery (see Section 2.3.2 and Couke, 2006).

2.3.5.1 Marfan syndrome

A French paediatrician called Antoine-Bernard Marfan was the first to describe the skeletal abnormalities, including an overgrown frame and soft, hyper-flexible joints which typify the connective tissue disorder eventually named after him (Marfan, 1896; Verstraeten, 2016).

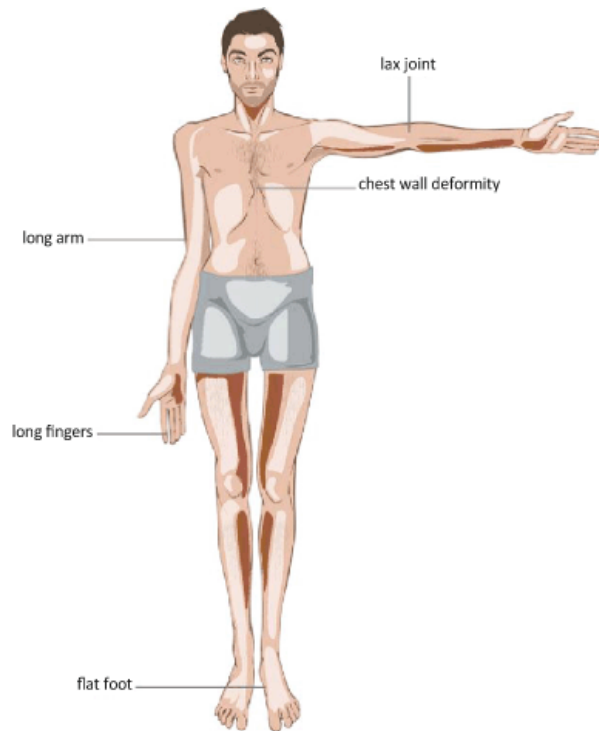


Figure 2.6: General manifestation of Marfan syndrome (Reprinted by permission from Springer Nature: image originally in “Genetics and Dismorphology” by Naga et al, Copyright 2015).

Since that time, many other clinical features have been linked to the disease. These include several eye, skin, and cardiovascular presentations such as lens displacement, short-sightedness, loose skin, stretch marks, aortic aneurysms and floppy heart valves. The cardiovascular problems are understandably the major culprits for the high rates of morbidity and mortality in patients who have Marfan syndrome (Cook, 2015).

The gene mutations that cause Marfan syndrome have been isolated to the *FBN1*, or fibrillin 1, gene (Dietz, 1991). *FBN1* encodes a type of protein which is used by the body to create tissue fibres, including bone. *FBN1* is particularly used for fibres that contain important qualities such as elasticity and structural support.

Patients with Marfan syndrome are highly predisposed to thoracic aortic aneurysms disease or aortic dissections, virtually every patient diagnosed with the syndrome has evidence of aortic disease at some point during their

lifetime (Hiratzka, 2010). The population incidence of Marfan syndrome is approximately 1 in 3,300 worldwide (Arslan-Kirchner, 2010).

2.3.5.2 Loeys-Dietz syndrome

Loeys-Dietz syndrome is a genetic disorder with similar characteristics to Marfan syndrome, in that it is a genetic dysfunction which affects connective tissue in the body. The disorder was first observed and described by Dr's. Bart Loeys and Harry Dietz at the Johns Hopkins University School of Medicine in 2005 (Loeys, 2005). A key defining feature of patients with the disease is the presence of aortic aneurysms or dissection (Loeys, 2006).

As well as aneurysmal formation, patients with Loeys-Dietz syndrome typically present with three common physical abnormalities, arterial tortuosity (see Section 2.3.5), wide spaces between the eyes and bifid uvula or a cleft palate, or a palate with a wide base and prominent ridge. Other features of these patients include: velvety and translucent skin which can bruise easily, a malformed skull, jaw or lower face, a blue tint to the whites of the eyes, patent ductus arteriosus (see Section 2.3.5), atrial septal heart defects, developmental delays or learning disabilities, spine abnormalities and joint hyper-flexibility (MacCarrick, 2014; Hiratzka, 2010).

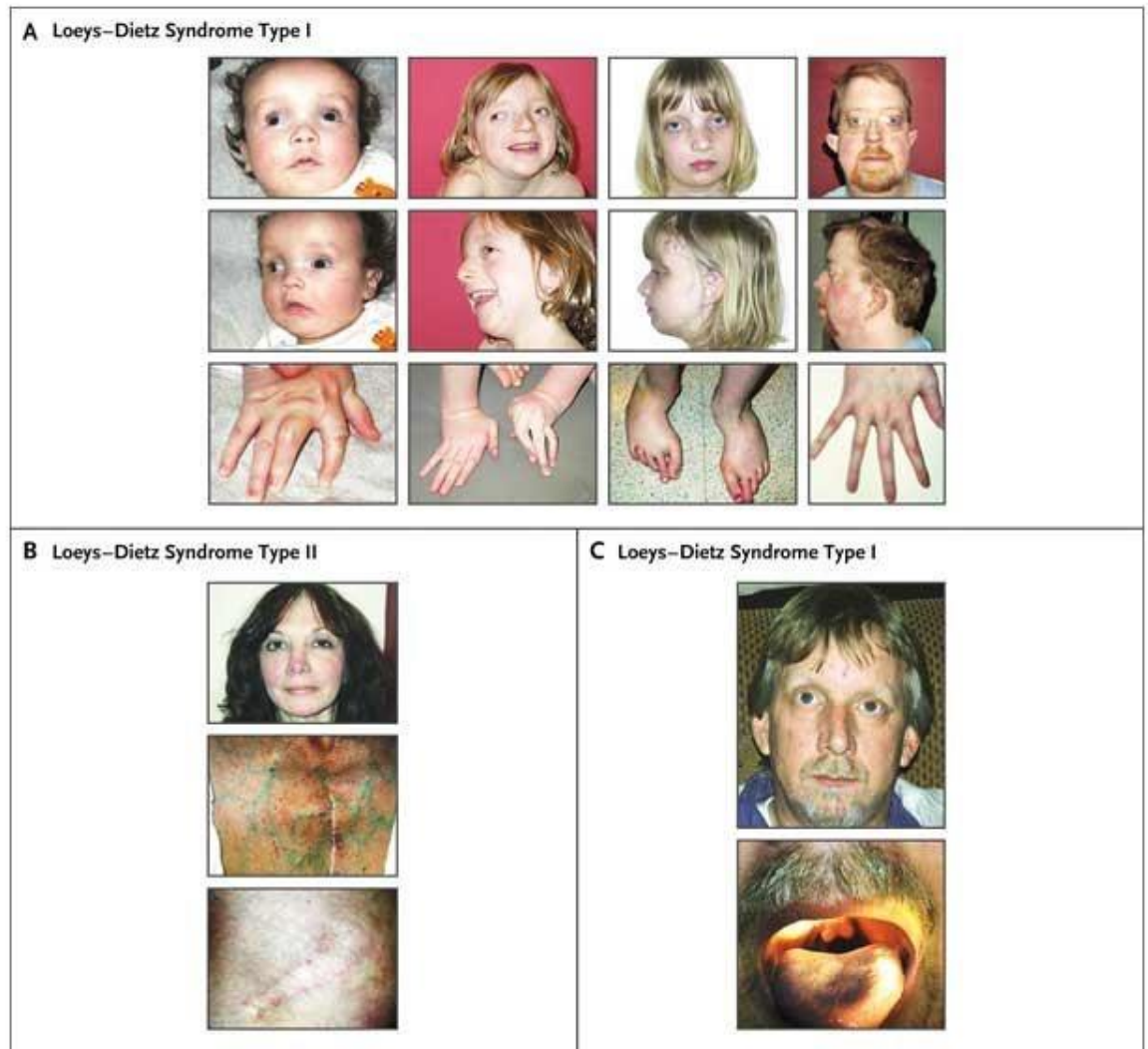


Figure 2.7: Characteristics of the Loey-Dietz Syndrome. Panel A shows typical facial characteristics of patients with Loey-Dietz syndrome type I at different ages. Panel B shows the facial characteristics of a patient with Loey-Dietz syndrome type II. The translucency of the skin is evident, with visible veins and distended scars. Panel C shows a patient who had type I with a bifid uvula (Loeys, 2006. Reproduced with permission from the New England Journal of Medicine, Copyright Massachusetts Medical Society).

As well as the aneurysmal formations and/or aortic dissections that define Loey-Dietz syndrome, the gene mutations that cause the syndrome have been identified as occurring in the *TGFBR1*, *TGFBR2*, *SMAD2*, *SMAD3*, *TGFB2* and *TGFB3* genes (Loeys, 2018). The four *TGF* genes are in the transforming growth factor category, these types of gene are essential for

tissue regeneration, cellular development and immune system regulation (Matt, 2009). The two *SMAD* genes are part of the *TGF* genetic process, they are activated by the *TGF* gene and combine to form protein-based cellular building blocks for creating tissues. Mutations in these genes lead to the production of non-functional cells which leads to physical abnormalities in blood vessels, cartilage and skin development (van de Laar, 2011). The population incidence of Loeys-Dietz syndrome is unknown, although mutations affecting *TGFBR1* and *TGFBR2* appear to be the most common.

2.3.5.3 Ehlers-Danlos syndrome

Ehlers-Danlos syndrome (EDS) is a group of genetic disorders that are characterised by abnormalities of the skin, joints and connective tissues, particularly stretchy or hyper-elastic skin, hyper-flexible joints and cardiovascular malformations. Although hypermobility was first described in 400 BC by Hippocrates (Beighton, 2011), and a comprehensive description of loose, fragile skin and hypermobile joints was published in late 19th century Russia by Tschernogobow (1892), the syndrome is named after two dermatologists – Edvard Ehlers and Henri-Alexandre Danlos – one Danish and one French respectively, who independently published their findings in the early 20th century (Ehlers, 1901; Danlos, 1908).

The system of classifying patients with EDS currently contains 13 different types. The type most associated with aneurysmal formation is Type IV, or vascular EDS. Patients who present with vascular EDS typically have very thin, fragile skin which bruises easily and has visible veins, they are also usually of short stature, have thin scalp hair and possess distinguishing facial features including large eyes, a small chin, sunken cheeks, thin noses and lips, and ears without lobes (Inokuchi, 2014). Internal arteries are particularly delicate and are predisposed to rupture, so surgical interventions are often limited to those life-threatening scenarios where aortic rupture occurs (Hiratzka, 2010), surgeons who have attempted arterial reconstruction in

these patients have found the task a formidable challenge and advise to carry out only the most straightforward procedures (Ascione, 2000).

The diagnosis of vascular EDS is based on mutations to the *COL3A1* gene. This gene provides instructions for making type III collagen, which is a protein that provides strength and support to many different types of bodily tissues (Kontusaari, 1990).

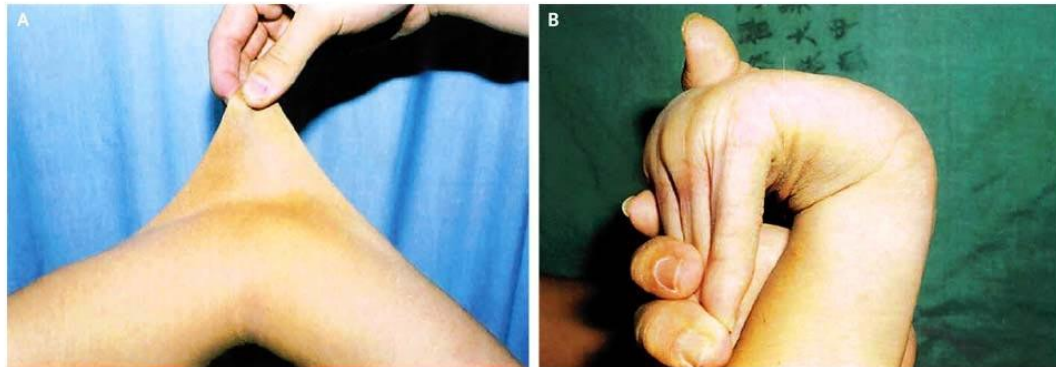


Figure 2.8: Examples of skin and joint presentations of EDS from Chen (2014). Skin hyperextensibility (A). Joint hypermobility at fingers (B). (Reproduced with permission from the New England Journal of Medicine, Copyright Massachusetts Medical Society).

The combined prevalence of all types of Ehlers-Danlos syndrome varies between 1 in 10,000 and 1 in 25,000 individuals worldwide. EDS type IV represents approximately 5 to 10% of cases (Germain, 2007)

2.3.5.4 Turner syndrome

Turner syndrome is a condition that occurs in females where they are partially or completely missing their X chromosome. It is named after Henry Turner, who in 1938 was the first to describe the disorder in detail (Turner, 1938).

As well as developmental, endocrine and reproductive problems, Turner syndrome also has a detrimental effect on the cardiovascular system. The

most serious manifestations of this are bicuspid aortic valves (between 10% and 25% of patients) and aortic coarctations (approximately 8% of patients, see Section 2.3.1). Both of these conditions carry with them an increased risk of aneurysmal formation and aortic dissection (Hiratzka, 2010; Bondy, 2007).

Some characteristics of Turner Syndrome (from Carr, 2014) include:

- Short stature
- Low hairline
- Fold of skin around the neck
- Constriction of the aorta
- Shield-shaped thorax
- Widely spaced nipples
- Elbow deformity
- Brown spots on the skin

Early identification and regular monitoring of the cardiovascular elements of Turner syndrome is necessary, including blood pressure, echocardiogram and clinical examination by a cardiologist. When significant cardiovascular problems occur, the most appropriate medical or surgical repair is advised (Gravholt, 2002).

Turner syndrome affects approximately one in 2500 live-born females (Nielsen, 1991).

2.3.6 Cardiovascular conditions associated with aortic aneurysms

Whether they are caused by a genetic syndrome, familial inheritance or developmental abnormalities, understanding how the following cardiovascular conditions manifest themselves, and the associated aneurysmal risk that they carry, is important when presenting an overview of aortic disease.

2.3.6.1 Bicuspid aortic valve

BAVs are one of the most common congenital heart defects reported, recent estimates put the prevalence of the condition between 0.5% – 1.4% of the general population (Braverman, 2011).

The earliest description of a BAV has been attributed to Leonardo da Vinci, who drew a bicuspid variant of the aortic valve in the early 16th century. The drawing was found amongst his numerous sketches regarding heart function and blood circulation, along with a note describing how the typical tricuspid valve has a more optimal configuration than a quadricuspid valve. (Mills, 1978; Braverman, 2005).

More recently BAV was referred to by Hunter in 1764, who described the case of a man with a BAV afflicted with severe disease and by Paget in 1844 who noted that patients with the condition were far more likely to develop disease on the leaflets of their aortic valves. In 1858, Peacock recognised that BAVs seemed more likely to harden and so become less flexible and effective than tricuspid configurations, and Osler (1886) saw a greater tendency for these valves to become infected. By the 1950's, clinicians began to appreciate how BAVs were intrinsically associated with aortic stenosis (where the aortic valve narrows over time), aortic regurgitation (where the aortic valve becomes leaky and blood begins to flow in the reverse direction), infection and aortic dissection (Campbell, 1953; Smith, 1955 and Bacon, 1959), or often a combination of these pathologies.

The leaflets of a BAV are usually of an unequal size, with a groove or ridge (known clinically as a raphe) noted in the larger leaflets, see Figures 2.9a and 2.9b, below. This groove is an indication of where the original tri-leaflet valve has fused together. The edges of the two BAV leaflets are limited in mobility when compared to tricuspid valves, the result of this is an increase in the stresses and strains being put on the valve during the course of contraction during a heartbeat. BAVs have been seen to undergo more significant folding and wrinkling while the flow of blood through the valve is more turbulent and irregular (Robicsek, 2004).

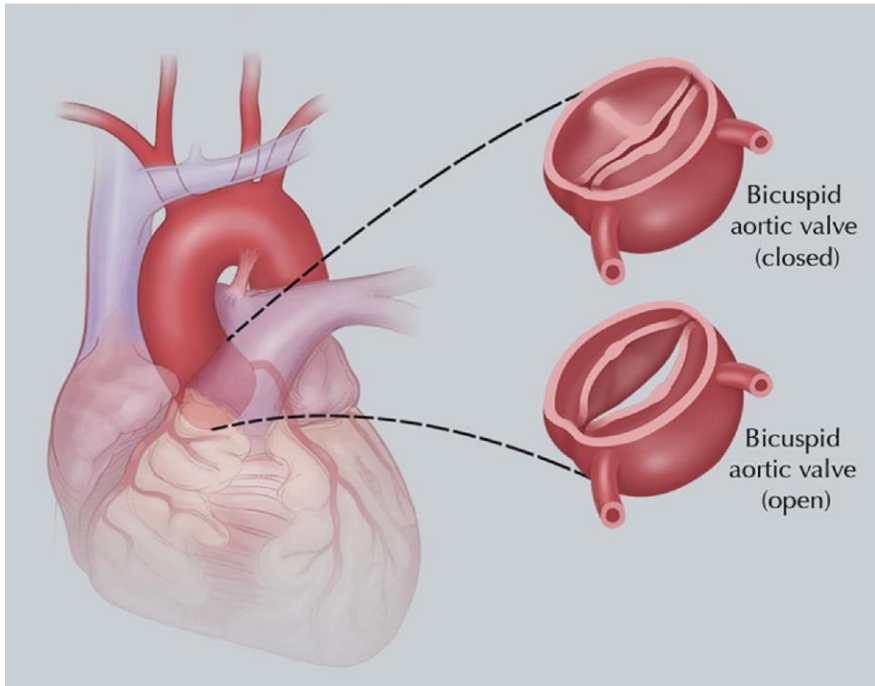


Figure 2.9a: Appearance of a Bicuspid Aortic Valve (Reprinted with permission from the Journal of the American College of Cardiology, Copyright Siu, 2010).

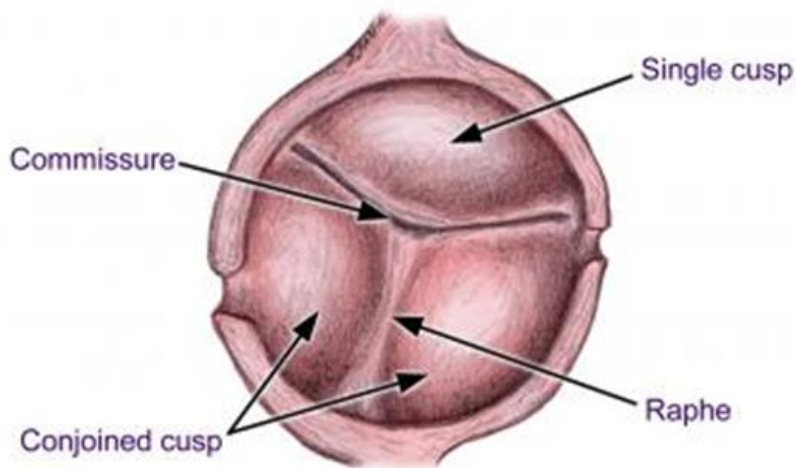


Figure 2.9b: BAV, detail (Bayne, 2016. Image reproduced with permission from Medscape Drugs & Diseases (<https://emedicine.medscape.com/>), Copyright 2016).

The first suggestion of a link between BAV and aortic aneurysms was made by Abbott in 1928. In the following years many studies have confirmed this link between BAV and aortic aneurysms, even if the BAV is functioning well and showing no signs clinical dysfunction (Nkomo, 2003; Morgan-Hughes, 2004). Patients with BAV have been found by Fazel (2008) to be more likely to develop aneurysms in all the proximal segments of the aorta (aortic root, ascending aorta and aortic arch), with 73% of his sample having some dilation of the aortic arch. Overall, aortic dilatation is one of the most common findings in patients with a BAV, with reported incidence rates between 30% and 70% (Losenno, 2012).

These aneurysmal formations represent a clear risk factor for aortic dissection, the connection of BAV with this critical outcome was highlighted by Larson and Edwards in 1984, who calculated a nine-fold greater risk of aortic dissection in BAV patients. Subsequent investigations have found that the rate is probably not quite so high, but individuals with a BAV are still at a significantly higher risk than the general population (Tzemos, 2008; Michelena, 2011).

2.3.6.2 Aberrant right subclavian artery

An aberrant right subclavian artery (ARSA; also known as 'Arteria Lusoria') is a physiological anomaly first described in 1735 by Hunauld. It has a reported incidence of between 0.5% and 2.0% in the general population (Epstein, 2002).

The irregular arterial structure of an individual with an ARSA is shown in Figure 2.4. Compared with the normal arrangement (see Figure 2.1) the innominate artery (also known as the brachiocephalic trunk), which appears closest to the aortic valve and then branches out into the right common carotid artery and the right subclavian artery, is missing. Instead, four arteries emerge directly from the aortic arch in the following order: the right common carotid artery, the left common carotid artery, the left subclavian artery, and

then finally the ARSA. In most cases, the ARSA then crosses behind the esophagus (not labelled in the figure) to supply blood to the right arm.

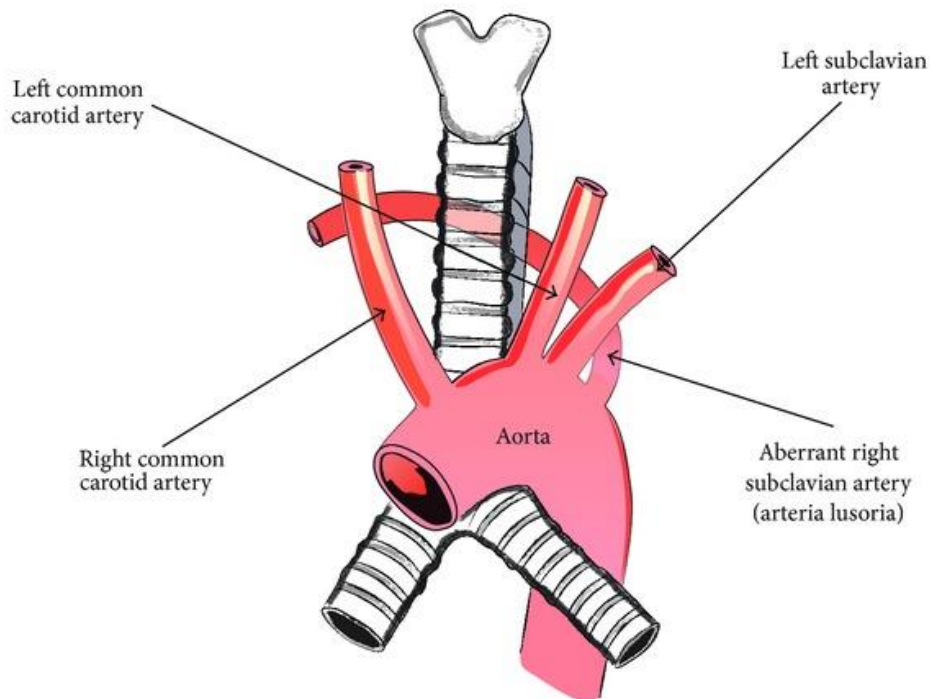


Figure 2.10: Aortic arterial structure with the presence of an ARSA (Polguy, 2014)

In many patients who have an ARSA, the aorta itself is also abnormal and is prone to aneurysm formation, dissection, and rupture (Hiratzka, 2010). It is possible for patients with an ARSA to have no symptoms that would indicate their presence, but they can also cause symptoms such as dysphagia (problems swallowing), shortness of breath or chest pains (Kedora, 2009).

2.3.6.3 Kommerell diverticulum

A Kommerell diverticulum (KD) is a developmental error that can occur within a number of aortic arch structural anomalies (see Figure 2.11). KD's can become aneurysmal when a weakened proximal section of an artery enlarges. The condition takes its name from a German radiologist, Dr. Buckhard F. Kommerell, who in 1936 made the first report in a living patient. Kommerell's original diverticulum was from a patient who had an ARSA (see Figure 2.10), but KD's are most frequently found in cases of right aortic arch with an aberrant left subclavian artery (van Son, 2002).

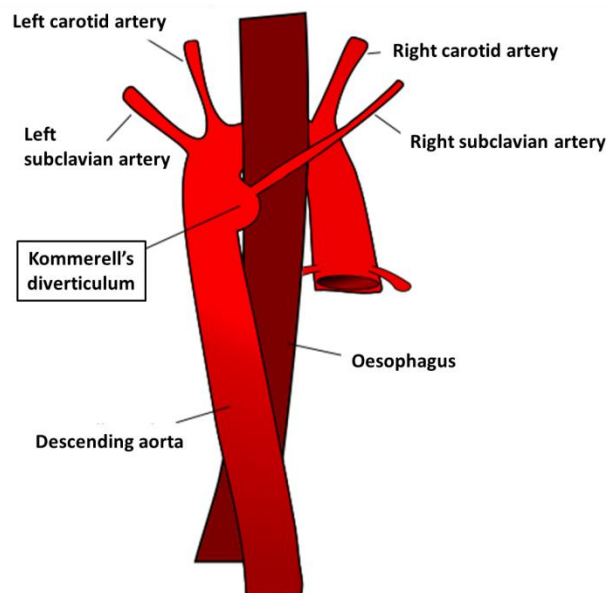


Figure 2.11: Diagram of a Kommerell diverticulum and its relationships with the oesophagus, the aorta and its collateral branches (posterior view) (adapted from Adert, 2013)

Kouchoukos (2007) described a surgical series of 10 KD patients over the course of 10 years, noting that the rarity of the condition meant that the natural history was not yet known with certainty. However, it has been observed that KD patients seem more susceptible to embolisation, internal compression of the trachea or oesophagus, aortic dissection and aortic rupture. Kouchoukos goes on to recommend a surgical technique for KD patients, reporting acceptable results.

2.3.6.4 Coarctation of the aorta

The first reported case of coarctation of the aorta (CoA), or aortic narrowing, was reported by Paris in 1791. It has since been recognised as a relatively common genetic abnormality, with both paediatric and adult cases being described before surgical treatment was feasible by Abbott (1908, 1928) and Lewis (1933).

CoA can occur anywhere in the aorta, but is usually found in the distal arch segment (see Figure 2.12). It is a relatively common abnormality that occurs in about 40 to 50 of every 100 000 live births, with a 2:1 ratio in males versus females (Hiratzka, 2010). CoA's are normally diagnosed in early life, and treated surgically at that time. Adult presentations are therefore typically due to subsequent complications or failures of the initial treatment. Choudhary (2015) reports aneurysmal formations in 15% of individuals, with the most common occurrences being noted in patients who had a patch aortoplasty procedure to treat their CoA.

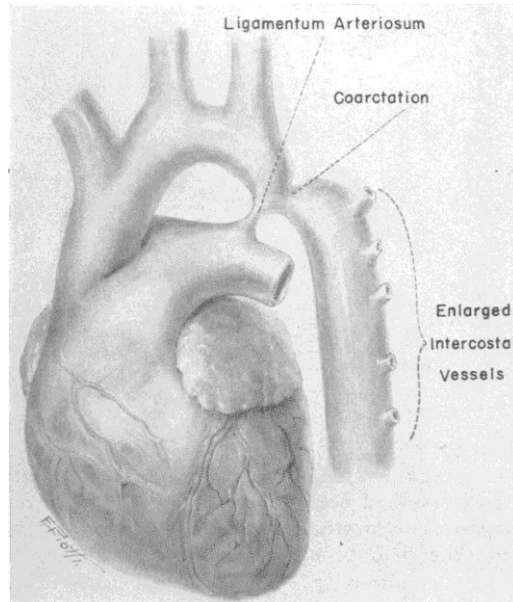


Figure 2.12: Coarctation of the Aorta from Robbins (1953). (Reproduced with permission from the New England Journal of Medicine, Copyright Massachusetts Medical Society).

2.3.7 Inflammatory diseases associated with thoracic aortic disease

Inflammatory diseases of the aorta are rare compared to genetic malformations, atherosclerosis and degenerative changes, which are the most frequent causes of aortic disease. However, these diseases can quickly damage the aortic tissue and result in aneurysmal formations (Caspary, 2016). Some of the most prominent diseases of this type, along with some introductory references, are listed below.

- Takayasu arteritis (Numano, 2000; Johnston, 2002)
- Giant cell arteritis (Evans, 1995; Salvarani, 2008)
- Behçet's disease (Sakane, 1999; Seyahi, 2016)
- Ankylosing spondylitis (Haroon, 2015)
- Cogan's syndrome (Kessel, 2014; Angiletta, 2015)
- Polyarteritis nodosa (De Virgilio, 2016; Guillevin, 2017)
- Kawasaki disease (Newburger, 2016)

2.3.8 Other pathologies related to aortic aneurysms

It is common for patients with an aortic aneurysm to experience other problems with their heart or heart valves. The presence of these additional abnormalities can make surgery more complex and therefore increase the risk of undesirable outcomes. The most common of these pathologies are covered in more detail in the following sections.

2.3.8.1 Aortic stenosis

Patients with a BAV or other congenital anomalies are at a greater risk of suffering from aortic stenosis (see Section 2.3.6.1), but the condition more commonly develops during aging as calcium deposits and scarring on the

valve leaflets increase. These degenerative developments damage the valve and restrict the amount of blood that can flow through it (American Heart Association, 2019). Figure 2.13 shows the differences in performance between a normal, tricuspid heart valve and a tricuspid valve with aortic stenosis.

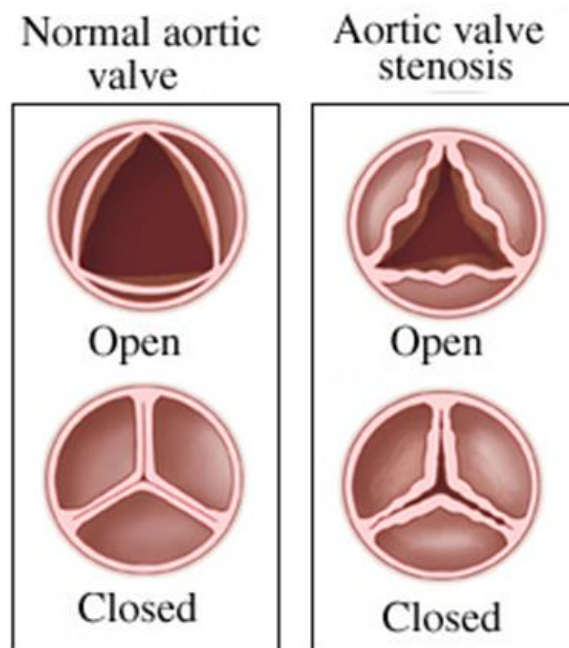


Figure 2.13: Normal aortic valve versus stenosed aortic valve (Šušak, 2013).

2.3.8.2 Aortic regurgitation

When an aortic valve is regurgitative it is because the leaflets of the valve are not closing properly. This causes blood to leak backward from the aorta into the left ventricle, as illustrated in Figure 2.14:

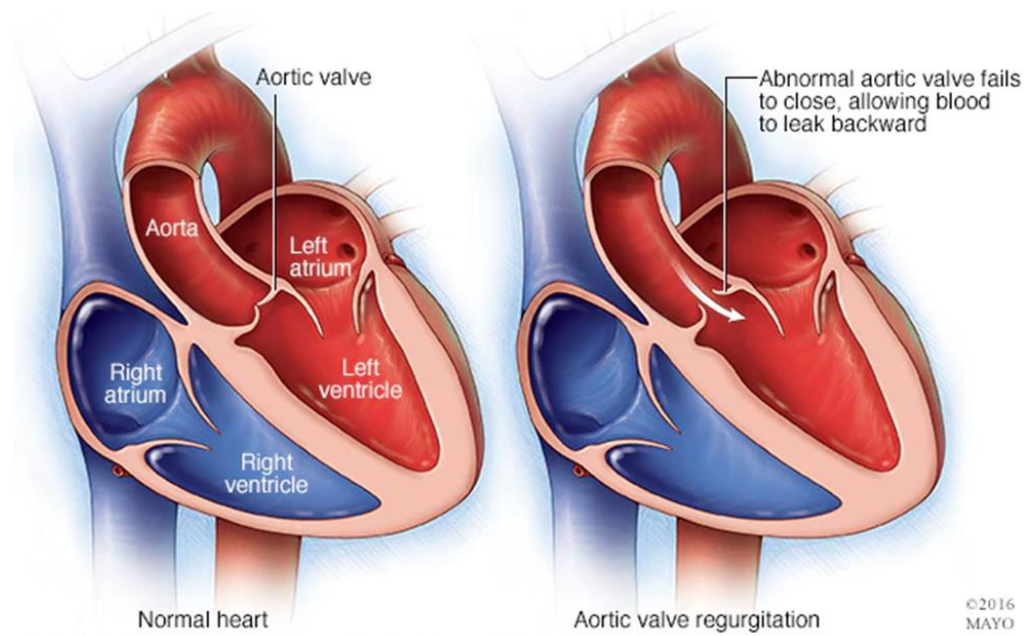


Figure 2.14: Aortic valve regurgitation (Mayo Clinic, 2019, used with permission of Mayo Foundation for Medical Education and Research, all rights reserved).

As the heart is not working efficiently, blood does not flow through the body as it should. Mild aortic regurgitation may have few symptoms, but more severe cases can provoke symptoms such as shortness of breath, fatigue or chest pain and if left untreated can lead to ventricular dysfunction and heart failure (Enriquez-Sarano, 2004).

2.3.8.3 Ventricular dysfunction

The left ventricle (see Figure 1.1) is the heart's main pumping chamber. Ventricular dysfunction occurs when the heart is not pumping as much blood as it should be. A common measure of how well the left ventricle of the heart

is pumping blood is the left ventricular ejection fraction (LVEF); this is a percentage expression of how much blood in the left ventricle is pushed out with each heart-beat. A healthy heart has an LVEF greater than 50%, moderate LVEF is defined as between 31% and 50%, poor LVEF is between 21% and 30% and very poor LVEF is 20% or less (Nashef, 2012). Patients with a reduced LVEF may experience shortness of breath, heart palpitations, fatigue, swelling in the lower extremities or a lack of appetite.

2.3.9 Symptoms

Thoracic aortic aneurysms can often develop without generating physical symptoms. Patients who have asymptomatic aneurysms are regularly diagnosed as a consequence of having an imaging scan (such as a chest x-ray, an echocardiogram or a computerised tomography (CT) scan) for some other health problem (Isselbacher, 2005).

Symptomatic patients, who account for between 5%–10% of cases (Sawyer, 2017), present with pain caused by the aneurysm. This pain can be due to the stretching of aortic tissue, or as the aneurysm grows it may begin to squash against other parts of the body.

If patients have pain, the site can indicate the location of the aneurysm, so proximal thoracic aortic aneurysms can cause pain in the neck and jaw, sternum or upper back. Aneurysms in the more distal descending aorta may produce pain in the left shoulder or between the shoulder blades, and abdominal aneurysms may trigger pain in the flanks, abdomen or lower back (Elefteriades, 2008).

Examples of symptoms that are associated with internal compression are: voice hoarseness, when nerves in the larynx are squashed; a high-pitched wheezing (known as 'stridor') if the larynx or trachea are compressed; difficulty breathing, or dyspnoea, if the lungs are affected; problems swallowing, if the oesophagus is compressed; and if the vena cava becomes

compressed this can result in plethora and oedema (swelling caused by an excess of blood).

Depending on how and where the aneurysm develops, it may stretch out the aortic valve structure and have a detrimental effect on its performance. The valve may become leaky or the leaflet movement may be impeded by the aneurysmal growth. This would then result in a heart murmur detectable upon examination (Hiratzka, 2010).

2.3.9.1 Aortic dissection

As mentioned in Section 2.3.3, aortic dissection often occurs as a consequence of an aortic aneurysm. This occurs when the aneurysm expands and the walls of the artery rupture, the aorta breaches and normal blood circulation is interrupted.

Aortic dissections are classified as either 'acute', which represent life-threatening clinical emergencies (Hagan, 2000), or 'chronic', which are still serious conditions but in these cases the arterial failure occurs more gradually (Beebeejaun, 2013).

Reports of aortic dissections have been made for over 200 years (Acierno, 2014), with one of the most famous early descriptions being made on the body of King George II after his death in 1760:

“...the next day Dr Nicholls, physician to his late Majesty, found the pericardium [the protective sac which covers the heart] distended with a pint of coagulated blood, probably from an orifice in the right ventricle, and a transverse fissure on the inner side of the ascending aorta 3.75 cm long, through which blood had recently passed in its external coat to form a raised ecchymosis [bruise], this appearance being interpreted as an incipient aneurysm of the aorta” (Leonard, 1979)

Estimated incidence of aortic dissections in the UK was 3,892 in 2010, based on Office of National Statistics 2010 census population figures. This corresponds to around 4 – 7 cases per 100,000 individuals (Howard, 2014). It is estimated that around a third of all dissection cases are chronic (Patel, 2014).

Patients who suffer from an aortic dissection report an abrupt onset of sharp, severe chest pain. Back or abdominal pain is also reported, typically linked to the location of the dissection.

2.3.10 Identification, diagnosis and surveillance

The clinical presentation of an aortic aneurysm can vary and may be similar to other more common health problems. If thoracic aortic aneurysms are to be identified at an early stage then a clear medical history, appropriate physical examination, suitable diagnostic studies, and skilled clinical management is important to achieve a positive outcome (Klein, 2005).

There are several diagnostic studies that can help to identify aortic aneurysms and offer useful information about size and location:

- Chest x-rays often raise suspicions about the presence of aortic aneurysms. They can clearly identify abnormal arterial formations, but are less sensitive when more detailed information is required (von Kodolitsch, 2004)
- Transthoracic echocardiography (TTE) can also provide suggestions of aneurysmal problems, but due to the limited coverage it provides it is not comprehensive enough for a full evaluation (Shiga, 2006). Transoesophageal echocardiography (TOE) has more value, especially if the aortic root and the aortic valve are the areas that the clinician is interested in, however it is a relatively invasive procedure, so is not used as a matter of routine (Holloway, 2011)

- CT scanning is currently the most widely employed technique for the study of the thoracic aorta (Di Cesare, 2016). The speed, convenience and precision of the technique give it an advantage over the other scanning methods. One drawback of CT scans is the amount of radioactive dose exposure that is involved. This makes repeat scanning, especially in younger people, a particular concern (Pearce, 2012).
- Magnetic Resonance Imaging (MRI) scanning has a similar, or perhaps greater, capability to CT scanning when comparing sensitivity and specificity of diagnostic images. It also has less of a radioactivity burden, making it preferable in younger patients or patients who may require multiple scans. Some weaknesses of MRI scanning are that the patient may feel claustrophobic in the scanning machine, the processing times for scans is longer and the presence of metal artefacts, such as aortic stenting or a pacemaker, can result in suboptimal images (Holloway, 2011).

Figure 2.15 shows an example of a CT scan with a large ascending aortic aneurysm indicated. For more details on imaging and aortic measurements see McComb (2016), who discusses reference values for treatment and normative sizes of aortic diameter. McComb found that while smoking did not appear to affect the diameter of an aorta, increasing age, male gender and increasing body surface area were all associated with a broadening of the artery.



Figure 2.15: CT scan showing a 7.5cm×8.3cm ascending thoracic aortic aneurysm. A indicates ascending; D, descending (Isselbacher, 2005. Reproduced with permission, copyright Circulation, AHA Journals 2005).

When a patient has been diagnosed as having a thoracic aortic aneurysm, it may not be appropriate to treat the malformation straight away as the risks of a poor outcome from an invasive surgery could be larger than the risks posed by the aneurysm itself. Many patients who have aneurysms therefore do not undergo immediate surgery, but are rather put under surveillance so the growth rate of their aneurysm can be monitored, and appropriate medical treatment given (Davies, 2002).

Size thresholds for patient monitoring and treatment vary based on various factors including age, body mass, ethnicity, the position of the aneurysm, aortic valve pathology and the presence of congenital disorders. Typically, a proximal thoracic aortic diameter of 3.5 cm is considered dilated and would require a follow up scanning regime to monitor potential growth (Wolak, 2008).

Current guidelines (Hiratzka, 2010) advise that in patients who have no genetic co-morbidities, surgical intervention should be administered for ascending thoracic aortic aneurysms when they reach 5.5 cm in size. In patients who have a genetic disorder, such as Marfan syndrome, the threshold for surgical intervention is set lower, at 5 cm. These guidelines are

based on data showing that at these sizes the risk of complications following an aortic rupture increases beyond the risk of complications from the surgical intervention, making the surgery preferable. Figure 2.16 illustrates the risks of rupture or dissection for increasing size of the ascending aorta, the 'hinge point' for greatly increased risk can be seen at 6cm, supporting the current strategy for intervention.

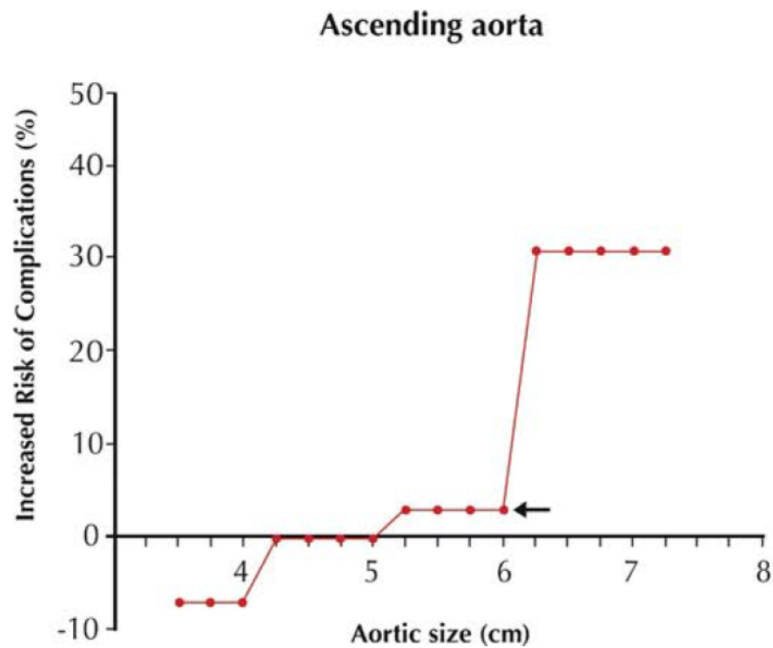


Figure 2.16: Illustration of hinge points for lifetime risk of rupture or dissection at various sizes of the ascending aorta (adapted from Coady (1997) in Elefteriades, 2010. Reprinted from Journal of the American College of Cardiology, Copyright (2010), with permission from Elsevier).

2.3.11 Treatment

2.3.11.1 Surveillance

Patients who have been diagnosed with a thoracic aortic aneurysm, but do not yet meet the criteria for surgery and have been placed under surveillance (see Section 2.3.9), do not constitute the target population of the following study. However, it is instructive to note the various types of medical advice

which is often given to these patients, along with regular scans to monitor aneurysmal growth.

Aneurysmal patients often have concomitant coronary disease, or other comorbidities including respiratory illnesses, high blood pressure and high cholesterol. The possibility of surgery at some point in the future means that these patients should aim to be in their optimal physical condition. If patients can positively modify their lifestyle to take exercise regularly, eat healthily, moderate alcohol intake and stop smoking (where applicable), this will increase their chances of having successful surgery if and when their aneurysm requires intervention (Smith, 2006).

2.3.11.2 Surgery

The indications for different types of surgical treatment depend on the location of the aneurysm and the presence of simultaneous disease in either the aortic valve or the arteries of the heart.

In general terms, there are two approaches: open surgery and endovascular surgery. These are covered in more detail in the following sections.

2.3.11.3 Open heart surgery

Historically, open surgery has been used to treat aneurysms located in the root, ascending and arch segments of the aorta. This involves making an incision in the sternum, opening the ribcage and using medical prostheses to replace the dilated segments of the aorta.

As mentioned above, patients who present with proximal thoracic aortic aneurysms often need secondary treatments for additional medical issues. Procedures that may be performed together with the aneurysm repair include: coronary artery bypass graft surgery (Eagle, 2004), valve replacement or repair (Nishimura, 2014), repair of cardiac septal defects

(Warnes, 2008), closure of vascular fistulas, and ablative therapy for arrhythmias (Calkins, 2007).

Over the years, several different styles of operation have been popularised including: the Bentall procedure (Bentall, 1968), or the Cabrol procedure (Kourliouros, 2011) where the aortic valve, aortic root and ascending aorta are all replaced; aortic valve sparing treatments, where the proximal aorta is diseased, but the valve does not require treatment (David, 2012); and 'elephant trunk' procedures (Svensson, 2004), where the aortic arch is replaced with a prosthesis that extends down the aorta, making it accessible for an either planned or probable subsequent secondary operation to repair a more distal aneurysm.

2.3.11.4 Endovascular surgery

Historically, endovascular surgery, or endovascular aneurysm repair (EVAR), has been used to treat aneurysms located in the descending and thoracoabdominal segments of the aorta. These procedures involve a more minimally invasive approach with a surgical incision through the groin. Catheters and guidewires are then used to deploy a prosthesis that replaces the affected area of the aorta (Moll, 2011).

Recently, investigators have suggested that endovascular treatments may become more popular with operators treating the proximal aorta as techniques, materials and the understanding of aneurysmal disease continue to advance (Klonaris, 2016; Harky, 2018)

2.3.12 Adverse treatment outcomes and post-operative care

2.3.12.1 Likelihood of adverse surgical outcomes

Patients who undergo open proximal aortic surgery have a relatively high risk of operative mortality. They are also at risk of life-changing complications

such as stroke or neurocognitive deficit, as well as other undesirable outcomes such as reoperations, infections, respiratory failure and changes in voice such as hoarseness or differences in tone (Hiratzka, 2010).

Reported operative mortality rates rise as the invasiveness of the procedure increases, expected mortality rates are summarized below:

- Patients who receive valve-sparing root replacements ('David' procedures, see Section 2.3.10.3) tend to be younger and have better overall health. Correspondingly, in-hospital mortality rates have been reported around 1% to 1.5% (David, 2007; Patel, 2008; Svensson, 2007a)
- In procedures where the aortic valve is bicuspid and the ascending aorta is replaced, Svensson (2007a, 2007b) reports a risk of operative mortality of 1.5%.
- Composite valve grafts and valve replacement with ascending aortic repair carry an operative mortality risk of between 1% and 5% (Crawford, 1989; Svensson, 2000).
- In arch replacement operations, a risk of death between 2% and 6% has been reported (Kazui, 2007; Sundt III, 2008; Spielvogel, 2005)
- Cases where the patient is returning for a reoperation on their aorta also carry with them some additional risk. Operative mortality rates for these patients are reported as being between 2% and 6% (Hirose, 2004).

Factors other than how invasive the operation is are also strongly associated with increased operative mortality, these include emergency priority, advancing age, concomitant cardiac disease and patient comorbidities such as renal dysfunction, poor lung function or irregular heart rhythms (Bashir, 2016).

Although rare, it should be noted that spinal cord injuries such as paraplegia can occur in these patients as a result of malperfusion. Paraplegia and paraparesis are usually associated with aortic dissection and circulatory arrest, but is reported to occur in approximately 1% to 3% of thoracic aortic aneurysm patients (Hiratzka, 2010; Sundt, 2004).

Permanent stroke complications are reported as occurring in between 2% and 8% of cases (Svensson, 2002; Svensson, 2007a). Protecting the brain is a key consideration when operating with cardiopulmonary bypass or circulatory arrest techniques (see Section 2.3.11.2).

Reoperation for bleeding is more common in the more difficult surgeries, this increased difficulty can increase the time a patient spends under cardiopulmonary bypass. Zehr (2004) and Motomura (2008) report rates of between 1% and 6% of this complication in their comprehensive series.

Motomura (2008) also reports an incidence of superficial infections of around 1% to 5% and a rate of less than 1% for more serious infection. Contamination, patient obesity or lung disease may be contributing factors.

Respiratory failure can occur in around 5% to 15% of patients. Standard tests for lung function can be carried out preoperatively to alert healthcare providers to the risks of this happening on a patient-by-patient basis (Hiratzka, 2010).

Changes to the voice can occur when the left recurrent laryngeal nerve (which loops under the aortic arch) is damaged during surgery. Reported occurrences of this complication differ, as do the length of time for healing, but rates could be as high as 30% in patients who undergo arch surgery (Ishimoto, 2002; Mulpuru, 2008)

2.3.12.2 Immediate post-operative care

The exact configuration of post-operative care will depend on the type of operation performed and the physical condition of the patient. All surgical patients would typically be admitted to an intensive care unit where their vital signs, peripheral pulses, urine output and neurological status (including lower extremity sensation and strength) can be monitored (Hiratzka, 2010).

Patients who have undergone open surgical repair for aneurysmal disease in the root, ascending or arch segments of the aorta will have had a median sternotomy incision (a vertical surgical cut made with a scalpel down the

centre of the chest, that allows surgeons access to work on the heart and aorta, see: Julian, 1957), this will need particular attention in order to avoid infection.

Cardiopulmonary bypass (CPB), where the circulation of oxygenised blood throughout the body is performed by a machine (Gravlee, 2008), is also a requirement for these procedures. The CPB technique carries with it the risk of patients suffering from post-operative fluid retention, electrolyte abnormalities, coagulopathies and hypothermia. Furthermore, patients who undergo repairs of their aorta that go beyond the root and ascending segments into the aortic arch are likely to have been subjected to a period of circulatory arrest and cerebral perfusion. When a patient is put into circulatory arrest, their body is cooled and blood flow to the brain is stopped whilst the surgery on the blood vessels that supply the brain takes place (Ziganshin, 2013; Tian, 2013). The surgical team will then continue to send oxygenated blood to the brain using a cerebral perfusion strategy. The additional cooling of the body, circulatory arrest and cerebral perfusion strategies in these cases can lead to postoperative brain injuries, so appropriate monitoring of the patient is required.

After patients are transferred from the intensive care unit onto a general ward, care is based around management of pain, helping the patient from their bed to getting around more normally, physiotherapy and monitoring their surgical wounds.

Upon discharge, patients and their families are advised on the importance of taking the appropriate medication, taking good care of their sternal wound and the need for regular outpatient follow-up. They are advised about the signs and symptoms of infection (redness, swelling or fever) and who to contact in case of any pain, unusual sensations and weakness or dizziness (Hiratzka, 2010).

2.3.12.3 Post-surgery lifestyle and healthcare

Lifestyle goals for patients who have had surgery on their proximal aorta should include:

- Maintaining an ideal body weight with regular low-impact aerobic exercise such as walking, tennis, golf or bike riding
- Eating a low-fat and low-salt diet to keep blood pressure under control
- Avoiding smoking tobacco or using stimulant drugs such as cocaine or amphetamines, as sudden increases in blood pressure could cause serious problems in the aorta (Eagle, 2002)
- Lifting heavy weights, or doing other forms of strenuous isometric exercise should be avoided for similar reasons of sudden increases in blood pressure (Hatzaras, 2007)
- More 'extreme' sports where there is a risk of trauma or stress to the chest (such as rugby, skiing or mountain biking) should be avoided (Nataf, 2006)
- Routinely taking prescribed medication

These are some generalised points to consider that are further refined on an individual basis. Patients should be able to continue to work in most occupations, but in a similar way to their lifestyle choices, jobs where heavy lifting or hard manual labour is required may trigger serious problems with a diseased or prosthetic aorta (Elefteriades, 2003). Patients should try to avoid putting themselves at risk in this way.

Patients who have aortic disease usually require active healthcare monitoring throughout the rest of their lives, regardless of what their treatment has been. This monitoring is a combination of treatment assessment, updated ideas about where the patient's care may be heading in future, and scans or imaging of the patient's aorta that can identify further aneurysmal growth (Erbel, 2014).

Clinical follow-up typically occurs more frequently in the first 12 months after surgery, with checkups at 1 month, 6 months and 12 months post-surgery

(Isselbacher, 2005). After this, if all is going well, scans and checkups may occur every 24 months or at the surgeon's discretion.

Patients who undergo aneurysmal surgery in the NHS are automatically invited to attend a post-operative cardiac rehabilitation course. The rehabilitation course is an affiliated national programme which patients can attend approximately 6 weeks after their operation. Patients are referred to their local hospital or community health centre for meetings with a specialised rehabilitation team who aim to help them achieve a healthy, active lifestyle and lower the risk of future heart problems. Many studies from international teams have found beneficial links for patients who attend these courses (Lindsay, 2003; Hedbäck, 2001; Williams, 2006).

2.4 Health-related quality of life

2.4.1 History and concept development

2.4.1.1 Quality of life

The term “quality of life” (QoL) has its origins in research conducted in the 1930's, but awareness and relevance of the term saw an increase in the aftermath of the Second World War (Pinto, 2017). It gained notable acceptance during the 1960's and 1970's as a socio-political goal proposed by policymakers for members of society to strive towards. Common usage began following a speech by the US President Lyndon B. Johnson in 1964 where he contrasted the objective of wealth acquisition with what he seemed to consider a nobler aim, an improvement in QoL:

“These goals cannot be measured by the size of our bank balance. They can only be measured in the **quality of the lives** that our people lead. [Americans] need a chance to seek knowledge and to touch beauty – to rejoice in achievement and in the closeness of family and community, and this is not an easy goal. It means insuring the beauty of our fields and our

streams and the air that we breathe. It means the education of the highest quality for every child in the land. It means making sure that machines liberate men instead of replacing them. It means reshaping and rebuilding our cities to make them safe, and make them a decent place to live. Yes, it means all of these things and more – much more”. (Johnson's Address at Rally in the Garden).

QoL as expressed in these terms involves not only health and healthcare, but acknowledges a wide-ranging and diverse mixture of influences including politics, economics, environment, housing and architecture, employment, income, social networks and recreation. In more recent decades, these socio-environmental factors have become characterised as ‘social determinants of health’ and several models have been produced to illustrate how the organisation and distribution of economic and social resources influences the quality of people’s lives. One of the most widely used models is Dahlgren and Whitehead’s ‘rainbow model’, formulated in 1991 (see Figure 2.17. This model attempts to map the relationship between the individual, their environment and health (Dahlgren & Whitehead, 1991; Raphael, 2006).

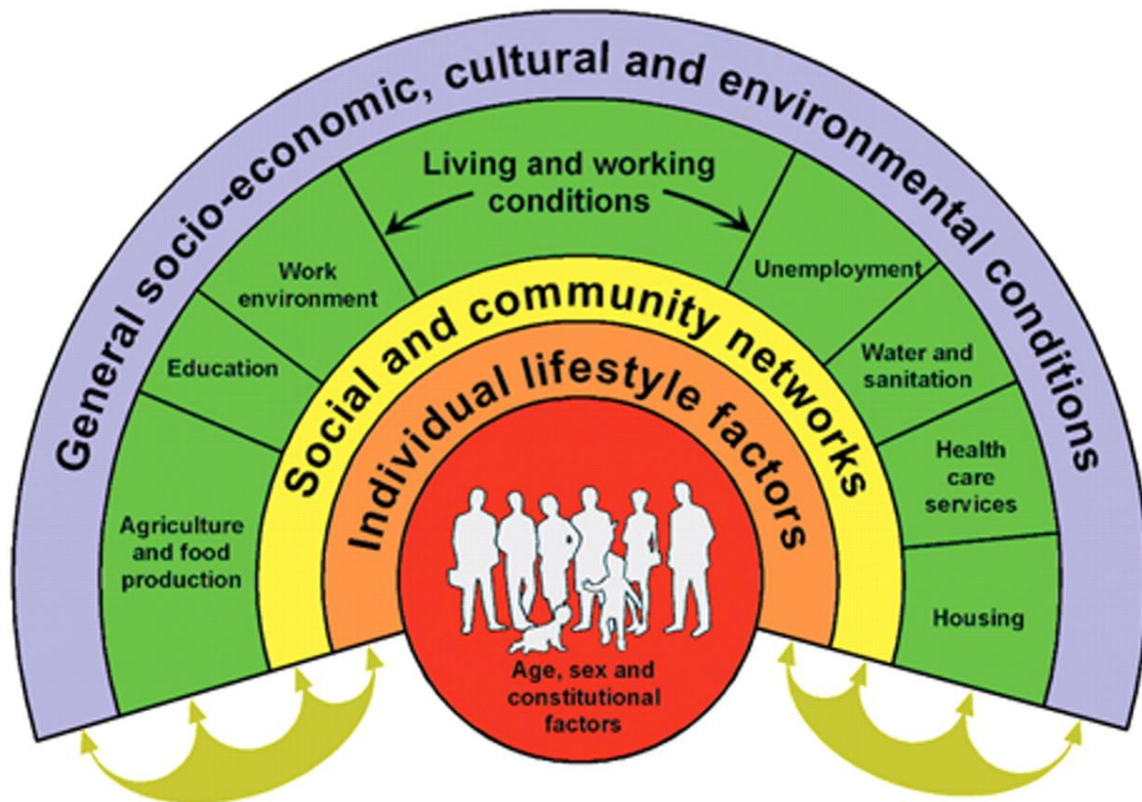


Figure 2.17: The Dahlgren-Whitehead ‘rainbow model’ of health determinants (adapted from Dahlgren and Whitehead, 1991. Image reproduced with permission from the Institute for Future Studies).

The extensive range of influences contained within QoL make it, at the very least, “a multi-level and amorphous concept” (Brown, 2004), with the result that there is no single accepted definition of QoL, even as its use has become commonplace in the last few decades throughout medical research and beyond (Barcaccia, 2013a).

In some respects, QoL suffers from its own abundance of possibilities. Scanlon (1993) suggests three interrelated questions that may contribute to the understanding of the concept:

- “What kinds of circumstances provide good conditions under which to live?”
- What makes a life a good one for the person who lives it?
- What makes a life a valuable one?”

These questions could also be asked from a number of different perspectives: the individual person, an interested or caring third party, an official in charge of allocating resources or from a larger societal/population perspective. It may therefore be understandable that a clear, shared conception of the methods and tools required to improve or sustain the value of a person's existence has not yet been reached.

Gasper (2010) argues that QoL should be understood as an umbrella term that covers many different meanings, as it "refers to an evaluation (an evaluative judgment) about selected aspects or the entirety of a life situation and that it doesn't refer to one unitary or objective entity". Although the nebulous nature of QoL definitions is in part due to these "evaluative judgment[s]" being individual and subjective, some researchers have argued that objective judgments should also be included (Meeberg, 1993; Cummins, 2005).

For example, QoL has been defined as "an overall general well-being that comprises objective descriptors and subjective evaluations of physical, material, social, and emotional well-being together with the extent of personal development and purposeful activity, all weighted by a personal set of values" (Felce, 1995).

When such a broad range of characterisations are possible, the study, application and interpretation of the concept of QoL requires careful consideration about what the term means. It has been suggested that some of the confusion around defining QoL is that researchers from different disciplines view QoL from different perspectives (Farquhar, 1995), and consequently have different purposes (Anderson, 1999). Economists focus on how scarce resources are allocated in order to gain benefits (Grabowski, 1990). Philosophers concern themselves with existential ideas and defining what may resemble a 'good life' (Ventegodt, 2003). Ethicists discuss how health-care decision-making is changing from being led by the concept of 'sanctity of life' to 'quality of life' (Weingarten 2007), and physicians appropriately concentrate on the health and illness-related dimensions (Devlin, 2017). So with no widely accepted single QoL definition to work

with, researchers should specify how they are using the concept within the parameters of their own investigations.

The individual nature of QoL means that the way in which it is measured must also be questioned. If a standardised questionnaire is used, can it truly be said to reflect an individual's outlook? Measurements must therefore be made using tools and techniques that are proven to be valid, discriminative, reliable and responsive to changes in QoL over time (Guyatt, 1993). Interpretability of results and reproducibility must also be considered (Terwee, 2007).

Another issue to appreciate when attempting to measure QoL in an individual, is how a typical questionnaire will take the fundamentally subjective concept of QoL and assemble a series of objective criteria in order to assess that individual's responses. Questionnaire tools used in this manner therefore need to be as customised as possible, and incorporate subjective experiences through the involvement of the subject group in the construction and development of the tool (Carr, 2001).

The broad, unwieldy nature of the concept of QoL suggests that realising a shared, universal understanding of what it is will remain challenging for some time (Moons, 2006). However, as QoL results are being increasingly used to inform political and economic decisions, these questions of definition, measurement, objectivity / subjectivity and shared understanding are more than just philosophical concerns. Different situations, alternative definitions and diverse points of view could have significant moral and ethical consequences (Barcaccia, 2013a).

2.4.1.2 Health status

QoL was first mentioned in relation to the medical field by Elkington in 1966. He highlighted the fact that new technologies, in particular kidney dialysis and transplantation, raised new questions for clinicians. Elkington made prescient insights that still carry weight today, he understood that medical

care was slowly changing and would begin to “focus on patients’ lives rather than patients’ bodies” (Sullivan, 2003). The three main issues he raised were broad themes drawn from personal experience, concerned with the effectiveness of medical care:

- How does a physician protect the QoL of an individual patient?
- How can QoL be improved in other patients in the future whilst also giving current patients optimal care?
- Which medical programmes should receive a greater proportion of society’s limited resources in order to achieve the best health and QoL results for all members of that society?

Traditionally, the healthcare community was focussed on outcomes based on mortality and morbidity, or post-operative changes in the biological function that had been targeted by a treatment. Many public health strategies are intended to standardise practice and guide improvement projects towards areas that can reduce the incidence and prevalence of avoidable deaths, strokes, operative infections or other negative outcomes at population level. Whilst this approach continues to be fundamental to healthcare and important to patients (SCTS, 2019), measuring QoL has seen an increase in prominence as medical treatment improves and lives are extended. Straightforward measures of mortality and morbidity were no longer seen as broad enough in scope to evaluate changes in the health of a population, ‘healthy life-years’ saw increasing prominence in public health policy and research not just ‘additional life-years’ (Bergner, 1985)

The World Health Organisation (WHO) first defined health in their constitution in 1946, and the wording remains the same as of the 48th edition published in 2014: “a state of complete physical, mental and social well-being, and not merely the absence of disease and infirmity” (Grad, 2002). The wording of this definition was highly influential for researchers as they began to develop tools that were intended to assess patient’s health. Key aspects of the WHO definition are the inclusion of psychosocial well-being and the emphasis on more than just the absence of disease signalling a

move away from the reductionist medical model towards the 'social determinants of health' (Larson, 1996).

One of the first attempts to measure and value health was the Health Status Index (HSI), devised by Fanshel and Bush in 1970. The HSI was a generic measure of health rather than a disease or population-specific measure, and the health states it defined were evaluated on a categorical scale. This scale was based on value judgements rather than in terms of an economic benefit, which was the standard practice at that time (Karimi, 2016). The eleven health states that the HSI ranged through from best to worst are as follows:

- Well-being
- Dissatisfaction
- Discomfort
- Disability-minor
- Disability-major
- Disabled
- Confined
- Confined-bedridden
- Isolated
- Coma
- Death

This early attempt concentrates on describing the physical functioning of the body. More recent tools developed for measuring general health status include the Medical Outcomes Study Short Form (SF) collection of questionnaires (Ware, 1998) and the EuroQoL 5 Dimension (EQ-5D) questionnaire (Rabin, 2001). These tools are rooted in the WHO definition of health and as such include categories which refer to the responder's psychosocial condition.

In 1984 Ware presented a “crude” framework for discussing disease and its impact upon a patient’s life (see Figure 2.18). Health status was argued to have the following elements, or domains:

- **Physiological aspects of disease**

This characteristic is at the center of the framework, because the focus of healthcare is curing or managing disease. It is comprised of the measurable physiological parameters of the disease such as symptoms, laboratory values and therapies.

- **Personal functioning**

This domain is defined as the performance or capacity to perform daily tasks, it includes self-care, mobility and other physical activities.

- **Well-being and psychological distress**

The third level of Ware’s personal health status model includes psychological effects of disease. This includes negative psychological states such as anxiety or frustration, but could also include positive changes and improved mental health.

- **General health perception**

This layer of the framework is intended to include how the individual perceives their overall health, taking into consideration the three previous health domains (physical functioning, personal functioning and psychological distress and wellbeing).

- **Social / role functioning**

The outermost layer of the model refers to an individual’s performance of their typical roles. This includes employment / schoolwork, ability to complete household tasks, or activities within the community.

Ware presented each domain in the framework as a layer. Each domain was hypothesised to have a two-way interaction with the other domains. For example, impairments in the performance of tasks (personal functioning) may result in frustration or anger (well-being / psychological distress).

Conversely, anxiety (well-being / psychological distress) as well as limiting some activities such as personal functioning, may also affect an individual's immune system (physiological aspects of disease).

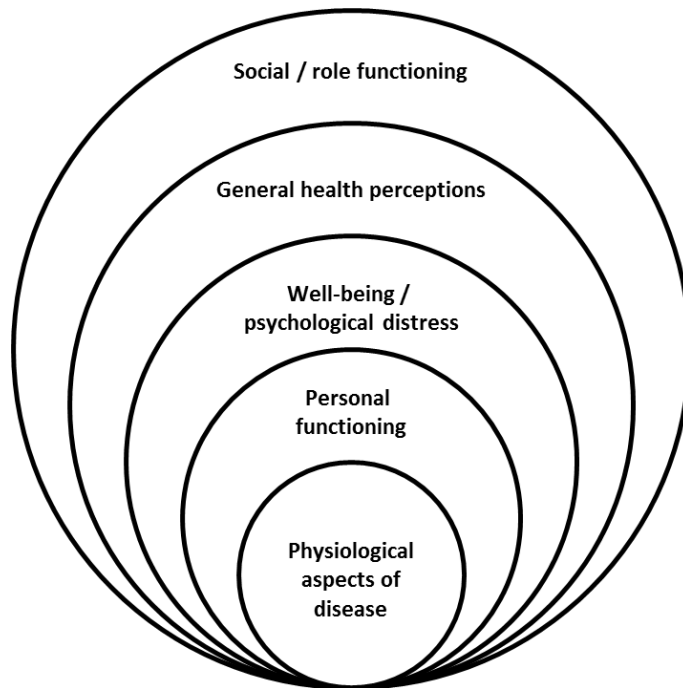


Figure 2.18: Ware's framework for discussing disease and its impact (adapted from Ware, 1984. Reproduced with permission from John Wiley and Sons).

In the same paper, Ware attempted to further categorise health status and well-being within the domains he proposed. Each framework domain was subcategorised into three "operational definitions", which would allow a healthcare professional to summarise a patient's health status whilst also incorporating QoL concepts:

- **Diagnostic indicators**
 - Blood pressure
 - Forced expiratory volume
 - Neurotic disorder
- **Physical**
 - Personal functioning
 - Role functioning

- Disability days
- **Mental**
 - General positive affect
 - Emotional ties
 - Psychological distress
- **General**
 - Self-ratings of health
 - Physical symptoms
 - Psychosomatic symptoms
- **Social**
 - Close friendship
 - Social contacts
 - Group activities

As the literature on health status measures developed and acquired increased recognition, the term health-related quality of life (HRQoL) was introduced.

2.4.1.3 Health-related quality of life

Kaplan and Bush (1982) referred to HRQoL when introducing the concept of 'quality-adjusted life years' (QALYs). QALYs are used in NHS cost-utility analyses as a measure of the value of a year in full health (Rawlins, 2004). In the wake of this, the term HRQoL was embraced by researchers in other influential papers (see for example Torrance, 1987) and expanded from there.

Lin (2013) notes that although QoL and HRQoL are often used interchangeably, the terms relate to different concepts. QoL is a broad concept that considers all aspects of human life, whereas HRQoL narrows the focus to concentrate specifically upon the effects of illnesses and the impact of healthcare treatments (Barcaccia, 2013b). This distinction is

important as it helps to separate out the elements of life that are related to how the individual maintains personal health and the wider socio-political determinants of health, such as education, home / workplace environment or religious / spiritual beliefs (Theofilou, 2012). Even though HRQoL can be seen as a subcategory of QoL it is still not an easy concept to define, Bowling (1995) asserts that “health-related quality of life is an equally nebulous concept”. This persistent difficulty in finding a straightforward definition of either QoL or HRQoL may be a consequence of the subjective nature of the terms and the wide range of determinants that contribute to them. The individualised judgments that underpin both QoL and HRQoL indicators are subjective and dynamic, meaning that defining either would rely on precisely characterising entities that are by their very nature imprecise.

Several definitions of HRQoL can be found in the literature. Torrance in 1987 related HRQoL directly to QoL: “quality of life is an all-inclusive concept incorporating all factors that impact upon an individual’s life. Health-related quality of life includes only those factors that are part of an individual’s health”. Non-health aspects of QoL, for example economic and political circumstances, are therefore not included when considering HRQoL, although the appropriateness of these omissions is often disputed (Anderson, 1999; Moons 2006)

Ebrahim’s (1995) definition similarly focuses on the aspects of QoL that are affected by health. For example, HRQoL is defined as “those aspects of self-perceived well-being that are related to or affected by the presence of disease or treatment”. This definition is sometimes stated in more focussed terms, where HRQoL “is used to identify the sub-set of the important or most common ways in which health or health care impact upon well-being” (Peasgood, 2014).

A third definition of HRQoL focuses on the value of health. For example, HRQoL can refer to the “values assigned to different health states” (Gold, 1996). These values, or utilities, are used to calculate QALYs and to measure the benefits of health technologies. The values used to calculate

the QALY are on a scale where zero is equal to dead and one is equal to full health. Values less than one are intended to reflect the loss of quality of life because of living in ill health.

One final definition of HRQoL by Hays and Reeve in 2008 used the following description: “how well a person functions in their life and his or her perceived wellbeing in physical, mental, and social domains of health”. “Functions” refers to an individual’s behaviours that can be observed by others, while “wellbeing” refers to an individual’s internal, subjective feelings and perceptions. These feelings and perceptions are not directly observable by others.

2.4.2 Quality of life vs. Health status vs. Health-related quality of life

As far back as the 1980’s there were concerns among researchers that the three terms were being used interchangeably, and with intentions that were indistinguishable from one another (Spitzer, 1987; Bergner 1989). These apprehensions around how the terms are defined and used persist in more recent publications (Moons, 2004; Karimi 2016).

The most straightforward distinction to be made is between QoL and health status. Ferrans in 1990 recognised that “quality of life is more than health status, clinical symptoms, or functional ability... health is only one dimension of quality of life”. This is shown by the range of factors influencing the definitions that were presented in Section 2.4.1.1. QoL includes many elements, such as an individual’s environmental and socio-economic context, that are not ordinarily considered to be a part of personal health. So whilst QoL is affected by health status, health status only describes a smaller subsection of QoL (Michalos, 2004). These two terms therefore describe different concepts.

Understanding and describing the differences between HRQoL and both QoL and health status is more of a challenge. This is because some HRQoL

definitions are similar to health status, and some are similar to QoL. If we consider HRQoL to concentrate on personal functioning and wellbeing, or that HRQoL is a subcategorised concept, focusing on the health properties of QoL, this does not seem to go significantly above and beyond the concept of health status. If we define HRQoL as relating to domains including physical, psychological, and social attributes then that is merely a description of 'health', as defined by the WHO. Rather than using disease symptoms or biological measurements, performance and wellbeing are used (Wilson, 1995). This makes it a determinant of health rather than QoL.

If we view HRQoL as a component of QoL that can be affected by health, it is difficult to distinguish it from overall QoL. It would be hard to identify key aspects of QoL that are not affected by health in at least some way. It is therefore sensible to approach the concept of HRQoL as an indicator of QoL derived from the perspective of healthcare; a measurement that contains both health status and QoL elements.

2.4.3 Measures of health-related quality of life

Choice of the most appropriate instrument for assessing HRQoL is dependent on the objectives for collecting data, the environment of the application, and methodological and practical considerations (Patrick, 1989). Several different types of instrument are available and a researcher should consider these and how they meet the particular requirements of the study in question (Fitzpatrick, 1998):

- **Disease-specific**

These tools have been developed to measure perceptions of a specific disease or health problem. A wide variety of instruments have been developed that are focussed on common healthcare problems. For example, the Arthritis Impact Measurement Scale is a self-administered questionnaire for use in rheumatic diseases (Meenan, 1980). It contains 45 questionnaire items covering nine dimensions:

dexterity, physical activity, mobility, household activities, activities of daily living, depression, anxiety, pain and social activities. The advantages of this approach are that the tools are more likely to have a high clinical relevance and be responsive (see Section 2.4.3.1) to any health changes that result from targeted interventions. Because the items contained within these tools are designed around diseases, patients are more likely to accept and complete them (also refer to Section 2.4.3.1) as questions are tailored and thus more relevant to their particular circumstances. This specialised approach does however have some drawbacks, as it means that aggregated health scores cannot be compared with the general population. Also, scores cannot be compared across different treatments and diseases, which may limit the application of these instruments when carrying out economic or population health evaluations. The focused nature of the instruments could also prove restrictive if, for example, any new or unforeseen side-effects appear that were not incorporated in the original tool.

- **Population-specific**

Population-specific instruments are designed to have an application to a particular demographic group, such as children or elderly people. For example, the Child Health and Illness Profile/CHIP is a tool developed for adolescents (Riley, 2004). The advantages of this approach are that population-specific instruments can be designed to have greater relevance to the group in question. For instance, in the case of the children's CHIP tool, a tailored format such as the use of cartoon illustrations rather than text can make these measures more accessible. This approach could enable individuals who are not typically consulted directly to report on their own health independently. Population-specific developed instruments may also be sensitive to systematic differences between population groups. The drawbacks of this approach are similar to the disease-specific measures, as they cannot be compared with any general population health measures

thus making the judgment of treatment efficacy across population groups difficult.

- **Dimension-specific**

Dimension-specific instruments assess one specific aspect of health status. The most common type of dimension-specific measure is one that assesses aspects of psychological well-being such as the Beck Depression Inventory (see Beck, 1988). Another commonly assessed dimension of outcome in trials of physically ill patients is pain, The McGill Pain Questionnaire is an example of a dimension-specific instrument developed for use in this way (Melzack, 1975). The instrument is based around a series of lists of adjectives that describe pain. The patient then selects the adjectives that best describe his or her personal pain status, the items chosen by patients are given quantities and summed to produce individualised scores for four separate aspects of pain experience: Sensory, Affective, Evaluative and Miscellaneous. The advantages of tools created using a dimension-specific approach are that they typically provide a more detailed assessment of a particular dimension of health than that given by disease-specific or generic instruments. A further advantage is that many of the instruments have been used for many years and in a wide range of situations, so there is a large amount of data available for comparing and interpreting results. A potential disadvantage of these types of tools is that judgments on psychological health were often made to assess differences between patients rather than being used as outcome measures. Further evidence of how appropriate and effective these instruments are for measuring changes over time is required and decisions must be made carefully to ensure they are sensitive enough to use in the context of a prospective trial (Fitzpatrick, 1998).

- **Generic**

Generic HRQoL tools are designed to measure very broad aspects of health and are potentially suitable for a wide range of patient groups

and the general population. The Medical Outcomes Study Short Form (SF-36) questionnaire (Ware, 1992) is one of the most widely used generic instruments (Brazier, 1992; Lins 2016). It is a 36-item questionnaire that measures health across eight dimensions of physical functioning, social functioning, role limitations due to physical problems, role limitations due to emotional problems, mental health, vitality, pain, and general health perceptions. Patient responses to the 36 questions are then summed to produce a 'health profile' of eight scores. The main advantage of generic tools is that they can be used in a wide range of healthcare situations. The results are also standardised, so they can be used across different patient groups in order to assess how effective different treatments or public health initiatives are. Generic tool data can be gathered from healthy populations in order to generate what may be considered 'normal' health state information, which can then be used as a comparator for different disease groups. However, the broad approach that generic tools take means sacrificing a level of detail which can limit their effectiveness in patient cohorts with specific disease problems. These tools would therefore be potentially less responsive to clinically relevant health changes.

- **Individualised**

Individualised measures are instruments in which the respondent selects domains that are of concern to them, but that are not predetermined by a list of questionnaire items (Ruta & Garratt, 1994). For example, the Measure Yourself Medical Outcomes Profile (MYMOP) is an individualised measure that allows patients to nominate and score two most important aspects of their lives (in the order of their importance) that contribute most to their overall QoL (Paterson, 1996; Ishaque, 2018). The advantages of this approach are that the concerns of the individual patient are addressed, rather than a researcher imposing a fixed idea of HRQoL that may not be applicable. This means that individualised instruments usually have a high content validity (see Section 2.4.3.1). Delivering these types of

instrument usually requires a face-to-face interview with the patient, and thus are resource intensive. This means that individualised measures are not as practical to implement as other types of instrument which involve, for example, a self-completed questionnaire.

- **Summary items**

Summary items are measures which ask respondents to summarise their HRQoL using a single question or a very small number of questions. Since 1974 the General Household Survey for England and Wales (see Thomas, 1994) has used two questions that together provide an assessment of chronic illness and disability: "Do you have any long-standing illness or disability?" and "Does this illness or disability limit your activities in any way?". The advantages of summary items are that they are as brief as possible and make the least demands on a respondents' time. This ease of item delivery means that it takes much less effort and resources to collect large comparative data samples, and regardless of how simple this type of measurement is, there is evidence that these items can be valid and reliable (Yohannes, 2011). The disadvantages of summary item measures are that the limited question scope and response variety results in crude results and limits any nuanced analysis of particular disease types, especially if only small differences are expected over time.

- **Utility measures**

The development of utility measures began in the early to mid-1990's, they grew out of economic theory with the intention of providing an estimate of individual patients' overall preferences for different health states (Drummond, 1993; Bakker, 1995). They are similar in scope to Generic HRQoL measures but they also incorporate evidence for the overall value of health states to society and can be used in cost-utility analysis. The EuroQol EQ-5D-5L (Herdman, 2011) consists of five items (5D) relating to mobility, self-care, main activity, pain/discomfort and anxiety/depression, with five levels of response (5L). On the basis

of their responses to the five items, patients are classified into a health state with a preference weight attached. Preferences for health states are derived from general population surveys using techniques such as the rating scale, standard gamble, and time trade-off. An advantage of utility measures is that they produce a single index. This helps with comparing alternative treatments for different health problems and also incorporates the idea of economic evaluation. The EuroQol EQ-5D-5L in particular is a widely used tool which has been validated in many different countries, typically included with the EQ-5D-5L is the EQ-VAS (Visual Analogue Scale) which allows respondents to self-rate their own health on a scale of 0 (worst imaginable health) to 100 (best imaginable health). Disadvantages are similar to generic measures, in that utility measures have a broad focus which make focused analysis of particular disease symptoms difficult.

The most useful distinction to make, and the one that is used most often in operational applications of HRQoL measures, is between those that are generic and hence widely applicable, and those that are specific to particular health problems or populations.

These instruments can be used in a number of applications, including clinical trials, economic evaluation and routine patient care. Different forms of instrument administration are possible, the main forms being patient or researcher completion of paper based questionnaires. Choice of HRQoL measure should be based on a number of criteria including certain psychometric properties (see Section 2.4.3.1), but also more general issues such as the appropriateness of an instrument for a specific application or patient cohort.

2.4.3.1 Attributes of health-related quality of life instruments

Following the identification of literature pertaining to instruments it is important that users apply the necessary criteria to select the most suitable instrument(s). As Fitzpatrick documented in 1998, there are eight criteria that should be considered in the selection of HRQoL tools: appropriateness, acceptability, feasibility, interpretability, precision, reliability, validity and responsiveness each attribute is explained in more detail below.

- **Appropriateness** – *Is the HRQoL tool appropriate to the questions which the study is aiming to address?*

A researcher must consider the nature of their patient study group, which domains of HRQoL are important and if the instruments available fit into those parameters (Guyatt, 1991). These considerations are often unique to each individual study, making objective recommendations about how to select a tool difficult, but researchers certainly need to consider how instrument items, scales and content will be received by responders (Ware, 1987). Deciding on the balance between specific and generic HRQoL instruments within the tool is also important. Where possible, both specific and generic instruments should be used to measure HRQoL (Cox, 1992; Devlin, 2010). In this way the most immediate effects of treatment on disease should be captured, as well as possible consequences that are harder to anticipate.

- **Acceptability** – *Is the instrument acceptable to patients?*

Indicators of acceptability include how long the tool takes to complete, what the response rates are and how complete the data is (Fitzpatrick, 1998). There are a number of factors that can influence acceptability including how the tool is administered, questionnaire design, and what the health of respondents is like at the time of completion. What type

of HRQoL measure is used can also influence how acceptable it is. For example, respondents completing individualised instruments usually find it more difficult than completing a pre-determined questionnaire (Ruta, 1999). General features of layout, appearance, and legibility are considered to be important influences on how acceptable a tool is. Language is also an important consideration, the instrument must be presented in a language that is familiar to respondents (Herdman, 1997). Acceptability issues should be considered at the study design stage, incorporating patients' views with pre-testing of the instruments (Sprangers, 1993).

- **Feasibility** – *Is the instrument easy to administer and process?*

Feasibility means considering how easy the instrument is to administer and process. These are important considerations for staff and researchers who collect and process the information produced by HRQoL measures (Erickson, 1995). Instruments that are difficult to administer and process may jeopardise the conduct of research and disrupt clinical care. An obvious example of this is the additional resources required for patient interviews versus a patient self-completing a questionnaire. The complexity and length of an instrument will also have implications for data collection and analysis.

- **Interpretability** – *How interpretable are the scores of the instrument?*

Interpretability is concerned with how meaningful the scores produced by an instrument are. There are three common approaches to interpretation in the literature. First, changes in instrument scores have been compared to previous scores produced by the same instrument (Testa, 1996). Secondly, attempts have been made to identify a minimal clinically important difference (MCID), which is presented as being the smallest change in score that is perceived as beneficial by patients (see for example Jones, 2005). Thirdly, normalised data from the general population can be used to compare and interpret scores (Garratt, 1994).

- **Precision** – *How precise are the scores of the instrument?*

Item scales within instruments have important implications for precision. A binary 'yes' or 'no' is the simplest form of response, but it does not allow respondents to report degrees of difficulty or severity. The majority of instruments use Likert type scales such as: strongly agree, agree, uncertain, disagree, strongly disagree (Sullivan, 2013). Visual analogue scales appear to offer greater precision but there is insufficient evidence to support this and they may be less acceptable to respondents (Fitzpatrick, 1998). The ability to capture the full range of HRQoL may vary in different instruments (Gardiner, 1993). Item Response Theory (IRT) may be applied to further determine the precision of an instrument. IRT assumes that a measurement construct, such as physical disability, can be represented by a hierarchy that ranges from the minimum to maximum level of disability (Lord, 2012). IRT has shown that a number of instruments have items concentrated around the middle of the hierarchy with relatively fewer items positioned at the extremes (Garratt, 2003). This approach allows researchers to understand both the intricacies of a patients' HRQoL and how precise the measurements used are.

- **Reliability** – *Does the instrument produce results that are reproducible and internally consistent?*

Reliability can be seen as the proportion of a HRQoL score that is signal rather than noise, or an assessment of how confident researchers can be in what the results of the instrument are telling them. As the measurement error of an instrument increases, so does the sample size required to obtain precise estimates of the effects of an intervention (Fitzpatrick, 1998).

Reproducibility assesses whether an instrument produces the same results on repeated administrations when respondents attributes (e.g. symptoms) have not changed. This is assessed by test-retest reliability (Weir, 2005). There is no exact agreement about the length

of time between administrations but in practice it tends to be between 2 and 14 days (Streiner, 2015). The reliability coefficient is normally calculated by correlating instrument scores for the two administrations. It is recommended that the intra-class correlation coefficient be used in preference to Pearson's correlation coefficient, which fails to take sufficient account of systematic error. Reliability correlations of above 0.7 and 0.9 are recommended for instruments that are to be used in groups and individual patients respectively (Fitzpatrick, 1998).

Internal consistency is measured with a single administration of a tool and it assesses how well items within a scale measure a single underlying dimension. Internal consistency is usually evaluated using Cronbach's alpha, which measures the overall correlation between items within a scale (Tavakol, 2011).

- **Validity** – *Does the instrument measure what it claims to measure?*

Validity can be assessed qualitatively through an examination of instrument content, and quantitatively through factor analysis and comparisons with related variables. As with reliability, validity should not be seen as a fixed property and must be assessed in relation to the specific population and measurement objectives (Hays, 2005). Qualitative evidence can be obtained from considering how the instrument was developed. This includes the extent of involvement of experts with relevant clinical knowledge in instrument development (Guyatt, 1994). More importantly, consideration should be given to the extent of patient involvement (Andersen, 2009).

Quantitative validity testing typically takes the form of construct validation. Construct validity is assessed by comparing the scores that the instrument produces and evaluating how these scores align with hypotheses related to the measure. Many instruments are multidimensional and measure several domains, such as physical functioning, mental health, and social functioning. These domains

should be considered when assessing construct validity. Factor analysis and principal component analysis can provide empirical support for the dimensionality or internal construct validity of an instrument (Joliffe, 1992). These statistical techniques can be used to identify separate health domains within an instrument (Garratt, 2001).

- **Responsiveness** – *Does the instrument detect changes over time that matter to patients?*

Responsiveness is usually assessed by examining changes in instrument scores for groups of patients whose health is known to have changed. Alternatively, patients may be asked how their current health compares to some previous point in time by means of a health transition question. There is no single agreed method of assessing responsiveness and a number of statistical techniques are used for quantifying responsiveness (Langfitt, 2006).

2.5 Patient reported outcome measures

2.5.1 Patient-centredness

The concept of a patient-centred approach to medical care has been discussed in the literature since 1969, when Balint began to explore the possibilities of “understanding the patient as a unique human being”. The idea found popularity throughout the 1970’s as healthcare treatment saw a shift from concentrating mainly on the biology of patients (a ‘traditional’ diagnosis, to use Balint’s terminology (1970)) to combining biology, psychology and social perspectives (an ‘overall’ diagnosis (Balint, 1970; Bensing, 2000)).

In 1976, Byrne and Long published a method that sought to categorise a medical consultation as either doctor- or patient-centred. They analysed 1850 general practitioner (GP) patient visits and concluded that the majority of doctors had a biological, or traditional, style of interpretation. That meant

that they responded to a patient's condition only through their own frame of reference. By the late 1980's, Stewart and colleagues were developing a 'patient-centred clinical method', with the aim of encouraging more effective patient / doctor communication and ultimately improving health outcomes (Stewart, 1989; 1995; 2003).

Over recent decades, patient-centredness in medicine has found an increasing number of advocates. Many practical and theoretical advances have arisen from studies on patient / doctor communication, such as the influential text by Ley in 1988 which found that improved communication led to an increase in patient satisfaction and compliance. Mead and Bower (2000) attempted to define the concept, and proposed five key dimensions that explained how patient-centredness differed from the 'traditional' biological model.

- **Biopsychosocial perspective**

This dimension is a central theme of many papers concerned with patient-centredness. It refers to the idea of extending the understanding of patient illness to include social and psychological factors. Stewart (2003), for example, maintains that a patient-centred consultation needs a clinician who has a "willingness to become involved in the full range of difficulties patients bring to their doctors, and not just their biomedical problems". According to Grol (1990), a patient-centred doctor is one who "feels responsible for non-medical aspects of problems". So the concept of patient-centredness can be seen as the widening of the scope of medicine from merely an organic disease to a far wider range of "dysfunctional states" (Silverman, 1987).

- **The 'patient-as-person'**

This dimension is concerned with understanding the personal meaning of the illness for that particular individual. For example, a

broken arm may not be experienced the same way by two separate individuals, and it is recognised that the medical treatment can be experienced differently too (Kaba, 2007). Mead and Bower (2000) suggested that this can have many dimensions, one obvious contributory factor being the economic instability that a period of poor health may produce. In order to develop a complete understanding of a patients' situation, and to provide effective clinical management, a doctor "should strive to understand the patient as a distinctive personality within his or her unique context." (Kaba, 2007).

- **Sharing power and responsibility**

Rather than focus on the paternalistic mode of healthcare that was encouraged in the 1950's (Parsons, 1991), Mead and Bower (2000) suggested a more democratic, equal partnership between doctor and patient. This represented a definite shift from a doctor guiding and a patient co-operating towards mutual participation in diagnosis and treatment. In this way, power and responsibility are shared. Byrne and Long (1976) recommend "encouraging the patient to voice ideas, listening, reflecting, and offering collaboration".

- **The therapeutic alliance**

Following on from the idea of partnership between doctor and patient, the alliance that is built is regarded as having its own intrinsic value. Treatment adherence may be increased if there is a mutual understanding and concordance based on friendly courteous conduct and a sympathetic manner (Wahl, 2005; Martin, 2000). Conversely, a negative relationship may produce misunderstandings or errors in judgment. A common understanding of the goals and requirements of treatment is crucial to any therapy, whether physical or psychological (Mead and Bower, 2000).

- **The ‘doctor-as-person’**

The final dimension considers the contribution that the doctor as an individual makes to the doctor / patient relationship. Balint (1993) described the traditional biological model of medical assessment as “one person medicine” meaning that the doctor is effectively undetectable when a patient’s clinical situation is described. In contrast to that approach, patient-centred medicine is portrayed as “two-person medicine”, positioning the doctor who is involved as an integral component of the healthcare encounter: “the doctor and patient are influencing each other all the time and cannot be considered separately”. Sensitivity and insight into the reactions of both parties can be used for therapeutic purposes (Mead and Bower, 2000).

In defining the conceptual framework of patient-centredness (see Figure 2.19 below), Mead and Bower (2000) also hypothesised a number of variables which have the potential to influence the degree of patient-centredness that a doctor may exhibit. At the centre of the model is the doctor-patient relationship. This relationship is expressed in the form of how the two parties behave towards one another, these behaviours are closely related to the five key dimensions discussed above.

The ‘Shapers’ are the external factors that shape the interaction, such as cultural norms or clinical experience, and may have an impact upon more specific elements such as gender or ethnicity. For example, cultural norms relating to gender may mean that it is more socially acceptable for females to discuss feelings and emotions than males (Chaplin, 2015).

The specific professional context of the clinical practice may also have an impact on patient-centredness. For example, GP’s may have overall knowledge of a disease, but could lack the specific understanding of an unusual problem which prevents them from having personal confidence in their diagnosis.

Finally, Mead and Bower (2000) point out that consultation-level influences may have the most immediate impact on the propensity of doctors to be patient centred. For example, time or workload pressures may limit possibilities for a satisfactory discussion potentially meaning that a treatment resolution between the doctor and patient is not possible. The conceptual framework presented also explicitly recognises that a doctor's tendency to be patient-centred will vary over time, and that the personal dimensions (i.e. the patient-as-person and the doctor-as-person) can require a significant period of time to develop.

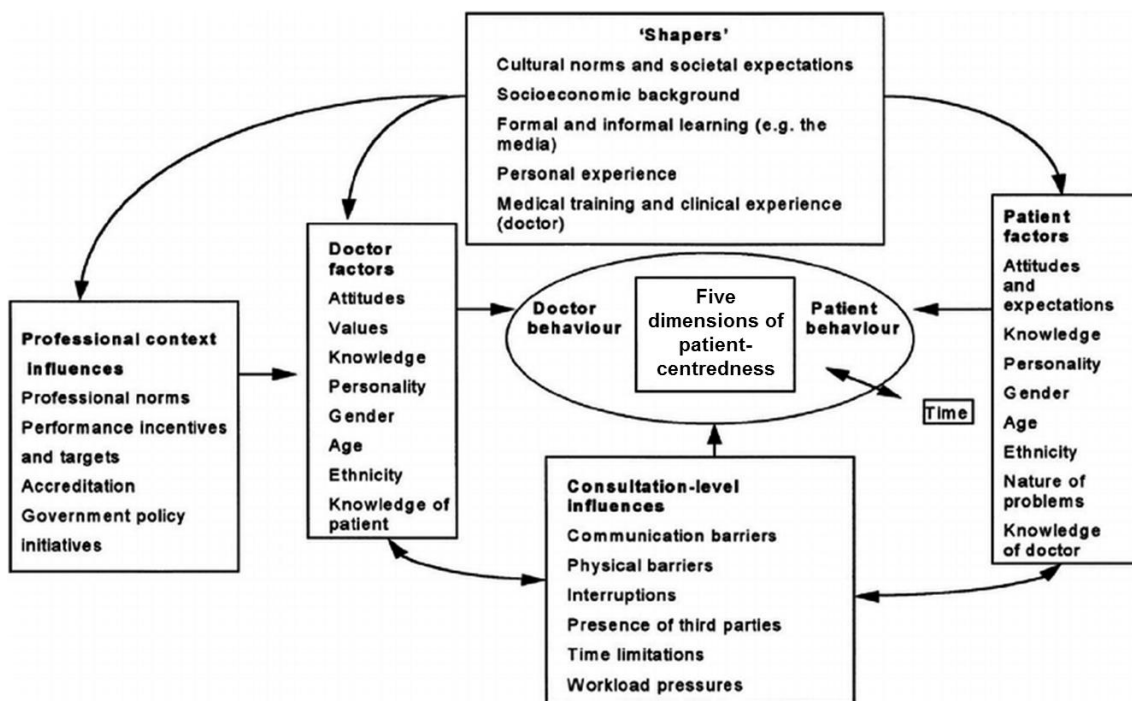


Figure 2.19: Factors influencing patient-centredness (adapted from Mead and Bower, 2000. Reprinted from *Social Science & Medicine*, Copyright (2000), with permission from Elsevier).

2.5.1.1 Communication with healthcare professionals

Communication skills are fundamental to the idea of a patient-centred mode of healthcare, at the turn of the century a consensus statement was released by 21 medical communication leaders with the objective of identifying the

'essential elements of communication in medical encounters' (Makoul, 2001). They concluded that there were seven sets of fundamental communication tasks that should apply during healthcare consultations:

- Build the doctor-patient relationship
- Open the discussion
- Gather information
- Understand the patient's perspective
- Share information
- Reach agreement on problems and plans
- Provide closure

A more recent consensus guideline for oncological treatment produced more detailed recommendations for doctors who care for adults with cancer (Gilligan, 2018). The first key point of 'Core communication skills' represents a contemporary view of how a skilled doctor may demonstrate the fundamental ideas of patient-centred care:

"1. Core communication skills

1.1. Clinicians should review the patient's medical information, establish goals and anticipate the needs and responses of the patient and family.

1.2. Clinicians should explore the patient's understanding of their disease and set a collaborative agenda with the patient and their family

1.3. Clinicians should use behaviours that actively foster trust, confidence and collaboration

1.4. Clinicians should provide information that is timely and useful to the patient. Clinicians should also check that the patient understands this information.

1.5. When patients display emotion, clinicians should respond empathically.”

Other key communication themes include ‘Discussing goals of care and prognosis’, ‘Discussing treatment options and clinical trials’, ‘Using communication to facilitate family involvement in care’ and ‘Communicating effectively when there are barriers to communication’.

2.5.1.2 Improving patient satisfaction and outcomes

There is compelling evidence that a patient-centred approach to healthcare can improve both patient satisfaction and outcomes. Little (2001) found that when GP’s adopted a positive, patient-centred approach (defined with five components: ‘Communication and partnership’, ‘Personal relationship’, ‘Health promotion’, ‘Positive and clear approach to problem’ and ‘Interest in effect on life’) that it was significantly associated with patient satisfaction, patient enablement (how capable the patient felt in terms of dealing with their health problem), a reduction in referral rates and a reduction in symptom burden 1 month after their appointment.

A systematic review of 40 patient-centred care articles (Rathert, 2013) reported similarly encouraging results. They reported strong evidence for patient-centredness increasing patient satisfaction and having a positive impact on patient self-management and adherence to medical instructions. However, the impact on clinical and long-term outcomes was more mixed, with some studies reporting improvements and some finding no difference. It was noted that observing the most important outcomes was a challenging task and that future studies would need to be carefully designed to fill that gap in knowledge (Rathert, 2013; Donabedian, 1988).

2.5.1.3 Patient-centredness in the United Kingdom

In the UK, policy documents released by the NHS have also increasingly highlighted the importance of a patient-centred approach to clinical management. Beginning in 2004 with an NHS Improvement Plan subtitled 'Putting People at the Heart of Public Services' (Department of Health, 2004), these policy white papers recognised that: "A new spirit of innovation has emerged, centred on improving the personal experience of patients as individuals, and this is now taking root in the NHS."

Subsequent policy documents reinforced this approach, all stating the intention of putting patients and their families at the centre of the NHS healthcare approach. The language used within these documents and the clear intention was to ensure that the design, delivery and evaluation of services become responsive to the needs and priorities of NHS users. In 2005, another NHS policy white paper titled 'Creating a Patient Led NHS' acknowledged that patient-centredness was not merely a small adjustment to current practice, but "a fundamental change in our relationships with patients and the public... [T]o move from a service that does things *to* and *for* its patients to one which is patient-led, where the service works *with* patients to support them with their health needs."

This shift in focus, along with a continued commitment to present "An NHS that gives patients and the public more information and choice, works in partnership and has quality of care at its heart." (Department of Health, 2008a), positioned PROMs as a fundamental means of assessing effectiveness of care from the patient's perspective: "This means understanding success rates from different treatments for different conditions. Assessing this will include clinical measures such as mortality or survival rates and measures of clinical improvement. Just as important is...the patient's own perspective which will be measured through patient-reported outcomes measures (PROMs)" (Department of Health, 2008b)

2.5.2 What are patient reported outcome measures?

PROMs are health-related questionnaires which aim to determine patients' views of their health symptoms, their functional status and their HRQoL. This is achieved by patients completing a PROM both before and after their treatment. (It is important to note that PROMs are not the same as PREMs (Patient Reported Experience Measures), PREMs focus on aspects of the humanity of care, such as being treated with dignity or being kept waiting (LaVela, 2014)).

Nelson (2015) identified five types of healthcare structure or situation that may benefit from the use of PROMs, along with stating what those benefits are:

- **Type 1:** A health system
 - Deliver patient-centred aspects of performance assessment
 - Determine value for money
- **Type 2:** Healthcare providers
 - Benchmarking PROM performance between providers (in the NHS, this could indicate a utility for commissioners)
 - Can be used as a gateway to quality improvement
- **Type 3:** Clinical trials
 - Screening. Identifying patients who may be at a higher risk of suffering post-operative complications
 - Can be used as evidence for differences in treatment outcome
- **Type 4:** Clinical practice
 - Have the potential to assist medical diagnosis
 - Can be used to monitor patient progress post-treatment
- **Type 5:** Information for patients or clinicians
 - Have the potential to influence choice of provider
 - Have the potential to influence choice of treatment

The routine use of PROMs is in keeping with the national promotion of patient-centredness. PROMs are a vital component in signifying the shift away from traditional biological health evaluations towards the biopsychosocial model (see Section 2.5.1). Black (2013) notes that the aim of most healthcare is to reduce symptoms, minimise disability and improve QoL, and that these are aspects which can be assessed only by patients. Patients often welcome being involved (Cox, 2007) (but not always, see Levinson 2005), and when patients do experience strong involvement, this has been shown to have benefits not just to patient health but also to healthcare institutions (Delaney, 2018). Considering patients' views also increases public accountability of health services and clinicians (Black, 2013).

2.5.3 Patient reported outcome measures in practice

Following on from the NHS policy reconfigurations detailed in Section 2.5.1.3, the first nationwide use of PROMs took place in 2008. This took the form of a voluntary audit of mastectomy and breast reconstruction patients (Jeevan, 2014). After the delivery of this programme had been established as feasible, a UK programme for PROMs in four elective surgeries was established. From April 2009 to October 2017 it was mandatory for all NHS providers who treat patients undergoing hip or knee replacement, groin hernia repair or varicose vein surgery to invite patients to complete a PROM questionnaire before and after their surgery. After a review of the national PROM programme took place during 2016, collection of groin hernia repair and varicose vein PROMs ceased in October 2017 while the collection of PROMs for hip or knee replacements is ongoing. The invitation to participate in PROM research typically happens in the pre-assessment clinic or on the day of admission. Black (2013) describes the content and process of PROM delivery:

“The preoperative questionnaire collects data on the patient's sociodemographic characteristics, the duration of their

condition, their general health, any comorbidities, and whether they are undergoing a repeat/revision procedure. In addition, they are asked to complete a disease specific PROM...and a generic PROM...Patients who complete a preoperative questionnaire are mailed a postoperative questionnaire...Non-responders receive one reminder letter. The questionnaire includes the same PROMs as the preoperative one plus single transitional items on their overall view of the result of surgery and the extent of any improvement. They are also asked to report on adverse outcomes (complications, readmission, and further surgery).”

Initial response rates to this initiative were encouraging, with around 131 250 eligible recruitments out of 245 220 eligible patients (54%) over the second year of delivery. These rates did vary from around 63% recruitment in the hip and knee replacement patients to around 38% for varicose vein surgery (Hutchings, 2014).

For these nationally mandated PROM returns, returned patient data is linked to data collected from Hospital Episode Statistics (see Herbert, 2017). This link allows a match between the PROM data and a systematically reported NHS admissions dataset. The link permits a more wide-ranging analysis upon a set of demographic, diagnostic and procedural information. The NHS providers of the elective surgery in question are identified and the change in PROM results is adjusted for case mix (Nuttall, 2015). In this way different healthcare providers can be compared to each other, and assessments can be made at a national level to identify any outlying institutions.

2.5.3.1 National PROM pilot for revascularisation

Between November 2011 and January 2013 in 11 English hospitals specialising in cardiac care, a pilot study was performed to assess the feasibility of delivering PROM instruments to patients who had undergone

coronary revascularisation. The patients may have undergone either cardiac surgery or a percutaneous coronary intervention (PCI). The main PROM instrument used for collecting pre- and post-procedure HRQoL data was the Coronary Revascularisation Outcome Questionnaire (CROQ; Schroter, 2004), a psychometrically validated patient based measure which as well as novel items contains items borrowed and modified from the Seattle Angina Questionnaire (Spertus, 1995), the Quality of Life after Acute Myocardial Infarction questionnaire (Valenti, 1996), the Angina Impact Questionnaire (Wilson, 1991), the SF-36 (Ware, 1992), the Menorrhagia Outcomes Questionnaire (Lamping, 1998a) and the Prostate Outcomes Questionnaire (Lamping, 1998b).

Official documentation regarding the outcomes of this pilot remain unpublished, but preliminary conclusions were that patient response rates were comparable to the nationally mandated PROM programmes at 61%, although there was considerable variability between hospitals the highest being 87% and the lowest 41% (unpublished National Cardiac Benchmarking Collaborative data). Early recommendations included the need for staff to have specific time allocated in order to administrate the PROM.

2.5.4 Experience of using patient reported outcome measures

In addition to the UK PROM approach, literature has been published describing experiences of PROM programmes from many international groups including Australia, Canada, Sweden, the Netherlands and the United States (Meehan, 2006; Cranley, 2004; Kettis-Lindblad, 2007; Haverman, 2011 and Crandall, 2010).

The perceived value of PROMs data varies in different settings and specialities, but in broad terms healthcare professionals do find value in PROMs, particularly in situations where they are useful for supplementing the clinical decision making process (Boyce, 2014). More specifically, studies

which reported positive aspects of PROMs found that they improved clinician's ability to assess the severity of patient symptoms (Crane, 2007; Berry, 2011), informed treatment decisions (Ayers, 2015) and enabled tracking of both treatment outcomes and general health and wellbeing over time (Forsberg, 2015; Katzan, 2011).

However, implementing PROM collection as a routine part of a healthcare pathway is not without challenges. Logistical concerns related to the extra workload placed on the staff administering the questionnaires and analysing and interpreting the subsequent data are often expressed (Miller, 2015; Franklin, 2015). Also, as technology continues to advance with service users beginning to expect quick, easy access to important healthcare information over the internet or on their mobile devices, PROM delivery needs to develop to meet these raised expectations (Lavalley, 2016). This prompts questions around the most informative and appropriate way to deliver PROM results (Kroenke, 2015) and the best way to tackle any legal or regulatory requirements in collecting and storing PROM data (Petersen, 2015).

2.5.5 Health-related quality of life and patient reported outcome measures in aortic surgery

A recent review of QoL papers focussed on patients undergoing surgery on their thoracic aorta (Jarral, 2015) found thirty relevant studies in the literature, however only twelve of these were focussed on the proximal aorta, and they included patients who had aortic dissections as well as elective aneurysm presentations.

Of the twelve which concentrated on the proximal aortic segments, seven included patients who underwent different forms of isolated aortic root replacement (Akhyari, 2009; El-Hamamsy, 2010; Franke, 2010; Golczyk, 2010; Khaladj, 2009; Lehr, 2011; Perrotta, 2010). These typically demonstrated a follow-up HRQoL that was comparable to healthy members of the general population. El-Hamamsy (2010) found an improvement in

physical functioning for patients who received a Ross procedure rather than a homograft, while Franke (2010) reported a superior HRQoL in all domains for patients who received a David procedure rather than a composite root replacement.

Four more studies reported outcomes after a mixture of different proximal aortic operations and again reported an acceptable HRQoL following surgery. Lohse (2009) concentrated on aneurysms within the ascending aorta, including concomitant aortic valve replacements, David, Bentall and Cabrol procedures, while Oda (2004) was interested in how more elderly patients (>65 years) coped with the impact of aortic surgery. Song (2012) examined patients with Marfan's syndrome and reported the differences in HRQoL between elective aneurysmal surgery and emergency type A dissections, the study found emergency surgery to be a significant predictor for impaired long-term HRQoL and reoperations. Another study by Stalder (2007), including both aneurysmal and dissection patients, identified the use of deep hypothermic circulatory arrest as a predictor of an impaired long-term deficit in physical functioning.

Two further studies described HRQoL outcomes after the surgical repair of type A dissections. Campbell-Lloyd (2010) reported a reasonable long-term survival rate and a HRQoL that was similar to that of healthy individuals in the general population, while Nakamura (2011) reviewed a small number of patients with cerebral malperfusion and demonstrated an initial improvement in functional status after surgery along with further improvement at follow-up.

2.6 Summary

The purpose of this review was to define and describe the pathology of proximal TAA and related diseases, to explore the concept of HRQoL and to examine the role of PROM instruments in how they record and report patients' experiences of treatment. The further intention of this chapter was to determine whether there was an existing, disease-specific, patient-based

questionnaire that is appropriate for measuring HRQoL and health status in patients undergoing surgical treatment for proximal TAA disease.

While there are useful instruments that have been used in similar clinical settings, and general health tools that have been used in similar patient populations, there was no single PROM instrument found that was appropriate for the measurement of HRQoL in a proximal TAA population. Subsequent chapters therefore describe the development and pilot of a new instrument designed to measure patient-based outcomes in these patients. Where possible, items from existing questionnaires reviewed in this chapter were considered for inclusion.

Chapter 3

Methodology

3.1 Introduction

The following chapter explores the philosophical and methodological framework for the development of a PROM tool aimed at patients who undergo elective surgical treatment for a proximal aortic aneurysm. The previous chapter highlighted the limited research that has been undertaken so far in this subject area. Considering the cost of performing major aortic surgery (Mishra, 2008), and the lack of evidence to date for the effectiveness of this surgery in terms of HRQoL and psychosocial recovery, this was considered an important area for further investigation.

3.2 Content of this chapter

Briefly, the main sections of this chapter will be:

- A summary of PROM methodology and an outline of the current research study problem
- An exploration of the pragmatic philosophy and theory that will underpin the research methodology
- An outline of the aims of the research study
- A rationale for gathering both quantitative and qualitative data, with associated research questions, descriptions of the types of data collected and the associated analytical approaches
- A discussion and definition of mixed methods research, including a categorisation and description of the type of mixed methods design used
- A research map

3.3 PROM methodology

The United States Food and Drug Administration (US FDA) centre have provided detailed guidance for PROM development (US FDA, 2006). They identify four main phases that should typically occur during instrument development, illustrated in Figure 3.1:

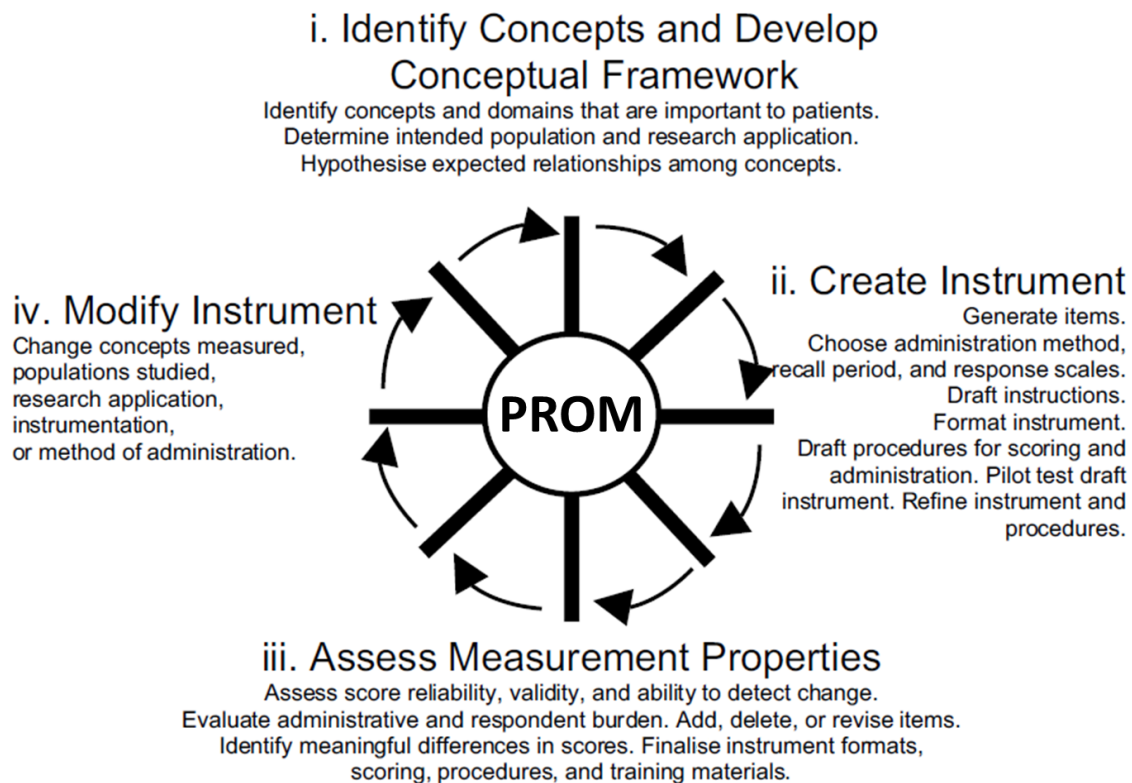


Figure 3.1: The PROM instrument development and modification process (adapted from the US FDA guidelines document, 2006)

3.3.1 Identifying concepts and developing a conceptual framework

An appropriate, clearly defined conceptual framework is a ‘fundamental consideration’ in PROM development (US FDA, 2006). Concepts and domains are generally chosen based mainly on patient interviews, along with expert opinion and literature review (see Rothman 2007 and Grady 2015 for

commentary and examples). It is also important at this early stage to clarify how the PROM is going to be applied; some PROM instruments are used to assess outcomes in clinical trials (Vodicka, 2015), while others evaluate treatment benefit (Schäfer, 2010; Nilsdotter, 2003) or monitor adverse events (Banerjee, 2013; Absolom, 2017). The intended patient population also needs to be identified, with particular considerations made towards the impact of the targeted disease and demographics such as age, sex, ethnicity and cognitive ability (Ju, 2017; McKenna, 2011).

3.3.2 Creating the PROM instrument

Items included in the PROM instrument can be generated from analysing interview transcripts (with patients, medical experts, family members or other stakeholders), reviewing the literature or focus group discussions (US FDA, 2006). Item generation should always be undertaken with the involvement of appropriate patients (see Marcovitch 2017 and Kingsley 2017). The way in which the PROM data is to be collected also requires clarification. Any special considerations around instrument administration procedures (such as interviewer instructions, instructions for self-administration or electronic / web-based / IT system requirements) need to be finalised at this stage. Malhotra (2016) reports a successful experience of using electronically delivered PROM instruments, and Hewlett (2016) highlights another secondary issue concerning the translation of PROM items into different languages.

The delivery strategy must also be reflected upon and tailored correctly. For example, if a patient should not expect to recover from their treatment until six months afterwards, sending a PROM instrument at two months would be incongruous. Similarly, if it is decided that more than one post-operative PROM is required then this must be balanced against a likely reduction in response rates (see Wood (2016) for a more detailed discussion on PROM response rates).

Instrument formatting, accompanying letter structure and decisions on item visualisation should also occur during this phase. Study resources should be assessed in order to ensure PROM delivery, data entry and data storage elements have the necessary capability. Scoring algorithms need to be finalised and the working version of the PROM instrument should be confirmed (US FDA, 2006).

3.3.3 Assessing the measurement properties

Lohr (2002) provides a detailed summary of the attributes and criteria that a PROM instrument should possess in order for it to be considered useful. PROM reliability, validity, responsiveness (or the ability to detect change) and interpretability (including a minimally important clinical difference) all need to be assessed and evaluated to determine its appropriateness for delivery. These attributes have already been discussed in some detail in Section 2.4.3.1.

For further insight into assessing PROM reliability and validity see Bolarinwa (2015), who provided a convenient review of the principles and approaches to assessing health research questionnaires, aimed particularly at researchers in developing countries. Responsiveness and minimally important change (MIC) are the focus of many papers in the literature including Christiansen et al (2015), who compare the performance of two independently designed shoulder outcome PROMs which aim to evaluate both pain and joint function. The paper used Receiver Operating Characteristic (ROC) curves to measure the ability of the scores to identify significant health improvements. ROC analysis was also used to identify the MIC for each PROM instrument. Chiarotto et al (2016) used a similar approach to assess how three different versions of the Pain Self-Efficacy Questionnaire (PSEQ) performed with regard to people suffering from chronic lower back pain. The original 10 item questionnaire and two newer, shorter versions with four items (PSEQ-4) and two items (PSEQ-2) were included in the comparison study. Ohanyan et al (2017) provided a short

report on the responsiveness and MIC characteristics of a recently developed PROM for people with the chronic urticaria skin condition called the Urticaria Control Test (UCT; Weller, 2014). The UCT results were compared with results from other established PROM instruments focussed on dermatological outcomes; the Urticaria Activity Score (UAS), the Chronic Urticaria Quality of Life Questionnaire (CU-Q2oL) and the Dermatology Life Quality Index (DLQI). These comparisons allowed the study coordinators to assess responsiveness and estimate the MIC for their UCT PROM instrument.

Devji et al (2017) meanwhile, presented a strategy for enhancing the interpretability of PROMs when carrying out a meta-analysis. The motivation for this study is the differences in the way that PROM results which use the same instrument, or PROM results with a common disease focus using different instruments, can be reported. These differences may lead to difficulties with interpretation and aggregation when carrying out meta-analyses. The paper discussed the strengths and limitations of various different methods of PROM result reporting, including mean differences, standardised mean differences, relative risks, odds ratios and MIC units. The ultimate aim of the research strategy is to objectively assess how PROM results are reported in meta-analyses, to provide recommendations for future PROM design methodologies and suggest a standardised approach to reporting PROM results.

3.3.4 Modification of the instrument

When a PROM instrument is used in a new patient population, item wording or appearance are changed, or PROM measurements are revised, this is characterised as a 'modification' (US FDA, 2006). Modified PROM instruments are considered to be different from the original, and properties are held to be version-specific. Additional validation is recommended to ensure the PROM instrument performs to an acceptable level following modification.

3.3.5 Applying a methodology to the current research question

The above representation of the current approach to PROM instrument construction show that undertaking such a project requires both qualitative (interviews, observations, focus groups) and quantitative (questionnaire validation using factor analysis, intraclass correlation coefficients, scoring algorithms, identifying minimally important clinical differences) research methods. Neither approach, if used in isolation, would successfully tackle the problem. This indicates that a mixed methods approach (Johnson, 2004) would be the most viable methodology for PROMs researchers to employ (for examples, see Bravo (2015), Dür (2015) and Martin (2018)), a research methodology that arises from the pragmatic school of philosophy and explained by Biesta (2010).

3.4 Pragmatic philosophy

The obvious challenge of placing PROM construction within a single research philosophy is that they should reflect individual patient experiences (which would be characterised as interpretivism (see Myers, 2008)), but they also generate quantitative data summarising the instrument properties and ultimately a numeric score that is representative of a pre-defined HRQoL domain (which would be characterised as positivism (see Crossan, 2003)). As a consequence of this methodological duality, PROMs research studies are perhaps best considered as being in the pragmatic tradition (Neale, 2015).

The foundations of pragmatism as a philosophy date back to publications by Charles Sanders Peirce in the late 19th and early 20th century (Pierce 1878, 1905). These ideas were subsequently developed by William James (1907) and John Dewey (Dewey developed his theories in a series of publications over the course of more than two decades, from 1917 to 1938) who in particular proposed a method of thinking called 'inquiry-based learning', where research questions are the primary driver of knowledge creation and

the production of knowledge flows from those beginnings. In fact, Dewey preferred to avoid the assumptions associated with the word “knowledge” and coined the term “warranted assertions” instead. Dewey maintained that inquiry produces outcomes, which produces warrants and that the knowing cannot be separated from the doing.

“For Dewey, the knower and the known were inseparable, bound together in a process of inquiry, with a simultaneous reliance on both belief and action... At the broadest level, Dewey’s pragmatism as a philosophy addresses the central question: What is the nature of human experience? Refocusing on inquiry as a central form of human experience requires reconsidering the philosophy of knowledge by replacing the older emphasis on ontology [how reality is perceived] and epistemology [how we know what we know] with a concentration on inquiries about the nature of human experience.” (Morgan, 2014)

This early thinking has been shaped for contemporary applications by Maxcy (2003), Hoshmand (2003) and Johnson (2004), among others. Unlike purely interpretivist or positivist research philosophies, which broadly conform to either subjective or objective ontologies, a pragmatic research philosophy accepts any relevant concept which can be used effectively to answer the research question at hand and effect a positive change. Pragmatics “recognise that there are many different ways of interpreting the world and undertaking research, that no single point of view can ever give the entire picture and that there may be multiple realities” (Saunders, 2012). Pragmatics also derive the meaning of ideas from their practical application. The essence of a pragmatic ontology could therefore be said to rely on actions and change (Goldkuhl, 2012).

Pragmatists feel that their approach values common sense and experience, and rather than adopting an epistemology that views knowledge as a “copy” of reality, they seek progress through empirical inquiry and prioritising the utility of outcomes (Legg, 2019).

Kroenke (2015) applies this strand of philosophical thinking to the creation of PROM instruments. He notes that the early advocate of pragmatism William James defined “truth” as that “which works” or that which motivates action to affect a positive change (James, 1967). The action “which works” in the context of a PROM instrument is therefore how it may allow an understanding of and improvement to patient care. In the 1980’s, another pragmatic philosopher stated that “It is the vocabulary of practice rather than of theory, of action rather than contemplation, in which one can say something useful about truth.” (Rorty, 1982). Kroenke goes on to draw a comparison between this perception of practical ‘truth’ and the reality of PROM implementation.

3.5 Aims and research questions of the current study

3.5.1 Aims

This study aims to develop a conceptual model that identifies key domains of HRQoL for individuals who have a thoracic aortic aneurysm. These domains will form the basis of a disease specific item set, which will be used to formulate relevant HRQoL questions. These questions will then be used in conjunction with preference based generic HRQoL measures (e.g. EQ-5D, SF-6D) to form a PROM instrument for patients who undergo surgery for a proximal aortic aneurysm.

3.5.2 Research questions

1. Is there an opportunity to develop a PROM instrument for patients undergoing surgical treatment for proximal TAA disease?

This first question has been responded to in part by the in-depth literature review presented in Chapter 2, where the decision to develop a proximal TAA PROM was justified by establishing the requirement for, and lack of, a disease-specific measure in this area.

2. What is the lived experience of patients who undergo aortic surgery on the proximal aorta?

This second question will be answered by the qualitative study phase, where patient interviews will be conducted, transcribed and analysed for key themes. The output will be a conceptual model of HRQoL in proximal TAA patients based on the data.

3. What themes specific to patients suffering aneurysmal disease of the aorta reflect a patient's health status / HRQoL, and how can these be relayed in a PROM?

The third question forms the bridging section between the qualitative and quantitative study phases, where the PROM will be constructed based on the emerging qualitative themes. The output will be a draft PROM instrument that can be piloted in a population of proximal TAA patients.

4. Is the administration of a PROM feasible in this patient population?

The fourth question forms the subsequent section of the quantitative study phase, where the PROM will be piloted and patient acceptance will be assessed via criteria such as response rate, administrative burden and data completeness.

5. Does the newly constructed PROM instrument perform well enough to be useful to future patients and clinicians?

The fifth question forms the final section of the quantitative study phase, where the PROM pilot results will be tested for reliability, validity and responsiveness. Recommendations on future administration and further instrument testing will be made.

3.6 Study rationale

The impetus for undertaking this project was the lack of a specifically tailored PROM instrument for the proximal aortic aneurysm surgery population. In

order to maximise the impact and utility of the PROM it was decided that a close application of the FDA guidelines (see Section 3.3), including gathering and analysing both qualitative and quantitative data, would be necessary. There is a standard methodological reasoning behind this approach, as Creswell (2013) explains it was required because we “need better contextualised instruments, measures, or interventions to reach certain populations”.

This established methodology of PROM instrument development (US FDA, 2006) requires both qualitative and quantitative strategies to succeed. As described above, this method is based within the pragmatic philosophical paradigm, and will use a mixed methods approach to research.

3.7 Mixed methods research

Johnson and Onwuegbuzie (2004) define mixed methods research as “the class of research where the researcher mixes or combines quantitative and qualitative research techniques, methods, approaches, concepts or language into a single study”. It is hypothesised that where a study has both interpretivist and positivist elements to it, that a mixed approach can provide a more complete, blended understanding of the research question than either a qualitative or quantitative analysis in isolation could do (Creswell, 2003).

The concept of a mixed methods research approach emerged from what commentators described as the “paradigm wars” (see Howe, 2003). During this period of the 1980’s and early 1990’s, members of the positivist / quantitative research community argued that only their approaches could lead to robust knowledge and an understanding of objective “truth”, they also maintained that qualitative methods lacked rigorous scientific precision. Conversely, interpretative/qualitative researchers believed that results based solely on statistical analysis and quantitative measurement gave a blinkered, inadequate interpretation of events. The human element was ignored and the

role of motives, reason, background knowledge and cultural behaviours, among other things, were side-lined (Siegel, 2018).

Rather than become entrenched in these philosophical battles, some researchers began to embrace a mixed methods approach (Tashakkori, 1998). This meant that instead of being at odds with one another, the objective and subjective poles of research could be melded together to create broad, balanced, complementary results.

This mixture of approaches should not, however, be adopted in an arbitrary, careless way. Proper consideration should be taken to make the correct methodological choices and integrate the appropriate results for the research question at hand (Bryman, 2006; Denscombe, 2008). It is also worth noting that the mixture of paradigms displayed in mixed methods research has the effect of diminishing the philosophical emphasis on specific ontological and epistemological perspectives. Of greater importance is achieving a consensus and producing outcomes which have practical value, as advocated by the underpinning pragmatic philosophy (see Section 3.4).

There are both benefits and challenges to consider when applying a mixed methodology approach to research. Johnson (2004, p21) provides a concise table of mixed methods strengths and weaknesses. Some accepted benefits include:

- Mixing qualitative and quantitative data can allow a greater understanding of the research problem and yield more complete evidence, in theory the investigator will gain both depth and breadth. “It makes intuitive sense to gather information from different sources, utilising different methods, which work together as an efficient design” (Almalki, 2016).
- Combining both numerical and thematic data can help avoid over-reliance on the former, and allows the researcher to acquire “soft-core views and experiences” (Jogulu, 2011), or the subjective factors necessary to help explain complex social interactions.

- The process of triangulation (Jick, 1979; Wilson, 2014), where the final results of the study may include both observations and statistical analyses, provides additional evidence and support for the findings.
- The use of a mixed methods approach can also help researchers to develop their skills. This is particularly important for those at an early stage of their career (Molina-Azorín, 2016).

Along with these advantages, selecting a mixed methods approach also presents challenges:

- It is more time-consuming and resource intensive to collect both quantitative and qualitative data.
- The research procedures can be complex and may be beyond the experience of the research team. Investigators are often trained in quantitative or qualitative methods and may need assistance to achieve good results in the alternative discipline (Brannen, 2005).
- Methodological intent will require clear presentation when published or presented, so that the audience can accurately understand the procedures and the findings.

3.7.1 Mixed methods research type

Several different outlines for a mixed methods study exist in the literature. Four of the most frequently used designs are:

- Triangulation mixed methods design

This type of study takes the form of a single phase project, with simultaneous collection of qualitative and quantitative data. The results are then combined with the intention of merging the two separate strands

of data into a composite model outcome. Some examples of this type of study are found in Graham (2005) and Casey (2009).

- Embedded mixed methods design

This type of study is conceived as providing supporting or secondary outcome data within the context of a larger interventional research study. The intention is to produce an output which supports the findings of the larger study, thereby enhancing the impact. Some examples of this approach to mixed methods design are found in Houtz (1995) and Rogers (2003).

- Explanatory Sequential mixed methods design

As the name suggests, the explanatory sequential design features sequential data collection in a two phase project. Here, the quantitative phase comes first, followed by the qualitative phase, with the concept that data collected during the second phase will build upon what was uncovered in the first. The intention is to better explain the phase one results, or to enable purposeful participant selection to better understand the initial findings. Some examples of this design approach are found in Lalor (2013) and McCrudden (2018).

- Exploratory Sequential mixed methods design

Again, this type of design features sequential data collection in a two phase project. But in this design the timeline of the approaches swaps round, so the qualitative phase comes first, followed by the quantitative phase. The concept of this approach is also that the data collected during the second phase builds upon the results of the first. The intention is to apply qualitative data to develop an instrument, or to identify categories that can be applied to a quantitative measurement. Some examples of this design approach are found in Stoller (2009) and Berman (2017).

The current study will employ methodological integration by gathering initial qualitative data, analysing it, and then using the qualitative results to build a new PROM instrument that will be tested quantitatively. The mixed method

type that seemed most appropriate for aim of study was therefore the exploratory sequential approach. Figure 3.2 shows the fundamental structure of the study plan.

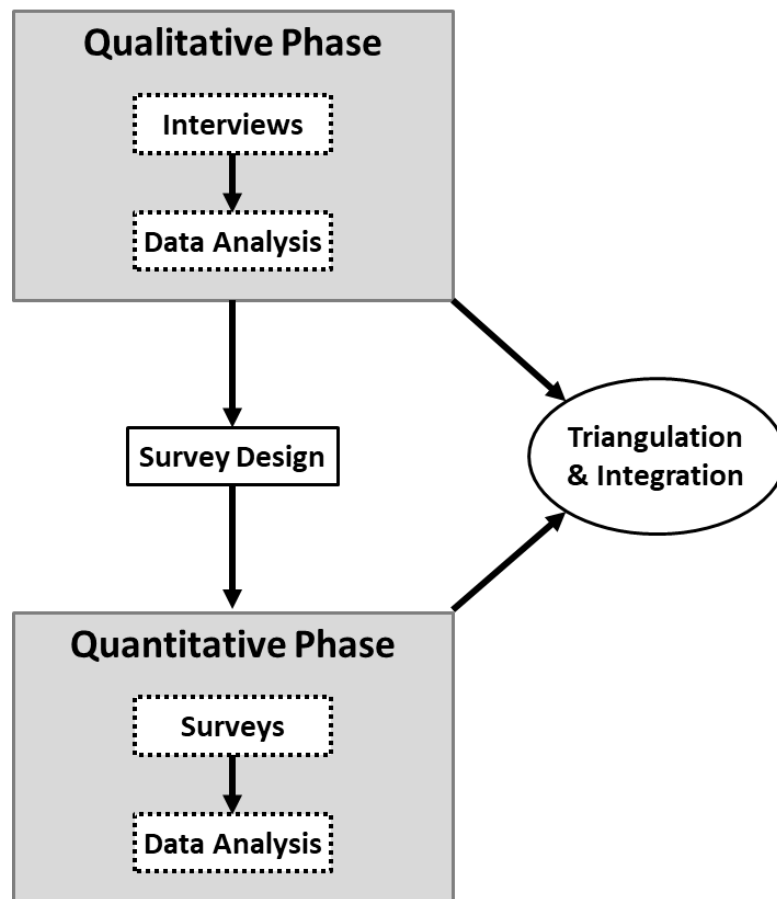


Figure 3.2: Exploratory sequential mixed methods design (adapted from Wu, 2012)

While Figure 3.3 illustrates in greater detail the two phases of implementation.

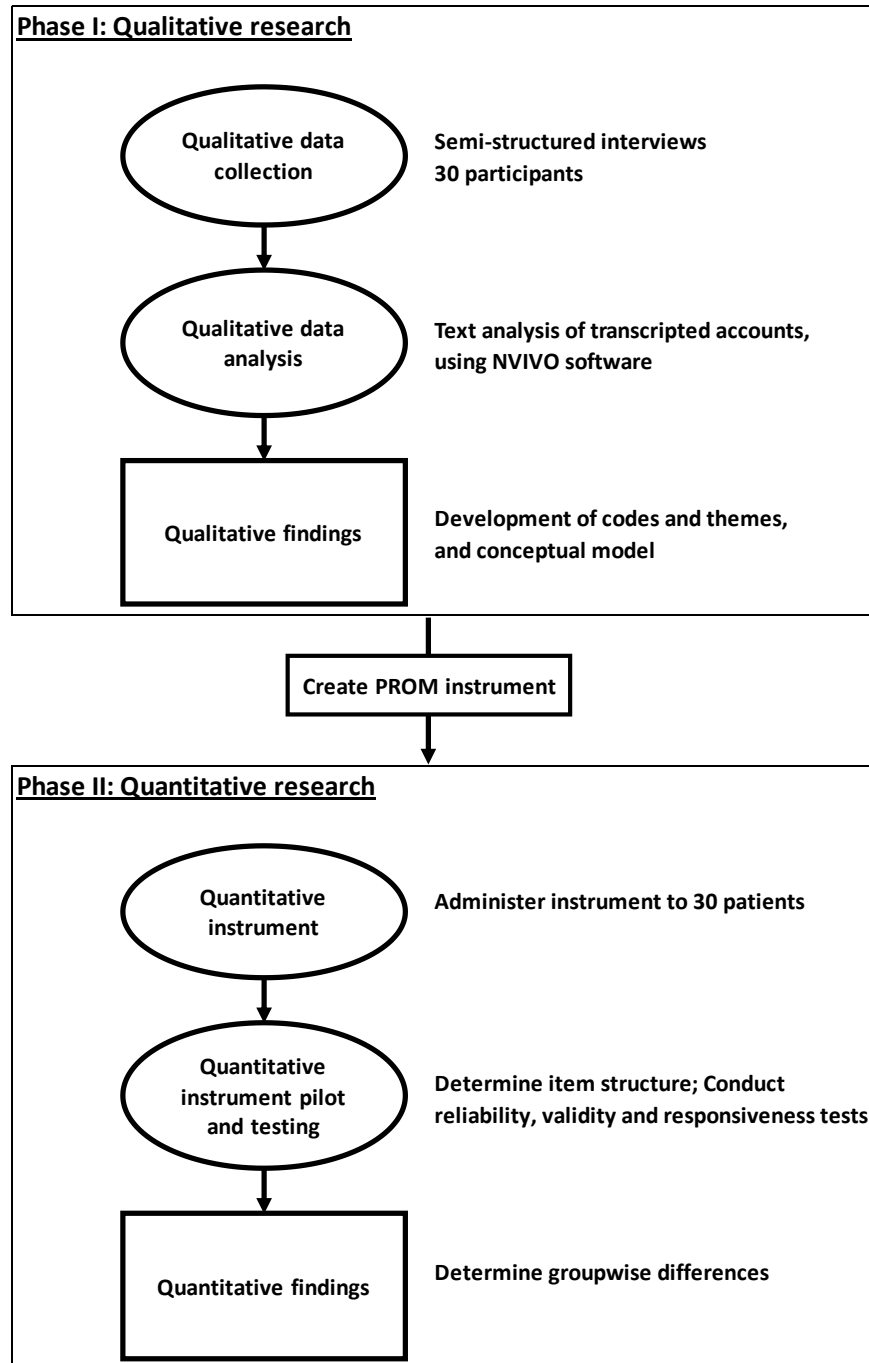


Figure 3.3: Exploratory sequential mixed methods design for the current study

3.8 Research study flow diagram

Figure 3.4 shows how the two phases of the exploratory sequential mixed methods design fit into the research study outline as a whole.

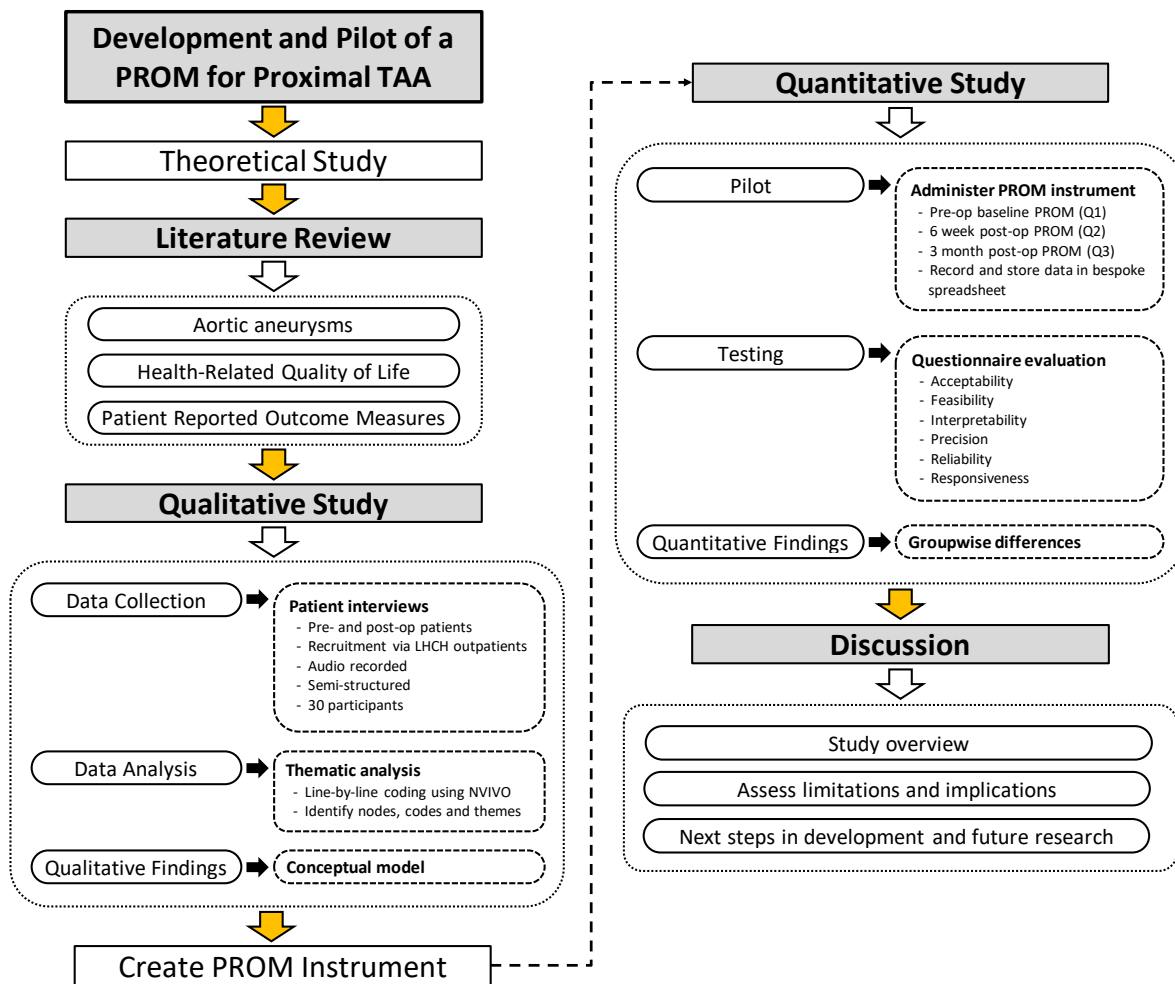


Figure 3.4: Research flow diagram for the current study

Chapter 4

Qualitative study

4.1 Introduction

This chapter presents the results of the qualitative study phase. This phase aimed to explore patient's experiences and perceptions related to proximal TAA disease, which included experiences of living with the disease and their perceptions of the healthcare they received. In-depth, semi-structured interviewing and thematic analysis were used with the intention of developing our understanding of the patients lived experience.

4.1.1 Appraisal of the current literature

Entering "aortic aneurysm" and "interview" as search terms into the PubMed US National Library of Medicine, National Institute of Health webpage (<https://www.ncbi.nlm.nih.gov/pubmed>) returned 46 entries (date of search, 10/05/2019). 10 (22%) of the published studies were only tangentially related to aortic aneurysms and did not include patient interviews. 24 (52%) studies made specific reference to abdominal aneurysms in the study title, while 8 (17%) made reference to endovascular repair, which also indicated abdominal aneurysm treatment. The remaining four studies (Luehr, 2017; Gavazzi, 2016; Melby, 2013 and Niclauss, 2011) did include patients who had treatment on their proximal aorta, but two were focussed on dissection patients only (Melby, 2013 and Niclauss, 2011). The third included coronary artery bypass grafting patients and valve replacements as well as ascending aorta repairs (Gavazzi, 2016) and the final paper (Luehr, 2017) included patients who had aortic arch replacements only, with any type of pathology. All four of the studies that included proximal aortic patients used interviews in order to establish long-term treatment complications, rather than enquire about patients' experiences. To date, no studies were found that explored the

lived experience of these patients, identifying a gap in the literature and making the current work an appropriate addition to current knowledge.

4.2 Objectives

The objectives of this study were:

1. To explore the experiences of patients with proximal aortic aneurysm disease in order to obtain an in-depth understanding of the extent and nature of how that disease impacted on their health status / HRQoL
2. To develop a conceptual model for health status / HRQoL in aortic aneurysm patients based on their personal experiences
3. To develop a questionnaire aimed at addressing health status / HRQoL in aortic aneurysm patients

4.3 Methods

4.3.1 Ethical approval

Ethical approval of the study was granted by the National Research Ethics Service Committee in Solihull on 16th February 2015 (Ref: 13/WM/0456). The confirmatory letter can be found in Appendix A.

4.3.2 Inclusion and exclusion criteria

The inclusion criteria for the qualitative study were:

- Participants must be aged 18 years or over
- Presenting specifically for previous or current proximal thoracic aortic aneurysm (TAA) disease
- Outpatient attendee at LHCH

The exclusion criteria were:

- Under 18 years of age
- Unable to provide informed consent
- Unable to read or write in English

4.3.3 Recruitment

Purposive sampling (see Robinson, 2014) was used to recruit patients attending routine outpatient appointments at Liverpool Heart and Chest Hospital (LHCH), a tertiary centre in the North West of England that treats, amongst other disciplines, patients with cardiac and aortic diseases. Recruitment was made using the following protocol:

- Suitable patients with a scheduled upcoming outpatient attendance were identified from the hospital Patient Administration System (PAS).
- An introductory letter was sent inviting the patient to take part in a research study (Appendix B), this included information about the purpose of the research, details on the interview subjects and structure and an incentive for participation in the form of payment for travelling expenses (up to a maximum of £50).
- Enclosed with the introductory letter was a Patient Information Sheet (Appendix C) which gave further details on the research study, how patient confidentiality would be managed and what the patient should do if they needed more information or wished to make a complaint. Patients were informed that participation was voluntary, would not affect their care and that they could withdraw from the study at any time, before or after the interview had taken place.
- A telephone call was then made to the eligible patients, to ask whether they would be willing to participate in the study. If the patient was

happy to be interviewed, they were met in the outpatient department and then interviews took place after their scheduled outpatient appointment.

- Participants were given the option of participating in the interview alone or having another person present.

4.3.4 Study participants and sample size

All participants provided written informed consent (Nijhawan, 2013) before their participation in the study, in accordance with Health Research Authority protocols (Appendix D). They were also offered a reimbursement of their travelling expenses (up to a maximum value of £50); many international studies have confirmed that monetary incentives can be useful in increasing the willingness of potential interviewees to participate in a study of this nature (Edwards, 2005; Singer, 2013; Kelly, 2017).

The patient sample size for the initial qualitative interviews was relatively large ($n = 30$) in order to ensure maximum variability. In order to support the patient findings, four specialist aortic surgeons (all based at LHCH) were also interviewed during the same period.

In qualitative research, unlike in quantitative data collection, it is not feasible to objectively predict optimal sample size prior to data collection. A frequently used concept for determining the sample size of nonprobabilistic studies is “data saturation”, which indicates the point at which no new information, ideas or themes are emerging from the data provided by the study participants (Guest, 2006). The goal was to acquire a dataset that was both “rich” (multi-layered, detailed and intricate) and “thick” (lots of data), in order to facilitate in-depth and high quality analyses (Burmeister & Aitken, 2012; Fusch & Ness, 2015)

The method chosen for analyses of the qualitative data was Thematic Analysis (see Section 4.3.6). Researchers using this method have suggested

that data saturation is not a useful method for determining an appropriate sample size, and some authors claim that this term may have little practical meaning (Malterud, 2016 in Braun, 2019). However, this has not discouraged some investigators from advising that between 6 and 12 interviews should be enough to achieve saturation in these types of studies (Guest, 2006; Ando, 2014). Hennick (2017) goes further, making a distinction between “code saturation” (range of issues identified) and “meaning saturation” (data richness and textured understanding achieved), suggesting that 9 interviews are enough to achieve the former and between 16 and 24 interviews for the latter. Another attempt by academics to employ a statistical methodology aimed at objectively estimating sample size for studies using thematic analysis (Fugard and Potts, 2015) received a largely sceptical response (Emmel, 2015; Braun and Clarke, 2016). In addition to these considerations, the researchers involved did not have a vast amount of qualitative study experience to draw upon. Sample size and data collection was therefore predicated on the following:

- A review of how many interviews had taken place in similar studies
- A pragmatic assessment of the number of interviews that would be possible within the time frame
- A continuing awareness throughout the interview process of the richness of the information collected and the diversity of the subject population, attempting along the way to broadly recognise how these elements would ultimately be applied to the research objectives

A short review of similar studies helped to support decisions around sample size for the qualitative phase. McElhone (2007) developed and validated a disease specific HRQoL instrument for adult patients with systemic lupus erythematosus (the LupusQoL), thematic analysis of 30 patient interviews took place at the qualitative stage. Welk (2013) used thematic analysis on 16 patient interviews to conceptualise and develop a PROM for neurogenic bladder dysfunction. An international study which used thematic analysis to

develop a conceptual HRQoL model for Hepatitis C (Armstrong, 2016) recruited 70 patients in total for interview (30 from France, 20 from Brazil and 20 from Australia). Forty six patients (38 from England, 8 from France) were interviewed in a study which aimed to define patient experiences within emergency care, with the intention of subsequently creating a PROM instrument for use in that clinical environment (Vaillancourt, 2017). Leffler (2017) proposed a conceptual model for the impact of coeliac disease on HRQoL amongst adults, also with the intention that the findings be used to construct a PROM; 21 patients were interviewed, with thematic analysis used to interpret the qualitative data. Tatlock (2017) interviewed 30 patients for a similar study concerning patients suffering with gout.

Planning to interview 30 patients therefore seemed to be a reasonable goal based on these comparable publications, and an achievable aim with regard to the study timetable and the availability of suitable patients.

4.3.5 Interviews

Two interview guides were developed (one for pre-operative patients and one for post-operative patients), these were based on the expertise of the primary investigator and clinical experts, and a review of the literature (Appendices E1 and E2). The interview guides were dynamic and flexible and were continually reviewed by the interview team (primary investigator, clinical supervisor and interview assistants) as the study progressed. Initial interview questions were general, to obtain patient perspectives, and then were divided into broad chronological domains, from diagnosis through to treatment and post-operative care, with specific probes within each domain.

The majority of the semi-structured interviews were conducted by the primary investigator (n = 17), and three other research team members, with experience of qualitative research with respect to patient experience, also assisted. The interviews lasted between 20 and 75 minutes. Interviews were audio recorded on a digital Dictaphone and transcribed verbatim using a denaturalised approach. The denaturalisation of interview data has been

suggested as being a preferable technique for thematic analysis studies (Neale, 2016). The interview guide was designed to probe the patient's thoughts and feelings about their experience of TAA's in a chronological way.

For the clinician interviews, each surgeon was asked open-ended questions about their perspectives on caring for TAA patients. These questions followed a similar order to the patient interviews as shown in the interview guide in Appendix E1 and E2.

4.3.6 Data collection

Digital recordings of the interviews were made using a Dictaphone. After the interview was completed, the digital files were uploaded onto the secretarial dictation system at LHCH in readiness for transcription. A hospital secretary familiar with the clinical terminology and experienced in typing up dictated recordings was employed to transcribe the interviews.

4.3.7 Data entry and thematic analysis

Interview transcripts were imported into NVivo software (QSR International Pty Ltd. Version 10, 2012) for qualitative data analysis.

Thematic analysis (TA) of the transcripts took place based on the six phase process that Braun and Clarke introduced in 2006 and have continued to develop over the following years (Braun and Clarke, 2013; Braun, Clarke and Rance, 2015). Although their method was initially conceived within the discipline of psychology, it has been used widely across many areas and has been specifically identified as offering a robust, practical solution for healthcare researchers who are undertaking qualitative data analysis (Braun and Clarke, 2014).

The six phase Braun and Clarke TA structure grew out of content analysis (see Mayring (2004) and Hsieh (2005)), and was heavily influenced by principles established by Boyatzis in 1998. Boyatzis was interested in how

qualitative data could be arranged into “codes” and “themes” so that pertinent sections of text in relation to the research question could be highlighted, collected and reported with clarity. His approach to and innovations with TA have been characterised as “a bridge between the language of qualitative research and the language of quantitative research” (Boyatzis, 1998), making the techniques apt for a mixed methods study design. Braun and Clarke then refined and popularised the TA approach to qualitative analysis in their landmark 2006 paper. Many well-cited publications covering a variety of specialities have used these techniques in the ensuing years, including studies relating to international entrepreneurship (Jones, 2011), gerontology (Wiles, 2012) and environmental psychology (Devine-Wright, 2010).

Bearing in mind that the investigators were relative novices in regard to qualitative research, it is also worth noting that TA is recommended as a suitable introductory method for this type of study “as it provides core skills that will be useful for conducting many other forms of qualitative analysis” (Braun and Clarke, 2006; Maguire, 2017). This argument is substantiated by Holloway (2003), who recognises the principle transferable skill of “thematizing meanings” as applicable to many other qualitative research methods, such as phenomenology, ethnography or grounded theory.

Briefly, the six phases of TA and how they were applied to the current study are as follows:

4.3.7.1 Phase 1: Data familiarisation

Familiarisation involves the initial immersion of the researcher in the data (Rabiee, 2004; Lacey, 2007). It is the process of listening to recordings, and then reading and re-reading the interview transcripts, and then making informal summaries about the information that has been gathered. This stage of immersion and insight was particularly important in the current study as the interviews were not all performed by the same researcher.

4.3.7.2 Phase 2: Initial code generation

After data familiarisation, preliminary code generation began. At this point in the analysis, transcripts were imported into the NVivo software package and systematically scrutinised. This stage allows for a broad interpretation of the data (Terry, 2017), any data segments that were considered relevant to the research question were tagged and given a short, meaningful title. Tuckett (2005) describes the process of code assignment as being “contingent on asking of the data segment, ‘What is being described (event, action, interaction) in the data text’? and ‘How is it understood (processes) – what does it mean’? ‘Why?’”. This interrogative thought process was used throughout the preliminary coding phase.

4.3.7.3 Phase 3: Identifying themes

After a comprehensive scrutiny of the interview data in phases one and two, the process of constructing a thematic sequence began. This stage involved a detailed inspection of the identified codes and the detection of any shared patterns, which then allows more extensive themes to be identified and categorised (Terry, 2017). Themes were generated using analytic induction and constant comparison within and between transcripts. This can be as straightforward as recognising a single complex code which incorporates several other codes within its meaning, these codes can then be “promoted” into themes (Charmaz, 2000). More frequently, the identification of codes and the relationships between them are knotty and complicated, requiring careful consideration and revision as they grouped together and given thematic status. The researcher must identify a dominant impression, “a core concept or idea, a central organising concept” (Braun, 2019), that is shared across a range of codes.

4.3.7.4 Phase 4: Reviewing themes

In this phase, the content of the identified themes was appraised and evaluated for coherence. Each code comprising a theme was examined and if they were found to be logical and consistent they were accepted. If a theme category or coding were found to be problematic then they were reworked. For example diet and alcohol began as separate sub-themes but upon reflection were found not to have enough variance to justify that choice, so they were merged into one. The results at the end of this phase were determined to accurately represent in a wider sense how the researcher viewed the data set as a whole (Braun and Clarke, 2006).

4.3.7.5 Phase 5: Defining themes

After thematic categories were identified, there was a requirement to provide a stronger sense of definition and meaning. The initial categorisation of themes focussed on broadly summarising a profusion of rich data, and then this step refined that approach into achieving a thematic narrative that sought to explain the in-depth meaning contained within the themes. “Clarity, cohesion, precision and quality” (Terry, 2017) were sought, with the aim of sharpening the thematic map produced in the previous phase. However, researchers must take care to ensure that the refined themes bring a more developed meaning to the overall data rather than oversimplifying and risk losing substance.

4.3.7.6 Phase 6: Producing a report

After the thematic analysis and categorisation was complete, the production of the report (in this case the thesis) summarised the analysis done up to this point. If it is possible to answer the original research questions using the assembled thematic structure with both illustrative and analytical precision, and to construct a compelling narrative that honours the complete data set,

then the researchers can take some reassurance from this. Braun (2019) also advises that adjustments to the thematic constructions and definitions can still be made at this stage, in order to make the final product as clear, consistent and representative as possible.

4.4 Results

4.4.1 Participant characteristics

28 post-operative and 2 pre-operative TAA patients (21 males, 9 females; mean age 61 years (standard deviation = 12), age range 35 to 84) were interviewed between March and August 2015. In the 28 post-operative patients, time since their operation ranged from 2 months to 10.5 years. Patient characteristics are shown in Table 4.1, where continuous variables are shown as median (inter-quartile range) and categorical variables are shown as absolute number (%).

Table 4.1: Qualitative study, patient characteristics

Characteristic	TAA patients (n = 30)
Age at interview	62 (53, 68)
Female gender	9 (30.0)
Ethnicity	
White - British	30 (100)
Marital status	
Married	22 (73.3)
Divorced / separated / widowed	5 (16.7)
Single	3 (10.0)
Index of multiple deprivation quintile	
1 (most deprived)	5 (16.7)
2	5 (16.7)
3	5 (16.7)
4	8 (26.7)
5 (least deprived)	7 (23.3)
Pre-operative, 'watch and wait' patients	2 (6.7)
	Post-operative patients n = 28
Body-mass index (kg/m ²)	27.1 (25.0, 31.3)
Canadian Cardiovascular Society (CCS) angina classification	
Asymptomatic	23 (82.1)
I	1 (3.6)
II	4 (14.3)
New York Heart Association (NYHA) functional classification	
I	10 (35.7)
II	15 (53.8)
III	3 (10.7)
Previous myocardial infarction	1 (3.6)
Diabetes	2 (7.1)
Hypertension	15 (53.6)
Ex-smoker	10 (35.7)
Peripheral vascular disease	2 (7.1)
Left ventricular ejection fraction between 30% and 50%	6 (21.4)
Logistic EuroSCORE	6.9 (6.4, 16.6)
Aortic segments operated on	
Root	22 (78.6)
Ascending	26 (92.9)
Arch	8 (28.6)
Concomitant cardiac surgery	
Aortic valve replacement	26 (92.9)
Coronary artery bypass graft	2 (7.1)

As seen in Table 4.1, the CCS Angina Grading Scale is a well-established method for the classification of angina severity. It was first published by Campeau in 1976 and has since been adopted and used in a wide range of international healthcare institutions and studies. It consists of four grades of increasing magnitude, described in Table 4.2:

Table 4.2: The CCS grading of angina pectoris

CCS Grade	Description
I	Ordinary physical activity does not cause angina, such as walking and climbing stairs. Angina with strenuous or rapid or prolonged exertion at work or recreation.
II	Slight limitation of ordinary activity. Walking or climbing stairs rapidly, walking uphill, walking or stair climbing after meals, or in cold, or in wind, or under emotional stress, or only during the few hours after awakening. Walking more than two blocks on the level and climbing more than one flight of ordinary stairs at a normal pace and in normal conditions.
III	Marked limitation of ordinary physical activity. Walking one or two blocks on the level and climbing one flight of stairs in normal conditions and at normal pace.
IV	Inability to carry on any physical activity without discomfort, anginal syndrome may be present at rest

Also seen in Table 4.1, The NYHA functional classification of heart failure was proposed in 1928 and has been revised several times since, most recently in 1994 (Criteria Committee of the NYHA). It places patients in one of four categories based on how much they are limited during physical activity. In a similar way to the CCS classification, it consists of four grades of increasing magnitude which are described in Table 4.3:

Table 4.3: The NYHA functional classification of heart failure

NYHA Grade	Description
I	Patients have cardiac disease but without the resulting limitations of physical activity. Ordinary physical activity does not cause undue fatigue, palpitation, dyspnoea (shortness of breath) or anginal pain.
II	Patients have cardiac disease resulting in slight limitation of physical activity. They are comfortable at rest. Ordinary physical activity results in fatigue, palpitation, dyspnoea (shortness of breath) or anginal pain.
III	Patients have cardiac disease resulting in marked limitation of physical activity. They are comfortable at rest. Less than ordinary physical activity causes fatigue, palpitation, dyspnoea (shortness of breath) or anginal pain.
IV	Patients have cardiac disease resulting in inability to carry on any physical activity without discomfort. Symptoms of cardiac insufficiency or of the anginal syndrome may be present even at rest. If any physical activity is undertaken, discomfort is increased

The third and final characteristic from Table 4.1 which may need additional explanation is the logistic EuroSCORE. EuroSCORE stands for the EUROpean System for Cardiac Operative Risk Evaluation, and was originally published by Roques et al in 2003. It is a widely used method of predicting the chance of an individual patient suffering in-hospital mortality after undergoing heart surgery, including patients who undergo surgery on the thoracic aorta. The logistic model produces a risk score for each patient based on their personal risk factors ranging from 0.9% (even in the fittest patients, cardiac surgery still carries some risk) to 100%.

4.4.2 Identification of qualitative themes

The interviews with TAA patients resulted in powerful accounts about how having TAA affected their daily lives across the 3 HRQoL domains of

physical, mental and social. Three main themes related to the effect of TAA on HRQoL emerged

- 1) Effect of disease and treatment (symptoms, personal adjustment to recovery and treatment satisfaction) on everyday life
- 2) Attitudes of and towards friends and family
- 3) Continued diagnostic monitoring for aneurysmal disease.

4.4.3 Effect of disease and treatment

4.4.3.1 Symptoms

All of the study participants explained how they experienced a range of symptoms, from having no symptoms whatsoever up to debilitating chest pain and blacking out into unconsciousness. Participants also discussed how their physical symptoms affected their emotional state and their psychological response and reaction to their illness.

4.4.3.1.1 Asymptomatic

The majority of participants (n = 18) recalled expressing surprise and disbelief when they were informed they had a serious health issue that would require surgical treatment as they had experienced no symptoms up to that point, for example:

"I didn't have any symptoms or nothing. Even now I wouldn't know I have an aneurysm because I have absolutely no pain." – P01

"No, I was never ever, there was never any symptoms as such like a shortage of breath or even under stress or under, I never felt any symptoms." – P12

Asymptomatic participants reported being incredulous when told about the seriousness of their condition, which may indicate the presence of a larger psychological burden when coping with aneurysmal disease, instead of the clear physical burdens which come with pain or discomfort. These participants exhibited a tendency towards disbelief of what they were being told by the doctors:

"They tell you that you have got a heart condition and you think what the hell are they talking about here?" – P18

"I said the crazy thing about it I feel great...I cannot understand why I am going to have this operation when I feel so good" – P21

Naturally, this situation led to individuals reporting surprise and at times shock, suggesting again that there is a psychological toll that these patients endure that is not reflected with typical physical symptoms:

"nothing at all like that,[shortness of breath], he just told me about it and I said "bloody hell", shock" – P28

4.4.3.1.2 Fatigue and collapse

Eight participants recalled that they were becoming tired and fatigued more easily. This was sometimes reported as being something they were aware of at the time:

"It was making me feel tired a lot as well" – P20

Another participant however identified their increased exhaustion only in retrospect:

"looking back now I probably had the symptoms from about then, where I was becoming more tired and lethargic" – P09

Two participants reported suffering a sudden collapse, one in the course of their normal activities and the other when exerting themselves on a demanding bike ride:

"I just collapsed and I think that it when it all stemmed from, that is where it all came from" – P19

"then the worst bit was cycling up a big hill in Maldon where I work I got the top then I collapsed in a ditch" – P25

These patients then in due course consulted their GP, which led to the discovery of the aneurysm and the subsequent procedure.

4.4.3.1.3 Angina

Several participants identified chest pain, or angina, as being a factor during their pre-operative experience and as an indicator that they would have to seek some medical attention, for example:

"when I was in my 30's I knew it was getting worse because I was having a lot of chest pain really," – P20

This was often described as happening when taking light exercise:

"probably up to about 18 months beforehand I started to get pains in my chest...and couldn't walk really far could not do all the things I used to do" – P05

"Yes I was ... getting a pain in my chest that's it, if I was walking somewhere maybe after 10 minutes I had to stop, stop and rest and try and carry on again" – P05

"Initially I had a slight tightness in the chest, I was exercising and particularly when I was out walking the dogs I would get just occasionally a slight tightness...I thought I probably ought to get this checked out. So I was diagnosed with angina" – P07

Angina does not always originate in the chest, but can also radiate through the arms, neck or jaw, as articulated by this participant:

"I was getting pains in the jaw line and a bit down my neck did not know what it was" – P03

As with the earlier identified symptoms of exaggerated tiredness or collapse, these unusual pains led to a GP attendance and ultimately the diagnosis of the individual's aneurysm.

4.4.3.1.4 Shortness of breath

All of the symptomatic participants (n = 12) spoke of experiencing shortness of breath, or dyspnoea, in the months to years before getting their aneurysmal diagnosis.

"I was getting out breath quite easily you know which was unusual for me" – P21

"yes I think I did slow down I don't think I realised it but my husband says now you were slowing down and you know getting a bit out of breath" – P30

This was again particularly felt during or after exercise or normal exertion.

"I can't even walk up the street. I get out of breath" – P02

"I was short of breath as well and you know I could not do much" – P20

Participants noted how their physical abilities were diminishing compared to what they were previously used to. The dyspnoea was described as 'getting puffed' by more than one participant, and was sometimes described together with the chest pain symptom.

"my quality of life was good although I was getting very puffed out which was starting to slightly puzzle me why and where that was coming from" – P08

4.4.3.1.5 *Dizziness*

One participant mentioned that they became dizzy during this pre-operative period, and that experience was the impetus for them to visit their GP:

"I had a dizzy spell went to my Doctor and he pressured me into having some tests" – P08

This quote also demonstrates the gap between relatively mild symptoms contrasting with the potential for catastrophic health outcomes. It is possible that the patient visited their GP dutifully, seeking reassurance rather than an extended course of treatment for an unexpected health problem. That their doctor had to then "pressure" the patient into having more tests suggests that their perception of risk was low and they were not anticipating a lengthy course of action.

4.4.3.1.6 *High blood pressure*

Three participants explained how the diagnostic tests for their known high blood pressure, or hypertension, led to the discovery of their aneurysm

"he said to me well your blood pressure is sky high...he offered me a chest X-ray and the X-ray showed that I had a slightly enlarged heart " – P03

"I had intermittent hypertension where I would go for an echo and they would say that my blood pressure was reading high" – P09

"my blood pressure was high and they sent me for an MRI scan" – P23

These quotes exhibit a category of patient who already appear within the healthcare system for one fairly common condition, high blood pressure, who then move to a more serious stage of diagnosis. High blood pressure is reasonably well understood condition, and patients will be aware of the risks. The difference between the previous 'Dizziness' patient being "pressured" into more tests and the hypertension patient taking up the "offer" of a chest

X-ray may reveal a gap in attitude between individuals who do not expect their symptoms to be an indication of a more serious condition, and those who do.

4.4.3.1.7 *Psychological*

The psychological effects of dealing with an aneurysmal diagnosis were discussed by many of the participants.

A feeling of having a life-threatening condition that could only be cured by a significant operation, not without its own risks, weighed heavily on the minds of many participants. The awareness that their aortic aneurysm could be growing without their knowledge and ultimately had the potential to dissect and cause disaster led to a range of significant negative emotions. Everything from anxiety and stress through to anger, horror, fear and depression were mentioned.

Feelings of impending doom were related by the following two participants, describing the potential for dissection as their aorta “bursting” or “popping”. These quotes go some way towards highlighting the powerlessness that these individuals feel over the aneurysmal disease:

"perhaps it's because I know it can burst so I go to bed at night thinking am I going to wake up in the morning...It stresses me to think that I could die at any time....you think Christ what if I die tomorrow, what about my poor husband." – P02

"and it's all horrible isn't it you don't expect to be told you have got this bulge that is going to pop and again if you feel absolutely fine and you cannot really see the point of the operation apart from stopping you dropping dead in the street" – P26

Fear was a common theme, and participants spoke of difficulties with coming to terms with their situation. One participant who was a health professional explained how having knowledge of the condition exacerbated her fear:

"Petrified, I was petrified and annoyed I had an aneurysm because I did not know how big it was, I am a nurse by profession so...I understood...the implications of it, I have done many a fast track to theatre you know blue lighting with patients that I know had aneurysm's and to find that I was in that position myself was [a] very, very scary and lonely time because the people around me kind of didn't understand the full implications of it, I probably knew too much about it and that was a really hard place to be...so I was very frightened sort of put my life on hold frightened of moving because I did not know what I was dealing with, did not know how big it was" – P09

This knowledge was seen as a disadvantage and also made the participant feel isolated, as people close to her did not share that understanding.

Many participants were worried about surviving the operation:

"you only know that you know you have got this major operation in front of you your head's swimming and you just think oh my god how am I going to cope with all this and what is going to happen am I going to come through it" – P13

Some participants had experience of losing a relative to the same condition which compounded their distress:

"it was scary really I was worried because then, that was what my dad died with an aneurysm...and I thought "Oh God, blimey", I thought, is that going to be me now. " – P14

One participant expressed their extreme emotional reaction to living with the aneurysm, as they experienced depression and suicidal thoughts:

"it's depressing more than anything it's a condition really it attacks you so hard some days that you just feel that you want to end it all" – P27

The experience of psychological distress was common and significant for most of the participants, which corresponds with previous research findings in this area.

4.4.3.2 Personal adjustment to recovery

After the patients had undergone surgery, several themes emerged from their recovery period. These ranged from managing expectations of healing times and lifestyle changes, to coping with how the surgery had affected and altered their bodies.

4.4.3.2.1 *Expectation of recovery time*

Participants articulated how they felt differently after their operation, both physically and psychologically. These two participants mentioned how they had to readjust their expectations of how quickly they would recover from their operation:

"I was surprised it took me so long to recover I just thought I would bounce straight back." – P04

"I can say it can take you 10 months after the operation I started feeling close to being back to normal." – P05

Another participant spoke of the profound differences they experienced within themselves after their operation, beyond physical symptoms and abilities they felt that their personality had changed and were having to adjust to a 'new normal'.

"I didn't prepare myself for how difficult it would be...I don't think I was prepared for that. To not be the same person after it...I was very introverted."
– P16

This account reflects a significant impact on personal wellbeing but also on the psyche and sense of self which goes beyond physical symptoms.

4.4.3.2.2 *Unexpected replacement valve noise*

Two of the participants who had received a mechanical aortic valve replacement spoke about their experiences of the prosthesis making noises that they were unaccustomed to:

"I was never prepared for the noise of your heart beating ... even though I was told about it ... so when you are lying there when you get to hear the valve, okay and when I am lying there I can hear, it is funny. But the worst and funniest thing is that I was on the golf team and you know it's very quiet and I was standing next to this bloke and I held my breath and he said what's that beating noise and I goes it my valve, he would go its spooky that it, put him off" – P25

"because I think as well having had the two valve replacements I can hear it is so loud you know the whole family..." – P30

These quotes seem to oscillate between making light of the situation – “put him off” – and having an understanding that the “spooky” heart sound makes people regard them in perhaps a negative or at the least an unfamiliar way. The replacement valve noise therefore has an effect on social interactions, possibly causing negative feelings for the patient such as embarrassment or shame. The contrast between describing the noise as “the worst and funniest thing” is interesting, the valve noise is perhaps a constant reminder both of a patient’s mortality and of their resilience and ability to overcome such invasive surgery and approach the unusualness of their situation with humour, or at least how strange the noise may seem to others.

4.4.3.2.3 *Limiting activities*

Participants reported limiting their activities in different ways. One individual was obviously keen to begin doing the things they were used to doing, but they restricted themselves as they did not want to produce any setbacks in their recovery:

"I have not yet lifted heavy objects yet because I do not know quite when I can do that they say 12 weeks but I think I could probably do it now but you know rather than risk it" – P12

Another participant reported limitations in their activities due to how they felt after their procedure, another example of the personal differences that can be brought about by the surgery:

"I am not as active now not even close, and I couldn't be" – P19

These differences in ability to recover could be caused by a variety of factors. The age of the patient and the extent of the operation performed may have an effect, and the capacity of the patient to adjust to a new post-operative sense of self may dictate how much activity they feel capable of.

4.4.3.2.4 Diet and alcohol

Two participants reported differences in their outlook towards their diet and to alcohol consumption. These comments may demonstrate the influence upon lifestyles that having a shocking health-related diagnosis and then a significant operation such as aneurysmal repair can have on a person's lifestyle.

"the only sort of things that we would do we changed our diet because we used to pig out we were foodies ... and I thought you know get rid of a couple of stone it has to help" – P29

"I am not drinking as much as I used to which is another good thing, I like to have a drink now and again but obviously not too many" – P20

These comments seem to reflect an increased attention to personal health and a mindfulness of how moderating food and alcohol can have a positive effect on mood and outlook. Losing weight "help[s]" and not drinking so much is a "good thing". Although patients had little influence over the growth of their TAA and the necessity for the ensuing operation, they do have a certain

amount of control over their own lifestyles. Exerting this control in what they feel is a constructive way may allow them to feel more optimistic about their health in the future.

4.4.3.2.5 *Back to wellness and 'normality'*

In contrast to the participants who reported feeling less capable and having to adjust to a reduced capacity for activity, or even changes in their personality, this participant felt that after a period of time they were back to a normal state:

"After the operation after 12 months everything is fantastic and I am feeling fine now... probably 12 to 18 months after surgery before you knew it, I suppose I felt no discomfort in my chest, I could feel a sort of change that week in myself, I could put on a shirt, it was getting comfortable." – P05

These participants even reported feeling better than ever, with improvements in both physical and emotional wellbeing:

"I am better now than I was, even before I was getting loss of breath at the start I am a lot better, I am not as narky " – P22

"you know I was healthy before the operation ... so I felt really good, it's as good as that now if not better, I feel as physically better as I did you know in my late 20's" – P05

"it was only after about 6 months ... I felt wonderful I felt like I was 16 again I felt, my heart felt, I had energy, ready to go out, I was walking and I felt absolutely brilliant." – P09

These comments are interesting as they touch on a range of physical, symptomatic and psychological aspects that patients clearly associate with their TAA disease and treatment. A routine action that would have previously been carried out automatically, such as putting on a shirt with no discomfort, is now a physical milestone on the road to recovery. Overcoming

symptomatic problems such as shortness of breath are conventional indicators of a good health outcome, but being less frustrated, bad-tempered or “narky” are the sort of psychological issues that a PROM is specifically designed to detect changes in, and that traditional indicators cannot. Two of these quotes also refer to the patient feeling young again, which could be interpreted as a new lease of life. This may display a perspective of how a successful recovery from debilitating disease and invasive treatments can rejuvenate people, living under the shadow of conditions like TAA disease and then coping with healing and renewal after intensive surgical treatment would take a toll on most people’s mental and physical strength. Once patients have come through this period, they unsurprisingly feel revitalised, full of “energy” and “as good as...if not better” than before their operation.

4.4.3.2.6 *Negative body image*

One female participant mentioned how they suffered from a negative body image coming from their sternotomy scar. This may be another way in which the course of treatment for aneurysmal disease affects patients psychologically as well as physically:

"the body image was really hard at first I could not look at myself, let alone anybody else look at me" – P09

"I get really up tight when somebody says when they see my scar, I don't often, I usually shy away from the camera" – P09

The presence of a sternotomy scar following cardiac surgery has been previously identified as a source of distress for women in particular. King (2009) found that although women appreciated that the presence of their scar was part of the trade-off between achieving better health and recovery from a life-threatening disease, they also found it an upsetting reminder of the health scare they experienced and the challenging surgery they had been through. Their scar made them feel less attractive and they perceived that they were being judged by others when it was visible in public. The

quotes above seem to reinforce these conclusions, and support the suggestion that patients could be both better prepared for the presence of a surgical scar in pre-operative meetings and better assisted to cope with the potential for a negative body image in post-operative rehabilitation or counselling.

4.4.3.2.7 Determination

As well as the pessimistic feelings that came with disease diagnosis, treatment and recovery. There were several participants who made a point to continue being positive, being determined to overcome their personal difficulties and not accepting the intrusion of negative thoughts:

"you know you got to get on with life...I can't stand negative ... like this lady ... she is sitting there like an 80 year old and you know I am thinking, you only get one life live it. That's what it's all about" – P14

A stoical, philosophical attitude was also in evidence, with a resolve to rise above their situation and not wallow in hopelessness:

"sometimes your dealt a rough hand aren't you but there is no point complaining and crying and moaning all the time just get on with it and enjoy what life you have got, none of us know what is going to happen." – P26

The substance of these quotes shifts the focus away from an introspective characterisation of personal events towards what appears to be an instinctive, broader understanding of how a positive approach to objectively undesirable situations produces optimism and possibly a smoother road to recovery. Rather than focussing on previous bad luck or subsequent hardships, making the most of their present situation may be the best way these patients know how to make sense of their experience.

4.4.3.3 Attitudes of and towards friends and family

The families and friends of participants unsurprisingly had a significant role to play when they were discussing their reactions to their experiences, and their support and attitudes towards the participants made deep impressions on how they lived through the events surrounding their aneurysmal treatment.

4.4.3.3.1 *Understanding the disease*

Several participants reported that when they tried to discuss their situation with their loved ones, they found that they did not have much knowledge of what aneurysms and their associated risks were, or the differences between aortic disease and cardiovascular disease:

"I don't think people realise what it is you know " – P14

"sometimes that you don't tell people because they instantly think you have had a heart attack." – P25

Even with family members who had no particular preconceived ideas about the problems that the participants were facing, there was sometimes a gap in understanding:

"they have just sort of been the same as me really not been able to sort of get their heads around it" – P12

This participant experienced a wide divergence between what they understood to be the risk of their aneurysm, and how their son seemed to be processing the information:

"in terms of family, a lot of people did not understand...my son, no didn't even register with him at all, I don't think he understood the severity, I did try and speak to him but then I thought well maybe ... apart from force it down their throats and say 'do you realise what this means?!'" – P09

This disparity in understanding and difficulty with enabling others to comprehend the significance of their situation may contribute to negative thoughts and feelings, patients may experience a cognitive tension between wanting people to appreciate the seriousness and potential consequences of TAA disease but also not want to worry their loved ones too much.

4.4.3.3.2 *Support*

Conversely, other participants were keen to express how supportive the people closest to them had been:

"Relationships were fine the wife was supportive" – P20

Relying on the strength of these cherished relationships was clearly something that these two participants felt was a comfort to them throughout their experience:

"I had good support from my husband...[he]'s really good and I do talk to him about it ... we have got a good relationship and we are open and we are honest with each other....I would not have got through it without him, he has just been wonderful" – P09

"but luckily I had family that were very strong you know" – P27

It is possible that patients who do not have reliable and sympathetic support from their family or friends will encounter a more difficult recovery period. Having access to some loving reassurance, being able to share responsibilities and knowing that there is someone present who can care for and assist you if there are difficulties would clearly be a comfort to patients both before and after TAA surgery.

4.4.3.3.3 *Not informing people*

Two participants reported another reaction to the news of their diagnosis. Although their nearest friends and family were involved, they did not necessarily tell people in their extended social and family network about their condition. This first quote shows that they are trying to protect these others, as the participant does not want them to experience any negative emotion or anxiety by being concerned about their situation:

"I just did not bother telling them so no big deal, I didn't need them to worry"
– P25

This reluctance to speak about their health and not want others to react may be part of a coping mechanism, minimising the effects of their problems externally so that they do not see them as insurmountable internally.

"to be quite honest with you we never talked about it, I come back and I didn't say nothing" – P28

These patients offer fairly blunt responses, which may be a further indication of how they have socially managed their experience of TAA disease and treatment. By not engaging in communication and preventing others involvement, they may be protecting themselves from unwelcome reactions. "No big deal" for the patient either way; if the reaction of their extended family and social network had been less than they might have anticipated, then they do not feel slighted. If the reaction made it obvious that they had placed extra concern and anxiety on these people, then they are protected from carrying that with them also.

4.4.3.3.4 *Preparing for the worst*

With the potential outcomes of their surgery including the possibility of death, some participants realised that they should prepare financially for that possibility. It is likely that these two individuals found some peace of mind in

knowing they had taken care of their family to the best of their ability, in case the treatment outcome had been a poor one:

"I just prepared that at least I'm getting an income coming in the family would get money if they needed to depending on what happened with me...I did put things in place just in case for my family" – P09

"I was more worried about me being the main bread winner and that was a concern for checking all life insurance policies and we were covered which was lucky so we would be ok if the worst happened so that helped me a little bit but that's all I thought about trying to get through the normal stuff just in case the worst happened." – P16

These comments highlight the unusually intense situation of knowing that treatment to resolve TAA disease carries with it the risk of death. They are a reminder that the strategy of surveillance, or 'watch-and-wait', exists because an invasive treatment for TAA disease can involve a greater threat of mortality. Patients naturally find reassurance in their situation by concentrating on "normal" personal administration tasks and relieving some amount of anxiety.

4.4.3.4 Continued diagnostic monitoring for aneurysmal disease

Patients who have surgery for aneurysmal disease are often genetically predisposed to the condition (see Chapter 2.3.4). This means that all patients are routinely offered follow-up monitoring to identify any further aneurysms which may occur. Interview participants were recruited from these follow-up monitoring clinics, so they had unique and current insights into how these made them feel.

4.4.3.4.1 *Reassurance*

This participant reported being reassured by these clinic visits, seeing them as check-ups and not letting negativity about what may be found affect them:

"I am doing more in some ways because I am not thinking about the problem I had, it's been fixed I just get checked up now" – P03

Another participant said that they appreciated getting their annual check-up:

"it is nice to know that we come back every year and get the MOT" – P21

The "MOT" terminology that this participant uses is striking, as it relates to the annual UK Ministry of Transport motor vehicle roadworthiness test. The patient is perhaps intending to defuse the threat of recurrent TAA disease by humorously minimising its importance and maybe slightly dehumanising themselves. Alternatively they are using this language to 'other' their disease. As TAA disease in isolation rarely has symptoms this disassociation makes sense, because even if there was a large TAA with a high risk of aortic rupture the patient may be oblivious. Patients with TAA disease are reliant on clinical experts to let them know if they have a problem with their own bodies, just as most car owners would not be aware if their car required a new oil filter or an exhaust fitting, they rely on garage mechanics to inform them whether their car is fit for travel or not.

4.4.3.4.2 *Apprehension about reoccurrence*

However, the same participant as in that last quote went on to admit that there was also apprehension and nervousness about the results of the annual check-up. This suggests that patients do not take for granted their 'aneurysm-free' status and may have underlying concerns about the possibility of further operations:

"you feel a bit conscious when you come back after you have had your scan and you think, oh, you know what are they going to find" – P21

The check-ups themselves, although necessary, are understandably a source of anxiety and trepidation. They are a reminder that although their TAA disease has been successfully diagnosed and treated in the past, there may be no physical symptoms or warning signs to let the patient know that they have a current problem.

4.4.4 Physical HRQoL

The physical element of participant's post-operative experience was discussed by all participants. This ranged from a brief outline of how they felt in themselves, to more complex, personal observations about their bodies capabilities and the changes that their health and healthcare had made.

4.4.4.1 Exercise

Several participants discussed how their activity capabilities and exercise regimes had developed and changed after their aortic aneurysm surgery. In a similar way to the activities item and the diet and alcohol item, this individual recognised that maintaining a healthier lifestyle was important in light of their treatment:

"I like physical work and I think it is probably good to keep in shape after something like this." – P08

Another patient highlighted the differences in what they felt capable of, and their mindfulness of what their limitations may be:

"I do small jobs, if it's a big job I will get somebody in to help" – P10

This quote is useful in emphasising how returning to exercise allowed this individual to feel like they were getting back to their normal state:

"I am back running now jogging, doing exercise, so yes I feel fine" – P12

These statements from patients reinforce the idea that TAA disease treatment represented a step-change in their awareness of personal health and how their own understanding of their physical capabilities seem to act as a yardstick both for recovery and for adjusting to a 'new normal'.

4.4.4.2 Lying in bed / sleep

This participant mentioned that sleeping could be challenging immediately after surgery. Their chest wound would understandably prevent them from making their normal movements and lying down in their usual manner when they were in bed.

"I have to get out of bed to turn over because I cannot do it any other way" – P13

These two patients also report that getting enough quality sleep in the initial weeks following surgery can be a challenge:

"when you come out of hospital and you go to sleep on your back, sleeping on your back propped up is just the most awful thing " – P25

"I did not sleep well for about two weeks" – P26

Not being able to get a good night's sleep may also restrict post-operative healing and generate a low mood. Redeker (2004) studied the sleep habits of patients after cardiac surgery and found an association between sleep patterns and changes in physical function and emotional wellbeing.

4.4.4.3 Intimacy

Participants also spoke about their reactions to others in terms of intimacy. This ranged from individual to individual from cuddling, kissing to sexual contact. This participant highlights the difficulties and breadth of feeling when becoming intimate with their partner:

"Some weeks, sometimes it is a bit of a roller-coaster, one minute were okay and then other times I will shy away from it" – P09

This patient emphasises the way in which pain dictated how much intimacy they felt comfortable with:

"the first couple of weeks there was no cuddles for anybody I can tell you that, because [of] the pain" – P25

Other studies have shown adverse effects on sexual activity in patients who underwent cardiac surgery. In the early post-operative period Foruzan-Nia (2011) found a significant increase in male impotence and premature ejaculation along with a decrease in libido, while females may experience unusual sensations in their breasts such as pain, numbness, tingling, burning, or heaviness (Steinke, 2013). Coupled with this is some evidence of a reluctance from healthcare professionals to discuss these issues with patients, with several studies demonstrating the reluctance of nurses to counsel patients with heart disease about their sexual health concerns (Hoekstra, 2012; Barnason, 2013 and Wang, 2019).

4.4.5 Psychological HRQoL

The psychological element of participant's post-operative experience was characterised by both negative and positive emotions. Even though their health problem had been treated successfully, there was still some discourse about how they were not necessarily content. However, this trepidation was also counterbalanced by individual's drive to not be overwhelmed by events.

4.4.5.1 Low mood / negative thoughts

After their operation, this patient reports feeling negative emotions. Thinking about "everything" suggests that they were stunned and exhausted by the treatment:

"you just think about everything and probably a lot of it was negative " – P19

This participant reports being scared and feeling lonely:

"then you go home and you think I am on my own, I am on my own what am I going to do and its frightening" – P27

Being discharged from a hospital full of health professionals back to a patient's home can clearly feel like a big step if they do not feel properly supported or able to take care of themselves.

4.4.5.2 Overcoming adversity

Although participants had in many cases been through a life-altering health situation, in a similar way to the determination item exhibited pre-operatively, they refused to be laid low by their circumstances.

"I would not, not do anything I would find a way" – P09

This participant endured through their aneurysmal treatment, and finds intrinsic value in that alone:

"Never mind, I am still here to tell the tale" – P14

These statements show the participants finding strength in their perseverance and survival.

4.4.5.3 Vulnerability

One participant reported feeling emotionally vulnerable when confronted with the prospect of being physically challenged:

"we walked past the shops and there were a gang of lads there and I felt really nervous and felt dead vulnerable" – P16

This quote highlights a potential link between a perceived possibility of either verbal abuse or physical threat from the group of youngsters, leading the patient to consider their own reduced physical effectiveness in such a situation in the aftermath of TAA surgery, the further consequence being a negative emotional response. It would not be hard to conceive a situation where post-operative TAA patients chose to avoid unpredictable scenarios such as this, maybe leading to their normal activities or social life being restricted.

4.4.6 Social HRQoL

The social element of participant's post-operative experience was also shown to have been affected by their health condition.

4.4.6.1 Work

This individual reports that they took 10 weeks to return to work at a reduced rate. Moreover they identify boredom as being a motivating factor for getting them back into a routine:

"I think I was back in work after 10 weeks, obviously not doing as much as I used to...I suppose I was getting bored at home, probably just wanted to get back into a routine" – P20

This participant quote demonstrates the challenge of returning to work, and the slow, structured way in which they had to build up their return in order to cope with the additional activity:

"I started work by doing 1 day, 2 days, 3 days, 4 days and I eased myself back in " – P25

Effective self-management and an innate understanding from the patients themselves of what will improve their psychological wellbeing, without

disturbing their physical and emotional recovery are suggested in these quotes.

4.4.6.2 Relationships

One particular participant spoke about struggling both with the regularity of people asking for information on their recovery:

"and in the end I just said when anything gets sorted I will let you know stop phoning up!" – P27

And also with their inevitable limitations and weaknesses during the recovery period:

"and my best friend ever, I kept saying to her I am a rubbish friend, why are you a rubbish friend, because I had to call you and make you come down" – P27

These quotes illustrate the emotional confusion that patients can feel during this post-operative period, and the negative thoughts and feelings that can occur when an individual's body has an invasive surgery and is placed in an unfamiliar state of recuperation.

4.4.6.2 Leisure time

Participants were also proud of themselves for getting over their operation and achieving noteworthy accomplishments after their TAA surgery. It seems clear that this participant feels satisfied and uplifted by what he has managed to achieve in his leisure time:

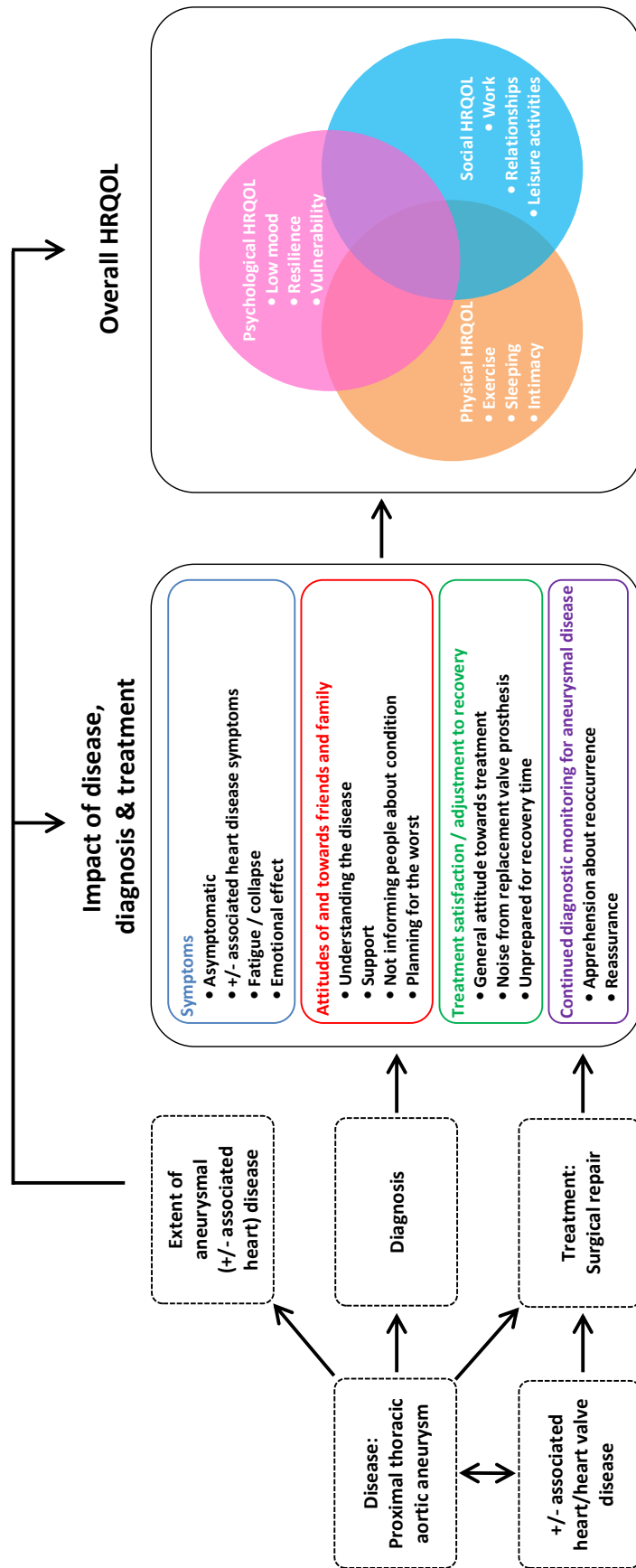
"they do not believe you could do the 35k having had, you know, the open heart surgery in January and that was quite a hard bike ride to do let me tell you some big bloody hills in that but so some people must feel a bit shocked that you can recover so quickly" – P25

They also seem gratified by the speed of their recovery and how they have managed to get back to doing what they enjoy in spite of what others may have thought.

4.4.7 Conceptual framework

Conceptual models are graphical representations that organise concepts and establish possible relationships among the concepts. Figure 4.1 summarises the results of the qualitative study, suggesting a conceptual model on TAA and HRQoL that links perceptions of receiving a diagnosis of TAA and undergoing subsequent surgical treatment, to satisfaction towards that treatment, attitudes of and towards friends and family and perceptions regarding the value of continued clinical monitoring of the TAA disease based on the data. The findings are then mapped onto the physical, psychological, and social HRQoL domains outlined in Sections 4.4.4, 4.4.5 and 4.4.6. The extent of the aneurysmal disease along with any associated heart disease may also influence the perceptions and experience of the disease, treatment and HRQoL.

Figure 4.1: Conceptual model



4.4.8 Perceptions of TAA patient QoL amongst health care professionals

A separate series of four interviews were conducted with aortic surgeons in order to identify their views on the impact of TAA disease on their patients' daily lives. They were asked how, in their opinion, patients responded to their diagnosis of TAA disease, how patients experienced TAA disease in their daily lives and how surgical treatment affected patients day-to-day activities, with particular regard to any physical and psychological changes. They were asked to describe changes patients might notice in the short- and long-term in relation to symptoms, physical, psychological and cognitive functioning, and any complications. Their responses were used to support the conceptual model of proximal TAA disease derived from the patient interviews and their insight into patient behaviours was used to further tailor the questionnaire items to the target cohort.

Discussion with the consultants also suggested a requirement for three separate PROM deliveries. A pre-operative PROM (Q1) and two post-operative PROMs (Q2 and Q3). Q1 would be delivered pre-operatively, Q2 at 6 weeks post-operation and Q3 at 3 months post-operation. The surgeons felt that the recovery from the surgery could be arduous, so patients may actually feel worse at 6 weeks post-operatively than they did beforehand. Then after 3 months the feeling was that although there may be some residual discomfort, patients should be returning to normal activities.

4.5 Questionnaire construction

The results of the qualitative research phase and the literature review formed the basis for construction of the proximal TAA PROM. Essential components identified for inclusion in the disease specific section of the PROM included: symptoms (or lack thereof), restrictions in physical activities, psychosocial function (anxiety, fear of death and / or pain, low mood, uncertainty, self-efficacy, frustration and avoidance of activities, impact of TAA disease on

family and friends, independence, interference with social activities) and cognitive function (reasoning, memory, attention and concentration). The general health measure included in the PROM was the EuroQol EQ-5D-5L (see Section 2.4.3 and Herdman, 2011) and the EuroQol EQ-VAS (Rabin, 2001). Permission for using these instruments for the purposes of this study was sought from and granted by the EuroQol Research Foundation.

Additional post-operative items identified for inclusion included physical and psychological complications, treatment satisfaction (including information received, expectations about the impact of the operation and the strategy for follow-up TAA monitoring) and a single 'Friends and Family Test (FFT)' question. The FFT is a commonly used patient experience tool that has been used in a variety of settings within the NHS, it has produced over 65 million pieces of feedback so far (NHS England, 2019) and was considered to be a concise and potentially valuable inclusion.

All 3 versions of the proximal TAA PROM include the four disease specific domains: Symptoms, Physical Function, Psychosocial Function and Cognitive Function. They also include the EQ-5D-5L and the EQ-VAS general health measures.

The Q1 pre-operative PROM includes 3 items not found in the post-operative Q2 and Q3 PROMs, these relate to time since diagnosis, previous operations for heart conditions and a list of co-morbidities.

The Q2 and Q3 post-operative PROMs are identical, apart from an introductory wording change from "six weeks" to "three months". They include 17 items not found on the Q1 PROM which relate to post-operative complications, treatment satisfaction follow-up hospital care and the FFT.

4.5.1 Item generation

Items were generated for each specified domain. Items were either borrowed or modified from the CROQ (Schroter, 2004), or newly created. The

Symptom domain includes 12 items: 3 items were borrowed from the CROQ, 1 item was an amalgamation of 2 CROQ items and 8 items were newly created. The Physical domain includes 9 items: 7 items were borrowed from the CROQ and 2 items were created as a result of splitting a single further CROQ item into 2. The Psychosocial domain includes 16 items: 13 items were borrowed from the CROQ, 1 was a modified CROQ item and 1 item was newly created. The Cognitive domain includes 3 items: 1 item was borrowed from the CROQ and 2 were modified CROQ items.

The items associated with these domains can be found in Table 4.4.

Where possible items were borrowed from the CROQ, which in fact sourced nearly half of its own items from other well-validated, psychometrically sound questionnaires. The language of each item was altered slightly to shift the focus towards the patient's "aortic aneurysm". However, some of the necessary items, especially in the Symptoms domain, were not well covered by the CROQ. New items were constructed where the necessary subjects were not adequately covered by the CROQ or other existing questionnaires. Interview themes and the relevant literature were consulted in order to identify further items for inclusion.

Particular effort was made to construct simple and specific questions. Unusual or ambiguous phrases (Schwarz, 1999), and double-barrelled questions (Streiner, 2015) were avoided. Attempts were made to keep the text of the items as short as possible, as longer items have been shown to have poorer validity (Holden, 1985). All items included required a response, as Fayers (2013) contends that including questions which are not applicable to a subset of respondents can have the consequence of increased missing data. Underlining and the use of bold font were used to emphasise the time of reference along with the terms "heart condition or aortic aneurysm" (Ringash, 2016). This was done to help the patient focus on the specific problem and the impact of their aortic aneurysm / surgical treatment on their day-to-day life.

Table 4.4: Items associated with PROM domains

Item	Symptoms domain	Physical domain	Psychosocial domain	Cognitive domain
1	Pain or discomfort in your chest **	Moderate activities, such as moving a table, pushing a vacuum cleaner, bowling or playing golf *	Family or friends being overprotective towards you? *	Have difficulty reasoning and solving problems (eg making plans, making decisions, learning new things)? **
2	Pain or discomfort in your back ***	Lifting or carrying groceries *	Feeling like a burden on others? *	Forget things (eg events that happened recently, where you put something, appointments)? **
3	Pain in your chest that radiates to other parts of your body (eg arms, shoulders, hands, neck, throat, jaw, back) *	Climbing one flight of stairs *	Feeling restricted in your social activities (like visiting with friends, relatives, etc)? *	Have difficulty doing activities involving concentration and thinking? *
4	Tiredness / lethargy ***	Climbing several flights of stairs *	Feeling worried about going too far from home? *	
5	Feeling dizzy, lightheaded or faint ***	Bending, kneeling or stooping *	Feeling restricted in tactile gestures (like being touched or giving hugs)? *	
6	Feeling unsteady or uncoordinated ***	Walking 100 yards *	Worried about your heart or aneurysm? *	
7	Shortness of breath ***	Walking half a mile *	Worried about doing too much or overdoing it? *	
8	Palpitations (a strong or irregular heartbeat) *	Bathing yourself **	Worried about dying? *	
9	General physical weakness ***	Dressing yourself **	Frightened by the pain or discomfort of your condition? *	
10	Changes to your voice (hoarseness, or differences in tone) ***		Uncertain about the future? *	
11	Problems with eyesight (eg blurred or double vision) ***		Depressed, or in a low mood? **	
12	During the past 4 weeks, how much trouble has your heart condition or aortic aneurysm caused you? *		Frustrated or impatient? *	
13			Anxious or upset? ***	
14			That your condition interfered with your enjoyment of life? *	
15			That it was difficult to keep a positive outlook about your health? *	
16			That it was difficult to plan ahead (eg holidays, social events, etc.)? *	

* CROQ item

** Modified CROQ item

*** Newly created item

4.5.2 Focus group

The appropriateness and acceptability of the proximal TAA PROM instrument was evaluated by a focus group discussion. Focus groups have been widely used in health research to explore the patient perspective (Carr, 2003; Côté-Arsenault, 2005). They are often included in mixed-methods studies to gather information on the suitability of questionnaire construction or to help interpret results (Creswell, 2017; Kroll, 2005). Tausch (2016) notes that the intention of having a group process is to help people recognise and clearly explain their views, this is considered to be a significant benefit that focus groups have over individual interviews. Tausch goes on to say:

“The group functions as a promoter of synergy and spontaneity by encouraging the participants to comment, explain, disagree, and share their views. Thus, experiences are shared and opinions voiced that might not surface during individual interviews.”

72 patients with a history of proximal TAA disease were identified from the LHCH clinical database. They were each telephoned and invited to attend a 2 hour focus group to discuss the structure, content and clarity of the PROM questionnaire. As an incentive for attendance, they would be given £10 towards their travelling expenses.

21 patients said that they would be interested in attending, they were sent an invitation letter in the post along with a copy of the pre-operative PROM (Q1) and one of the post-operative PROMs (Q3). They were asked to scrutinise these 2 questionnaires and consider:

- Is there anything that could be added or altered that would improve them?
- Are there any questions or terms that do not make sense, or are difficult to understand?
- Is any of the wording or the layout incorrect or inappropriate?

8 patients ultimately attended on the day itself. Those patients were arranged into 2 groups of 4, each with a member of the research team who guided the discussions and recorded their findings using a pen and paper.

Patient responses to the PROM instrument were generally positive:

- They were pleased that they would have the opportunity to report their own experiences.
- They felt that the questions covered the range of experience they had encountered.
- The length, layout and font were acceptable.
- Completing 2 post-operative PROM questionnaires was also acceptable.

However, patients did suggest some changes to the draft PROM instrument that were incorporated into the items listed in Table 4.4:

- The order of the items in the physical domain should be from easy to difficult, rather than difficult to easy.
- Another item in the Physical domain that related to “Bathing and dressing yourself” should be split into 2 separate questions.
- The item regarding “Depression” in the Psychosocial domain should be expanded to “Depression, or in a low mood”, as patients felt that the original term may be confusing. It was suggested that respondents may believe the question is relating to clinically diagnosed depression only, so less severe depressions may be missed.
- In the Cognitive domain, it was suggested that the first 2 items (“Have difficulty reasoning and solving problems” and “Forget things”) would benefit from examples of what was meant by this, in a similar way to some of the questions in the Symptoms domain.

4.6 Summary

These findings develop understanding of the complicated relationships between TAA disease, surgical treatment, treatment satisfaction, symptoms, patients' adjustment to recovery, TAA patient's attitudes of and towards family and friends, continued diagnostic monitoring for aneurysmal disease and HRQoL. The data outline perceptions of the disease and treatment in the context of a patient's everyday life.

The early post-diagnosis phase is characterised by physical and psychological components. Psychological reactions included shock, stress and anxiety, and occasionally depression and suicidal thoughts. Physical symptoms included overwhelming tiredness, sometimes resulting in collapse. Patients diagnosed with concomitant heart disease also suffered with shortness of breath, chest pains and dizziness. Attitudes towards the surgical operation itself were typified by acceptance and apprehensiveness. Post-operatively, participants described a period of adjustment related to physical condition, improvement in diet, awareness of their body's limits and weaknesses, challenges with sleep and intimacy, anxiety, depression, vulnerability and a negative body image. Also firmly present was a stoic determination to overcome these concerns, by drawing strength and motivation from family, friends and fellow patients. Where a heart problem had also been treated, participants tended to report a reduction in physical symptoms.

The conceptual model of proximal TAA disease is proposed based on an extensive literature review, in-depth patient interviews and agreement of importance of concepts by aortic surgeons.

Patient-based focus group discussions found the resultant PROM based on items derived from the conceptual model to be appropriate and acceptable, with minor changes to clarity and layout.

The subsequent chapter describes the methods and results of the pilot field test undertaken to evaluate the reliability, validity, responsiveness to change

and clinically important differences of the newly constructed proximal TAA PROM.

Chapter 5

Quantitative study

5.1 Introduction

This chapter presents the results of the quantitative study phase, this phase aimed to pilot the proximal TAA PROM. Preliminary field testing was undertaken to perform an initial evaluation of the PROM instrument. Standard quantitative assessments for questionnaires, including acceptability, feasibility, interpretability, precision, reliability, validity and responsiveness were addressed in the context of this newly proposed PROM.

5.2 Objectives

The objectives of this study were to:

- Plan and implement a practical approach to how TAA PROMs are administered
- Collect and store the data received from the completed TAA PROMs appropriately
- Carry out statistical analysis to assess the reliability and validity of the TAA PROM, including:
 - Assessing the internal consistency of the TAA PROM scores
 - Assessing the test-retest reliability of the individual items of the TAA PROM
- Present the results of the TAA PROM pilot study, and measure any changes in domain scores before and after aneurysmal surgery

5.3 Methods

5.3.1 Inclusion and exclusion criteria

All patients included in the study were inpatients at LHCH between October 2017 and March 2019. In order to be invited to participate, patients had to comply with the following inclusion and exclusion criteria:

The inclusion criteria for the quantitative study were:

- Participants must be age 18 years or over
- Presenting electively for surgery on proximal thoracic aortic aneurysm (TAA) disease
- Inpatient at LHCH

The exclusion criteria were:

- Under 18 years of age
- Unable to provide informed consent
- Recorded diagnosis of more acute aortic syndromes, such as aortic dissection, trauma, intramural haematoma or penetrating atherosclerotic ulcer

5.3.2 Pilot study recruitment and approach

Potential participants were identified from the consultant aortic surgeon's electronically networked theatre diaries. Proximal TAA cases were identified based on the theatre session listing within a Microsoft Outlook diary entry and confirmed by visual inspection of relevant letters (such as letters to the patient's GP about an outpatient attendance) and diagnostic documentation (such as typed results of CT scans confirming the aneurysm size) on the LHCH electronic patient record system.

Patients meeting the inclusion and exclusion criteria were sent an introductory letter in the post inviting them to take part in the study (see Appendix F1) along with a copy of the pre-operative PROM instrument (Q1; Appendix F2) and an envelope to return the completed questionnaire with a freepost sticker attached. This was scheduled to happen approximately two weeks before they were due to have their proximal TAA operation.

Patients who returned the Q1 PROM then routinely received a further letter and post-operative PROM at both 6 weeks (the letter can be seen in Appendix G1 and the questionnaire (Q2) in Appendix G2) and 3 months (the letter can be seen in Appendix H1 and the questionnaire (Q3) in Appendix H2) after their operation. Patients received both questionnaires regardless of whether they completed and returned the Q2 PROM.

If the Q1 PROM was not returned, the patient was considered to have declined the opportunity to take part in the study and the follow-up questionnaires were not sent.

5.3.3 Data collection and analysis

Mailing and response data were collected using customised Excel spreadsheets. Categorical variables are presented as absolute number and %, associations were investigated using chi-squared tests or Fisher's exact tests as appropriate. For continuous variables, firstly distributive normality was established using the Kolmogorov-Smirnov test and by visual inspect of frequency distributions. Then in the case of normally distributed variables, figures are presented as mean \pm standard deviations and comparisons are made using the Students t-test. In the case of non-normally distributed variables, figures are presented as medians (inter-quartile range) and comparisons are made using Mann Whitney U-tests.

Interpretability was measured using the technique described in Morris (2013), the precision of individual scores was calculated at the 90% confidence interval level by multiplying the standard error of measurement

(SEM) by the two-tailed z value at 90%. This approach was used as an attempt to identify potentially minimal important changes (MIC) within the PROM domains, the EQ-5D index and the EQ-VAS. These MIC calculations demonstrate the differences between both the disease specific domains and the general health measures from baseline to 6 weeks and 3 months. Differences from baseline are displayed in cumulative distribution frequency (CDF) charts.

Item response theory (IRT) (see Embretson, 2013), as described with application to health outcomes and health status measures by Hays (2000) and McHorney (2000), is a technique used to assess the precision of measurement tools such as questionnaires. The approach has overtaken what was traditionally achieved with classical test theory (CTT) methods such as item-total correlation analysis, stepwise regression and factor analysis (Coste 1997). A variation of IRT known as the Rasch model is increasingly applied as the 'gold standard' of questionnaire development, this method aims to ensure more accurate, efficient and reliable items.

The Rasch model generates a linear metric scale in logit-units, representing the construct being measured, on which both the questionnaire items and respondents persons are located (Prieto, 2003). Then, the probability of any particular item response by any particular individual is given by a logistic function of the difference between the item location and person location (for an example, see Tennant, 2007). Items and respondents are then judged for model conformity using fit statistics.

Sample size is an important consideration when undertaking psychometric evaluations, so the development of CTT, IRT or Rasch modelling falls outside of the scope of this pilot study. Recommended minimum sample sizes for relatively straightforward models begin at 100 (Linacre, 1994), but others suggest that at least 200 (Orlando, 2002) or 500 cases (Tsutakawa, 1990) are required before these types of analyses can be considered useful and informative.

Internal consistency of the proximal TAA PROM was calculated using Cronbach's alpha (see Tavakol, 2011). Cronbach's alpha measures the internal consistency of a question or scale, in practice this means that it provides an estimate that represents the degree to which a group of questions (or 'items'), within a questionnaire domain, produce similar scores or appraise the same concept.

Cronbach's alpha is presented as a number between 0 and 1. Acceptable values have been suggested to lie between 0.70 and 0.95, a low score indicates that items may have poor inter-relatedness or perhaps have been assembled in an unrelated manner (Bland and Altman, 1997). Whereas if the score is too high, it may suggest that some items are redundant as they are testing the same concept using an altered form of words.

The reproducibility of questionnaire results is assessed by carrying out a test-retest analysis. Suitable patients are given the questionnaire and asked to complete it twice within a relatively short time frame, usually no more than two weeks.

The output of a test-retest study is known as the intraclass correlation coefficient (ICC). It gives an indication of the extent to which measurements can be replicated, not only the degree of correlation but also the level of agreement between results from Phase 1 and Phase 2 responses. As with Cronbach's alpha, the values range from 0 to 1 with values closer to 1 representing a stronger reliability. ICC values less than 0.5 indicate poor reliability, between 0.5 and 0.75 moderate reliability, between 0.75 and 0.9 good reliability and greater than 0.9 excellent reliability (Koo, 2016).

Changes in the reported PROM scores between the baseline Q1 measurement and both the 6 week Q2 measurement and the 3 month Q3 measurement are presented as box and whisker charts (McGill, 1978). These charts show the median and the IQR in the 'box' and the range in the 'whiskers', the mean value is represented with a blue dot. Comparisons of scores have been made using the Wilcoxon signed rank test for continuous, non-parametric, paired results (Chan, 2003).

Statistical analyses were conducted using SAS for Windows v9.3 (SAS Institute, Cary, NC), in all cases a *P* value < 0.05 was considered statistically significant.

5.4 Results

5.4.1 Study participants and sample size

A total of 125 patients were found to be eligible to receive a proximal TAA PROM. In the pilot period 58 patients returned a completed copy of the Q1 PROM instrument, giving a response rate of 46.4%. The final number of patients who returned completed Q1, Q2 and Q3 PROM instruments and were therefore included in the main pilot analysis was 30 (response rate of 24% of original sample).

The cohort diagram in Figure 5.1 below clarifies the responses and reasons for attrition during the study period.

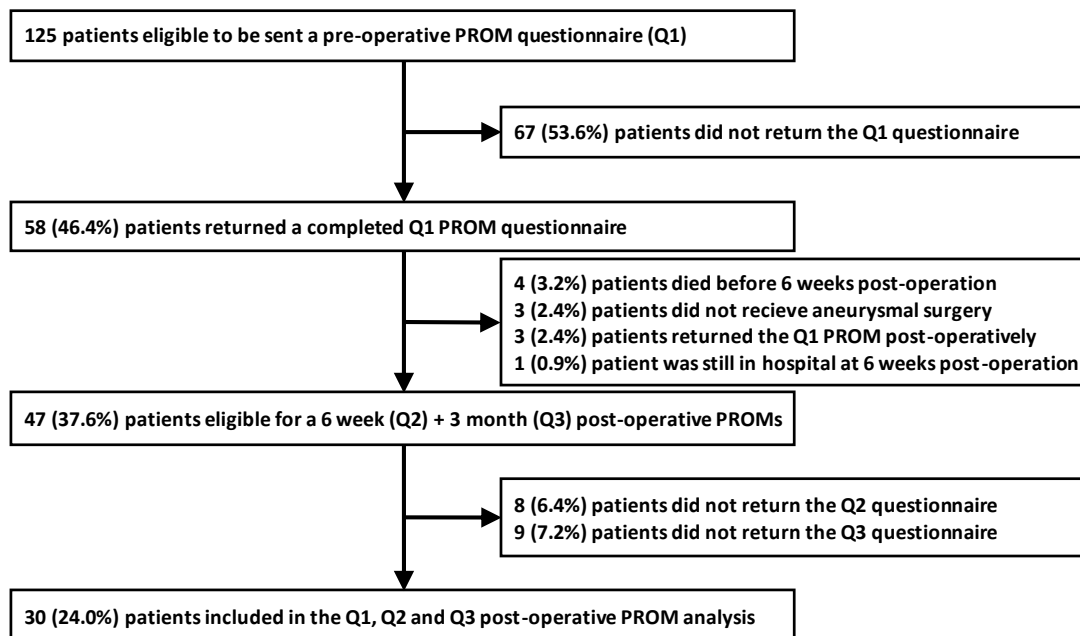


Figure 5.1: Cohort diagram showing pilot study response rates and attrition categories

5.4.2 PROM pilot patient characteristics

5.4.2.1 Respondent pre-operative characteristics (n=58)

Respondents' ages ranged from 27 to 88 years old at the time of their operation, with the majority (n = 36, 62.1%) being 60 or older. Nineteen (32.8%) of the respondents were female. Patient socio-demographic characteristics and co-morbidities are presented in Tables 5.1a and 5.1b.

In addition to the characteristics of the overall cohort, a statistical comparison was made between study responders and non-responders. It was found that patients who responded to the invitation to participate in the PROM pilot were more likely to be older (median age of 64 years (IQR = 54 to 72) vs. median age of 56 years (IQR = 43 to 67); Mann Whitney U $p = 0.01$).

Two other characteristics that may also have influenced response rates had p -values < 0.05 , although small numbers mean these findings should be treated with caution due to low statistical power. Patients who had suffered a previous myocardial infarction were more likely to respond than those who had not (6 / 58 (10.3%) vs. 1 / 67 (1.5%); Fisher's exact $p = 0.049$). Also perhaps understandably, patients who had not suffered a previous stroke were more likely to respond than those who had (2 / 58 (3.5%) vs. 9 / 67 (13.4%); Fisher's exact $p = 0.049$).

There were no other statistically significant differences between the responders and non-responders on a range of clinical and demographic variables, as shown in Tables 5.1a and 5.1b. This shows that apart from some potential age bias, the invitation to the study was reasonably acceptable to patients across a range of different presentations.

Table 5.1a: Eligible patients' socio-demographic characteristics and pre-operative co-morbidities

	All patients eligible for PROM (n = 125)	Responders (n = 58)	Non-responders (n = 67)	P value
Age at operation (years)	60 (50, 68)	64 (54, 72)	56 (43, 67)	0.01
Female gender	37 (29.6)	19 (32.8)	18 (26.9)	0.47
Body mass index (kg/m ²)	27.2 (24.5, 30.0)	27.2 (24.9, 30.0)	27.2 (24.1, 29.4)	0.92
Canadian cardiovascular society (CCS) angina grade				
Asymptomatic angina	102 (81.6)	48 (82.8)	54 (80.6)	0.76
Angina only with strenuous exertion	5 (4.0)	3 (5.2)	2 (3.0)	0.66
Angina with moderate exertion	16 (12.8)	6 (10.3)	10 (14.9)	0.44
Angina with mild exertion	2 (1.6)	1 (1.7)	1 (1.5)	>0.99
Angina at rest	0 (0)	0 (0)	0 (0)	-
New York heart association (NYHA) heart failure class				
I	50 (40.0)	25 (43.1)	25 (37.3)	0.51
II	53 (42.4)	24 (41.4)	29 (43.3)	0.83
III	22 (17.6)	9 (15.5)	13 (19.4)	0.57
IV	0 (0)	0 (0)	0 (0)	-
Previous myocardial infarction	7 (5.6)	6 (10.3)	1 (1.5)	0.049
Previous percutaneous coronary intervention (PCI)	5 (4.0)	4 (6.9)	1 (1.5)	0.18
Previous cardiac operation	13 (10.4)	6 (10.3)	7 (10.5)	0.99
Diabetes	8 (6.4)	3 (5.2)	5 (7.5)	0.72
Hypertension	51 (40.8)	26 (44.8)	25 (37.3)	0.39
Smoking status				
Previous smoker	44 (35.2)	22 (37.9)	22 (32.8)	0.55
Current smoker	13 (10.4)	4 (6.9)	9 (13.4)	0.23

Table 5.1b: Eligible patients' socio-demographic characteristics and pre-operative co-morbidities (cont.)

	All patients eligible for PROM (n = 125)	Responders (n = 58)	Non-responders (n = 67)	P value
Creatinine ($\mu\text{mol/L}$)	83 (75, 96)	81 (73, 97)	85 (77, 96)	0.35
Creatinine > 200	3 (2.4)	1 (1.7)	2 (3.0)	>0.99
Renal dysfunction	2 (1.6)	0 (0)	2 (3.0)	0.50
Respiratory disease	41 (32.8)	17 (29.3)	24 (35.8)	0.44
Previous stroke	11 (8.8)	2 (3.5)	9 (13.4)	0.049
Neurological dysfunction	2 (1.6)	0 (0)	2 (3.0)	0.50
Peripheral vascular disease	3 (2.4)	1 (1.7)	2 (3.0)	>0.99
Presenting with non-sinus heart rhythm	14 (11.2)	8 (13.8)	6 (9.0)	0.39
Coronary disease	16 (12.8)	11 (19.0)	5 (7.5)	0.055
Left ventricular ejection fraction category				
Good (> 50%)	100 (80.0)	46 (79.3)	54 (80.6)	0.86
Moderate (30 - 50%)	24 (19.2)	11 (19.0)	13 (19.4)	0.95
Poor (<30%)	2 (1.6)	1 (1.7)	0 (0)	0.46
Ventilated pre-operatively	1 (0.8)	1 (1.7)	0 (0)	0.46
Poor mobility	2 (1.6)	1 (1.7)	1 (1.5)	>0.99
Index of multiple deprivation (IMD) quintile				
1	33 (26.4)	13 (22.4)	20 (29.9)	
2	19 (15.2)	11 (19.0)	8 (11.9)	
3	23 (18.4)	13 (22.4)	10 (14.9)	
4	22 (17.6)	11 (19.0)	11 (16.4)	0.40
5	26 (20.8)	10 (17.2)	16 (23.9)	
Missing	2 (1.6)	0 (0)	2 (3.0)	
Logistic EuroSCORE	9.4 (6.9, 16.8)	9.8 (6.9, 17.8)	9.4 (6.4, 15.0)	0.39

5.4.2.2 Respondent intra-operative characteristics

Eligible patient's intra-operative characteristics are shown in Table 5.2. This data includes the types of operation carried out on the patient, including any coronary revascularisation and valvular replacements or repairs, along with information on the segments of the aorta that were operated on, the operative times and an anonymised breakdown of responsible consultant. One consultant operated on over half of the eligible patients included in the pilot study (n = 64; 51.2%), with a second consultant accounting for a further 21.6% (n = 27). The remaining 34 patients were split fairly evenly between another three consultant surgeons. This data shows that over a range of clinical procedure variables, there were no statistically significant differences between responders and non-responders.

Table 5.2: Eligible patients' intra-operative characteristics

	All patients eligible for PROM (n = 125)	Responders (n = 58)	Non-responders (n = 67)	P value
Concomitant operation				
Coronary artery bypass grafting	19 (15.2)	12 (20.7)	7 (10.5)	0.11
Valve	118 (94.4)	56 (96.6)	62 (92.5)	0.45
Aortic valve	115 (92.0)	54 (93.1)	61 (91.0)	0.75
Mitral valve	3 (2.4)	0 (0)	3 (4.5)	0.25
Other cardiac operation	6 (4.8)	4 (6.9)	2 (3.0)	0.41
Aortic Segments				
Root	79 (63.2)	35 (60.3)	44 (65.7)	0.54
Ascending	113 (90.4)	55 (94.8)	58 (86.6)	0.12
Arch	34 (27.2)	17 (29.3)	17 (25.4)	0.62
Cardiopulmonary bypass time (mins)	240 (197, 295)	228 (203, 283)	260 (195, 304)	0.32
Aortic cross clamp time (mins)	194 (155, 242)	184 (156, 217)	206 (155, 248)	0.26
Cases requiring circulatory arrest	33 (26.4)	17 (29.3)	16 (23.9)	0.49
Circulatory arrest time (mins)	38 (32, 59)	37 (31, 61)	40 (33, 55)	0.53
Consultant responsible for treatment				
Consultant A	64 (51.2)	29 (50.0)	35 (52.2)	
Consultant B	27 (21.6)	11 (19.0)	16 (23.9)	
Consultant C	12 (9.6)	6 (10.3)	6 (9.0)	0.41
Consultant D	11 (8.8)	4 (6.9)	7 (10.5)	
Consultant E	11 (8.8)	8 (13.8)	3 (4.5)	

5.4.2.3 Respondent post-operative characteristics

Eligible patient's post-operative outcomes are shown in Table 5.3. This data includes in-hospital mortality (n = 3, 2.4%, across the entire cohort), along with major morbidity (strokes, paraparesis (partial paralysis of the lower limbs), renal failure and reoperation) and lengths of stay for these patients in intensive care, post-operatively and as a whole. Again the data and the statistical analyses shows no statistically significant differences in post-operative characteristics between the responders and non-responders.

Table 5.3: Eligible patients' post-operative in-hospital outcomes

	All patients eligible for PROM (n = 125)	Responders (n = 58)	Non-responders (n = 67)	P value
In-hospital mortality	3 (2.4)	2 (3.5)	1 (1.5)	0.60
Permanent stroke	2 (1.6)	1 (1.7)	1 (1.5)	>0.99
Transient stroke	1 (0.8)	0 (0)	1 (1.5)	>0.99
Paraparesis	1 (0.8)	1 (1.7)	0 (0)	0.46
Return to theatre for reoperation	9 (7.2)	4 (6.9)	5 (7.5)	>0.99
Acute renal failure	2 (1.6)	0 (0)	2 (3.0)	>0.99
Deep sternal wound infection	0 (0)	0 (0)	0 (0)	-
Intensive care LOS (days)	3 (2, 6)	3 (2, 7)	3 (2, 6)	0.19
Post-operative LOS (days)	8 (6, 13)	8 (6, 13)	8 (5, 13)	0.99
Total hospital LOS (days)	10 (7, 15)	10 (7, 15)	10 (7, 16)	0.78

5.4.2.4 Analytical cohort characteristics

In order to establish the suitability of the analytical cohort (i.e. the 30 participants who completed the PROM at all 3 time periods, see Figure 5.1) to accurately represent the characteristics of the proximal TAA patient population as a whole, it was necessary to repeat the pre-, intra- and post-operative analysis, this time comparing those patients in the analytical cohort (n = 30) with the remaining eligible patients who did not respond (n=95). The data on demographic and clinical differences is presented below in Tables 5.4a, 5.4b, 5.5 and 5.6. The 30 Q3 PROM responders did appear older, and a higher proportion were female compared to non-responders, but this was not statistically significant.

There were no statistically significant differences between the patients in the analytical cohort and the other eligible patients identified, suggesting that there was no identifiable bias involved and that the results in the analytical cohort (n=30) could be reasonably extrapolated to the wider patient population (n=125). Nevertheless, the small sample size should be noted and any findings should be treated with a degree of caution.

Table 5.4a: Socio-demographic characteristics and pre-operative co-morbidities, stratified by analytical cohort

	Patients in analytical cohort (n = 30)	Other eligible patients (n = 95)	P value
Age at operation (years)	66 (53, 71)	58 (48, 67)	0.09
Female gender	12 (40.0)	25 (26.3)	0.15
Body mass index (kg/m ²)	26.5 (24.6, 29.0)	27.4 (24.4, 30.6)	0.55
Canadian cardiovascular society (CCS) angina grade			
Asymptomatic angina	23 (76.7)	79 (83.2)	0.42
Angina only with strenuous exertion	1 (3.3)	4 (4.2)	>0.99
Angina with moderate exertion	5 (16.7)	11 (11.6)	0.53
Angina with mild exertion	1 (3.3)	1 (1.1)	0.42
Angina at rest	0 (0)	0 (0)	-
New York heart association (NYHA) heart failure class			
I	13 (43.3)	37 (39.0)	0.67
II	10 (33.3)	43 (45.3)	0.25
III	15 (15.8)	7 (23.3)	0.34
IV	0 (0)	0 (0)	-
Previous myocardial infarction	1 (3.3)	6 (6.3)	>0.99
Previous percutaneous coronary intervention (PCI)	1 (3.3)	4 (4.2)	>0.99
Previous cardiac operation	2 (6.7)	11 (11.6)	0.73
Diabetes	2 (6.7)	6 (6.3)	>0.99
Hypertension	12 (40.0)	39 (41.1)	0.92
Smoking status			
Previous smoker	11 (36.7)	33 (34.7)	0.85
Current smoker	1 (3.3)	12 (12.6)	0.19

Table 5.4b: Socio-demographic characteristics and pre-operative co-morbidities, stratified by analytical cohort (cont.)

	Patients in analytical cohort (n = 30)	Other eligible patients (n = 95)	P value
Creatinine (µmol / L)	81 (74, 92)	83 (75, 97)	0.78
Creatinine > 200	1 (3.3)	2 (2.1)	0.56
Renal dysfunction	0 (0)	2 (2.1)	>0.99
Respiratory disease	9 (30.0)	32 (33.7)	0.71
Previous stroke	1 (3.3)	10 (10.5)	0.46
Neurological dysfunction	0 (0)	2 (2.1)	>0.99
Peripheral vascular disease	0 (0)	3 (3.2)	>0.99
Presenting with non-sinus heart rhythm	1 (3.3)	13 (13.7)	0.18
Coronary disease	4 (13.3)	12 (12.6)	>0.99
Left ventricular ejection fraction category			
Good (> 50%)	24 (80.0)	76 (80.0)	>0.99
Moderate (30 - 50%)	6 (20.0)	18 (19.0)	0.90
Poor (<30%)	0 (0)	1 (1.1)	>0.99
Ventilated pre-operatively	1 (3.3)	0 (0)	0.24
Poor mobility	1 (3.3)	1 (1.1)	0.42
Index of multiple deprivation (IMD) quintile			
1	9 (30.0)	24 (25.3)	
2	5 (16.7)	14 (14.7)	
3	7 (23.3)	16 (16.8)	
4	5 (16.7)	17 (17.9)	0.78
5	4 (13.3)	22 (23.2)	
Missing	0 (0)	2 (2.1)	
Logistic EuroSCORE	9.6 (6.8, 17.4)	9.4 (6.9, 16.5)	0.79

Table 5.5: Intra-operative characteristics, stratified by analytical cohort

	Patients in analytical cohort (n = 30)	Other eligible patients (n = 95)	P value
Concomitant operation			
Coronary artery bypass grafting	6 (20.0)	13 (13.7)	0.39
Valve	28 (93.3)	90 (94.7)	0.67
Aortic valve	27 (90.0)	88 (92.6)	0.70
Mitral valve	0 (0)	3 (3.2)	>0.99
Other cardiac operation	2 (6.7)	4 (4.2)	0.63
Aortic Segments			
Root	17 (56.7)	62 (65.3)	0.39
Ascending	27 (90.0)	86 (90.5)	>0.99
Arch	8 (26.7)	26 (27.4)	0.94
Cardiopulmonary bypass time (mins)	226 (197, 245)	250 (199, 304)	0.21
Aortic cross clamp time (mins)	178 (161, 210)	200 (155, 248)	0.16
Cases requiring circulatory arrest	8 (26.7)	25 (26.3)	0.97
Circulatory arrest time (mins)	35 (30, 41)	39 (33, 61)	0.21
Consultant responsible for treatment			
Consultant A	11 (36.7)	53 (55.8)	
Consultant B	9 (30.0)	18 (19.0)	
Consultant C	2 (6.7)	10 (10.5)	0.06
Consultant D	2 (6.7)	9 (9.5)	
Consultant E	6 (20.0)	5 (5.3)	

Table 5.6: Post-operative in-hospital outcomes, stratified by analytical cohort

	Patients in analytical cohort (n = 30)	Other eligible patients (n = 95)	P value
In-hospital mortality	0 (0)	3 (3.2)	>0.99
Permanent stroke	0 (0)	2 (2.1)	>0.99
Transient stroke	0 (0)	1 (1.1)	>0.99
Paraparesis	0 (0)	1 (1.1)	>0.99
Return to theatre for reoperation	2 (6.7)	7 (7.4)	>0.99
Acute renal failure	0 (0)	2 (2.1)	>0.99
Deep sternal wound infection	0 (0)	0 (0)	-
Intensive care LOS (days)	3 (2, 6)	3 (2, 6)	0.66
Post-operative LOS (days)	8 (6, 10)	8 (6, 14)	0.37
Total hospital LOS (days)	8 (7, 12)	10.5 (7, 16)	0.23

5.4.3 Acceptability

5.4.3.1 Response rates

A total of 125 Q1 PROM questionnaires were posted out to patients awaiting proximal TAA surgery. Of those, 58 (46.4%) were initially returned and usable. Of those patients who returned Q1, 30 were also able to return Q2 and Q3 giving an internal overall response rate of 63.8%. Looked at in the wider context of the overall eligible patient population, the analytical cohort of 30 / 125 produces a response rate of 24%.

Rates of response and attrition in the four original treatments which were allocated a nationally mandated PROM were published in a “Special Topic” document in 2016 (Health and Social Care Information Centre, 2016), data from the report is included in Figure 5.2:

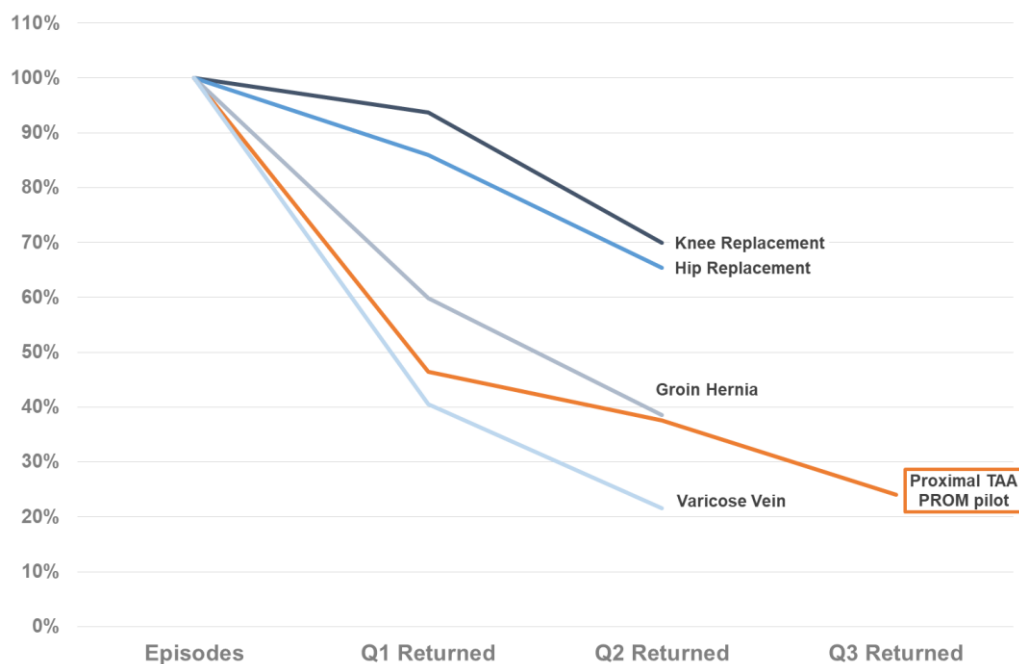


Figure 5.2: Attrition rate: Percentage of participants at each stage during the proximal TAA PROM pilot, including a comparison of national PROMs 2013/14 data (Contains information from NHS Digital, licenced under the current version of the Open Government Licence).

Figure 5.2 shows the Q1 response rate along with the attrition rates at Q2 and Q3 for the proximal TAA PROM pilot. Even though the delivery strategy for the current study included an additional post-operative PROM delivery, the pattern of response falls between the groin hernia PROM and varicose vein PROM response configuration for 2013/14.

5.4.3.2 Missing data

In the analytical group of 30 patients, the rates of missing data per questionnaire item were small in most cases. For the Q1 questionnaire, all items had a missing data rate < 10% (n missing < 3). For the Q2 questionnaire, 6 items had a rate of missing data > 10% (3 items had 13.3% missing data (n missing = 4) and 3 had 16.7% (n missing = 5) missing data). For the Q3 questionnaire, no items had a rate of missing data > 10%.

Overall, across the three Q1, Q2 and Q3 PROM questionnaires, 191 key items were identified. Multiplied by 30, that results in 5730 individual items of data. Out of these there were 123 items of data missing in the analytical cohort dataset, or 2.1%.

5.4.4 Feasibility

5.4.4.1 Respondent feasibility

The reasonable response rates reported above go some way towards demonstrating the feasibility of the PROM pilot in this patient population. One of the unique elements of this study was the requirement for patients to respond at both 6 weeks and 3 months post-operatively. This makes it difficult to compare response rates with previous PROM programmes, which typically included a single follow-up survey. However, the reality is that the response and attrition rates appear to be at the lower end of what was experienced in the national programme, although strategies to improve this –

such as exploring how feasible it would be to deliver the Q1 PROM at a pre-operative clinic, or testing Q2 and Q3 deliveries at different post-operative time points – could be adopted.

5.4.4.2 Administrative staff feasibility

The PROM pilot was administered by a single investigator in addition to their usual employment tasks. Document printing, patient identification, mailing and data input were all manageable with the rate of eligible proximal TAA patients admitted to a single aortic surgery provider. If the numbers of eligible patients were to increase, or the delivery of the PROM was to change in any significant way, the administrative burden may become more challenging.

5.4.5 Interpretability

Interpretability was measured using the following CDF charts, allowing the MIC₉₀ thresholds to be identified.

Figures 5.3a and 5.3b show the MIC₉₀ for the Symptoms domain at 6 weeks and 3 months, respectively. At 6 weeks the MIC₉₀ was 5.4 and at 3 months it was 8.7

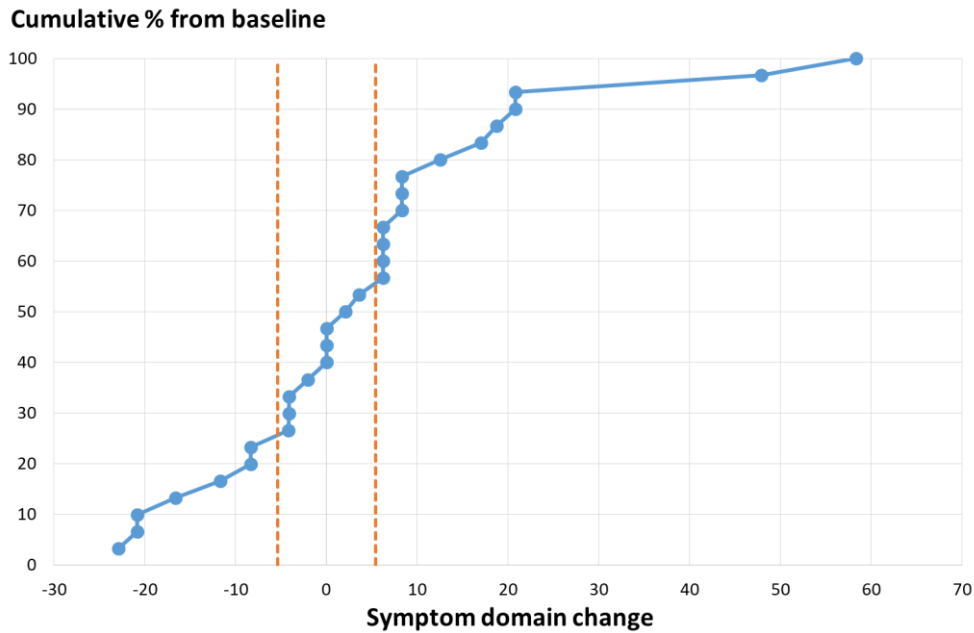


Figure 5.3a: CDF chart showing the MIC₉₀ (dotted orange lines) for the Symptoms domain at 6 weeks

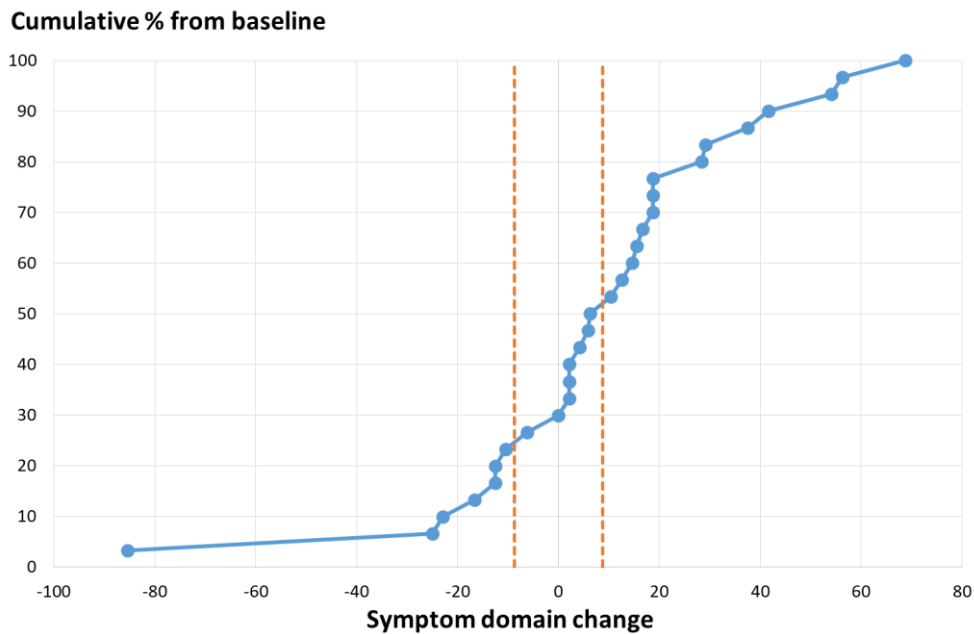


Figure 5.3b: CDF chart showing the MIC₉₀ (dotted orange lines) for the Symptoms domain at 3 months

Figures 5.4a and 5.4b show the MIC₉₀ for the Physical domain at 6 weeks and 3 months, respectively. At 6 weeks the MIC₉₀ was 5.9 and at 3 months it was 8.1

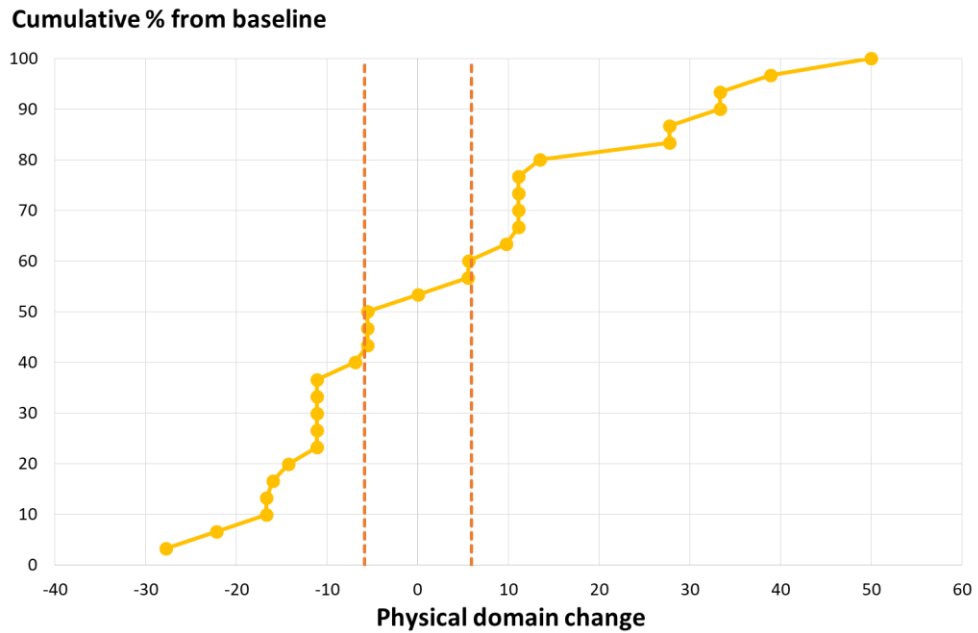


Figure 5.4a: CDF chart showing the MIC₉₀ (dotted orange lines) for the Physical domain at 6 weeks

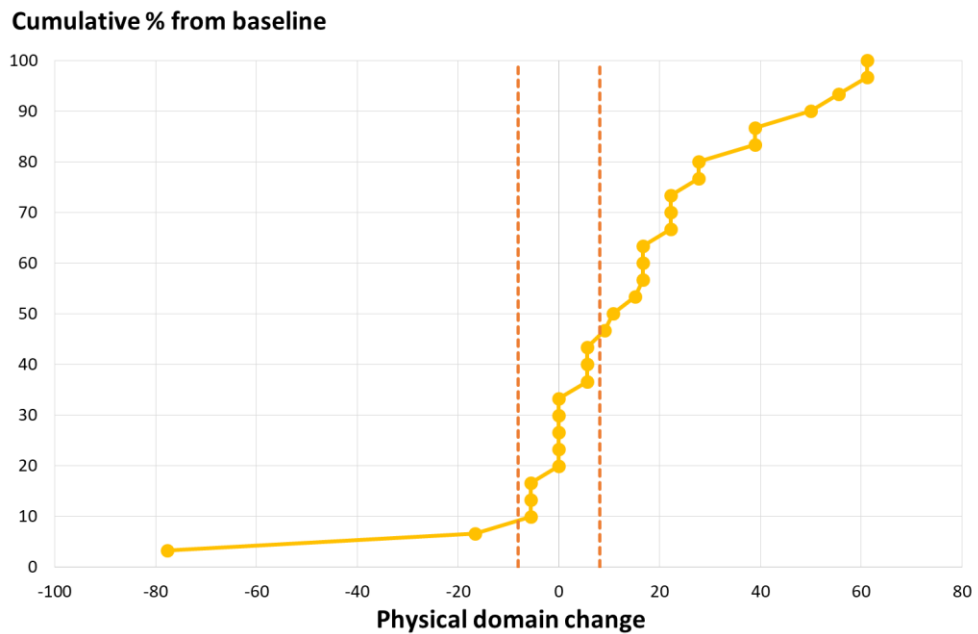


Figure 5.4b: CDF chart showing the MIC₉₀ (dotted orange lines) for the Physical domain at 3 months

Figures 5.5a and 5.5b show the MIC₉₀ for the Psychosocial domain at 6 weeks and 3 months, respectively. At 6 weeks the MIC₉₀ was 5.3 and at 3 months it was 6.2

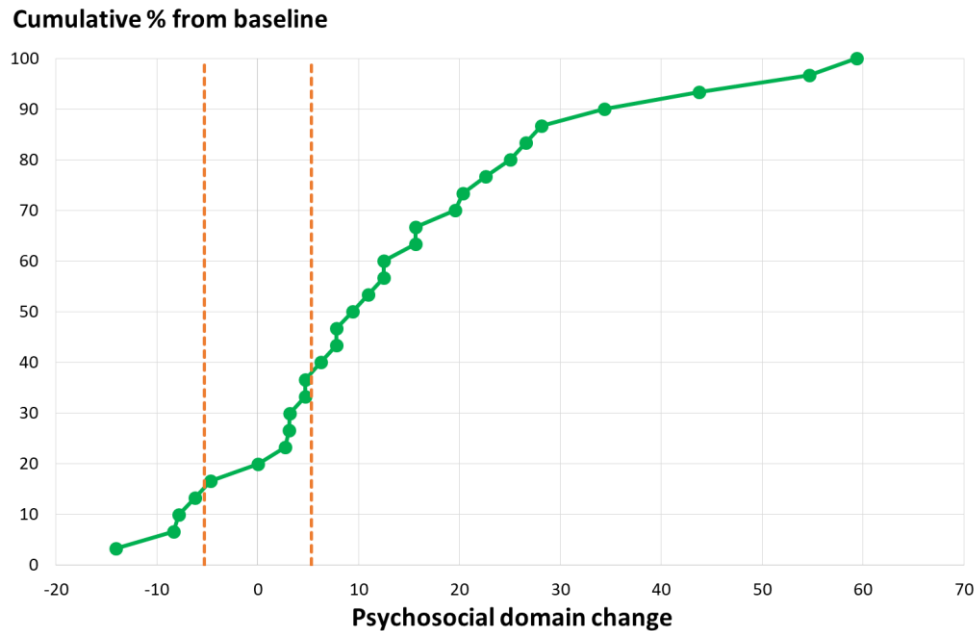


Figure 5.5a: CDF chart showing the MIC₉₀ (dotted orange lines) for the Psychosocial domain at 6 weeks

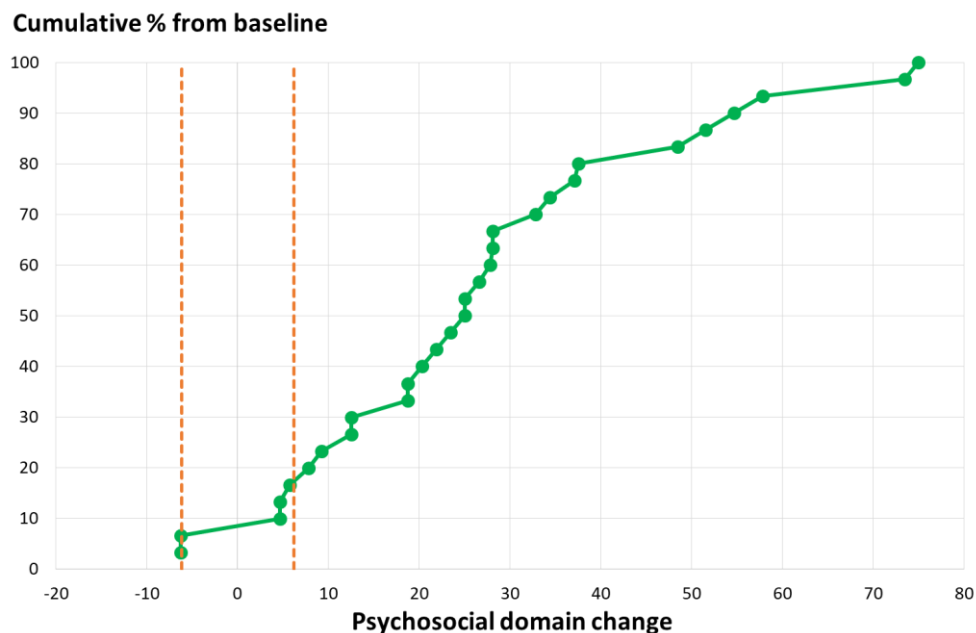


Figure 5.5b: CDF chart showing the MIC₉₀ (dotted orange lines) for the Psychosocial domain at 3 months

Figures 5.6a and 5.6b show the MIC₉₀ for the Cognitive domain at 6 weeks and 3 months, respectively. At 6 weeks the MIC₉₀ was 7.4 and at 3 months it was 9.4

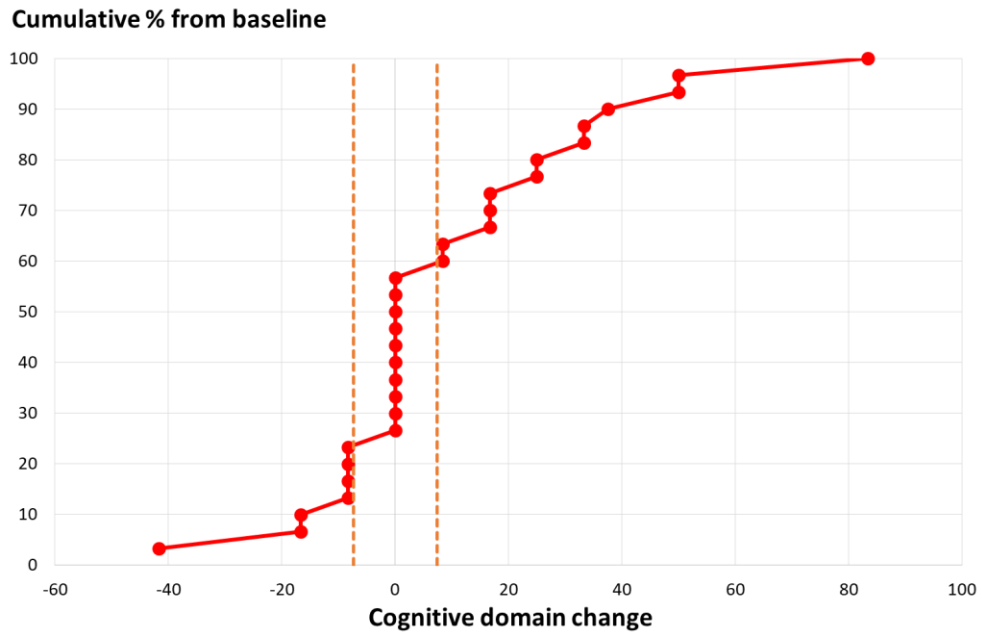


Figure 5.6a: CDF chart showing the MIC₉₀ (dotted orange lines) for the Cognitive domain at 6 weeks

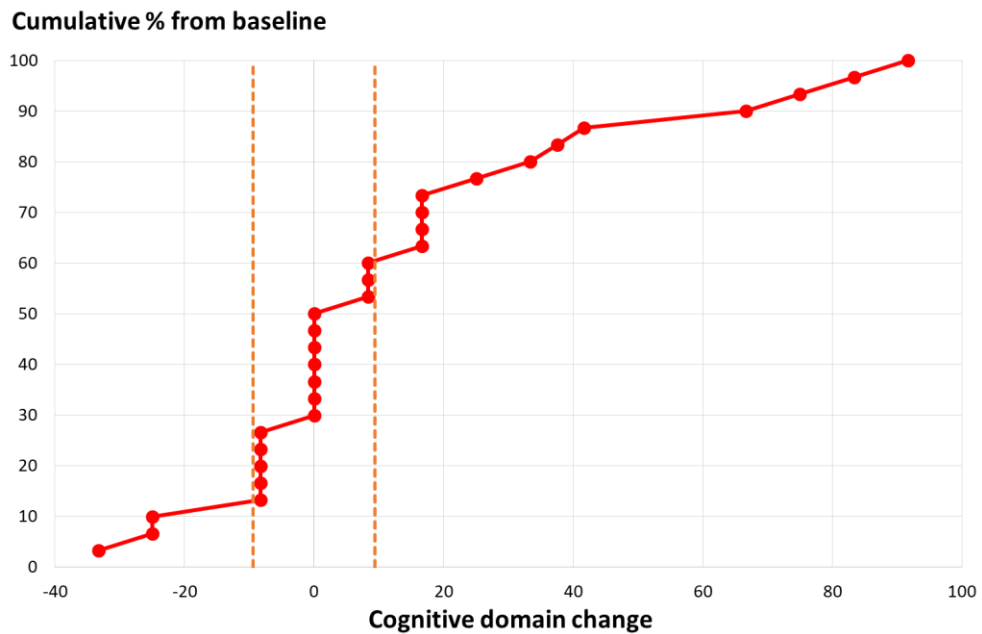


Figure 5.6b: CDF chart showing the MIC₉₀ (dotted orange lines) for the Cognitive domain at 3 months

Figures 5.7a and 5.7b show the MIC₉₀ for the EQ-5D Index at 6 weeks and 3 months, respectively. At 6 weeks the MIC₉₀ was 0.09 and at 3 months it was 0.11

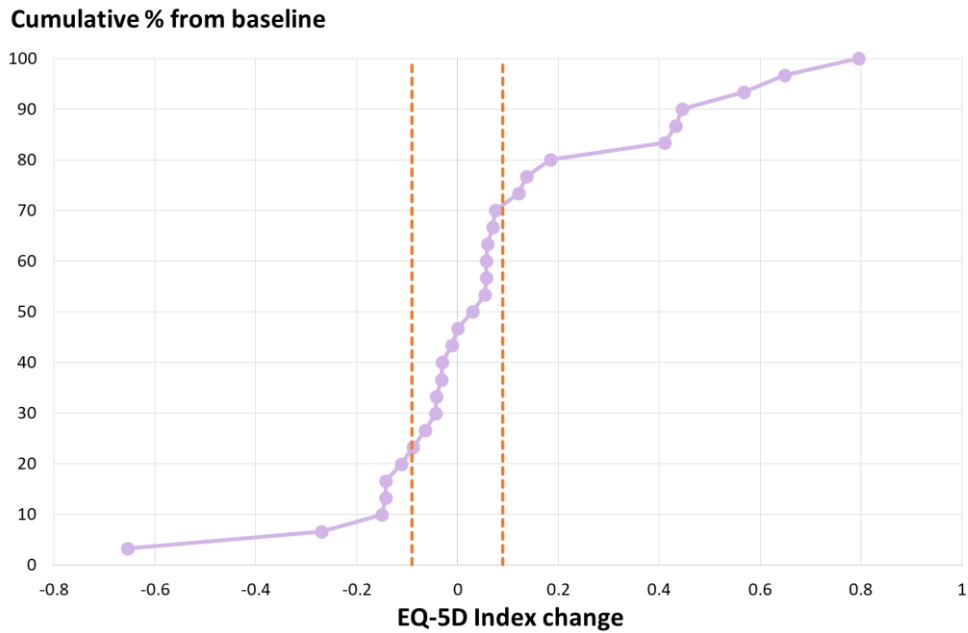


Figure 5.7a: CDF chart showing the MIC₉₀ (dotted orange lines) for the EQ-5D Index domain at 6 weeks

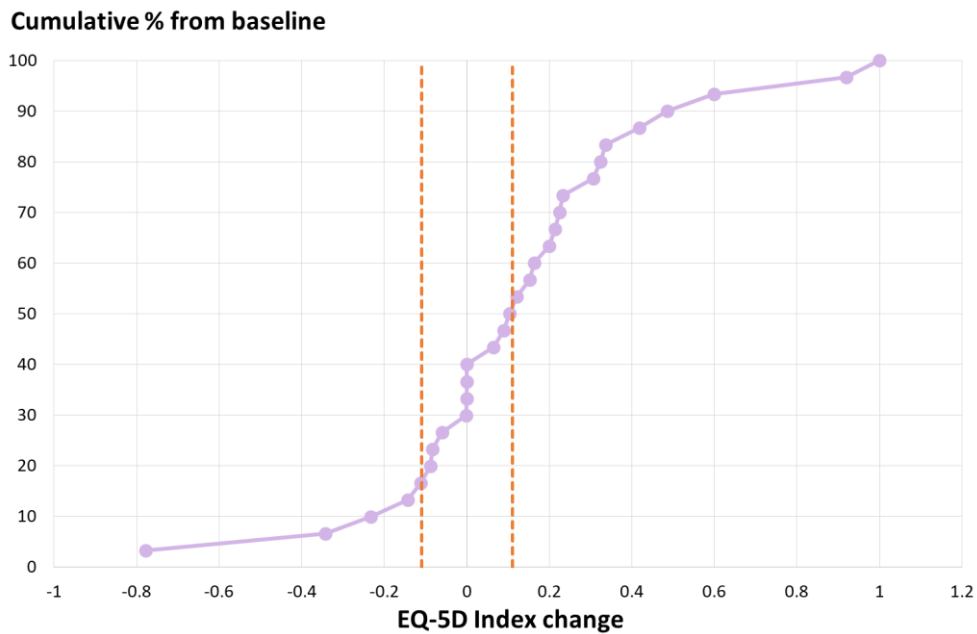


Figure 5.7b: CDF chart showing the MIC₉₀ (dotted orange lines) for the EQ-5D Index domain at 3 months

Figures 5.8a and 5.8b show the MIC₉₀ for the EQ-VAS domain at 6 weeks and 3 months, respectively. At 6 weeks the MIC₉₀ was 7.4 and at 3 months it was 8.1

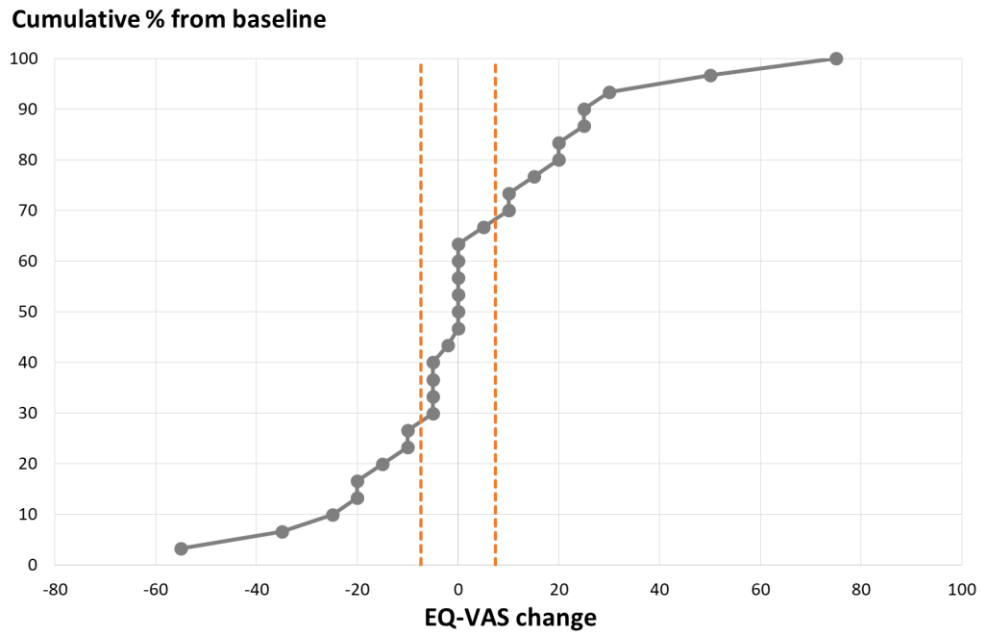


Figure 5.8a: CDF chart showing the MIC₉₀ (dotted orange lines) for the EQ-VAS domain at 6 weeks

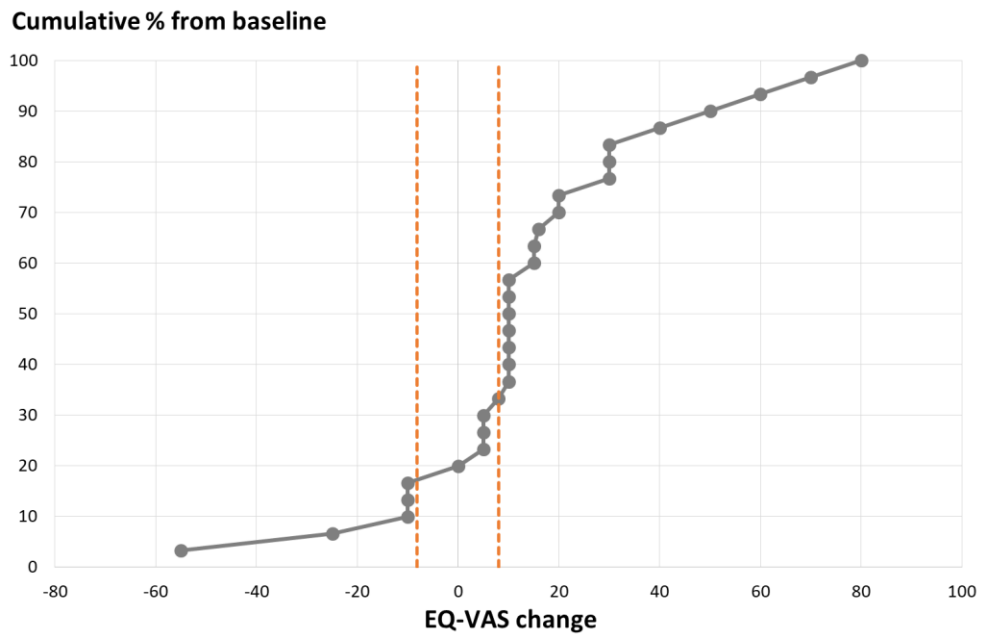


Figure 5.8b: CDF chart showing the MIC₉₀ (dotted orange lines) for the EQ-VAS domain at 3 months

5.4.6 Reliability

5.4.6.1 Internal consistency

Table 5.7 shows the Cronbach's alpha scores for the four PROM domains, based on the baseline Q1 responses.

Table 5.7: Cronbach's alpha scores for PROM domains

	Symptoms	Physical	Psychosocial	Cognitive
Item 1	0.896	0.933	0.955	0.811
Item 2	0.907	0.935	0.952	0.824
Item 3	0.900	0.934	0.952	0.759
Item 4	0.902	0.934	0.951	
Item 5	0.909	0.941	0.956	
Item 6	0.912	0.939	0.950	
Item 7	0.899	0.933	0.950	
Item 8	0.901	0.950	0.957	
Item 9	0.900	0.952	0.950	
Item 10	0.907		0.952	
Item 11	0.911		0.954	
Item 12	0.902		0.951	
Item 13			0.952	
Item 14			0.949	
Item 15			0.948	
Item 16			0.950	
Overall	0.911	0.946	0.955	0.855

In the main, the alpha scores show a very high level of internal consistency as they are all >0.70. Although some perhaps demonstrate that there are redundant items present, as it has been suggested that alpha scores >0.90 indicate questions that are testing the same response only in a different

guise (Streiner, 2003). This is especially true in the Psychosocial domain. Future work would need to repeat the analysis with a larger cohort in order to confirm these results.

5.4.6.2 Test-retest reliability

60 patients independent of the main analytical cohort were invited to take part in the test-retest study, 29 (48.3%) patients responded to both questionnaires and were included in the analysis. Figure 5.9 shows the cohort diagram, clarifying the response rates and reasons for attrition in the test-retest study.

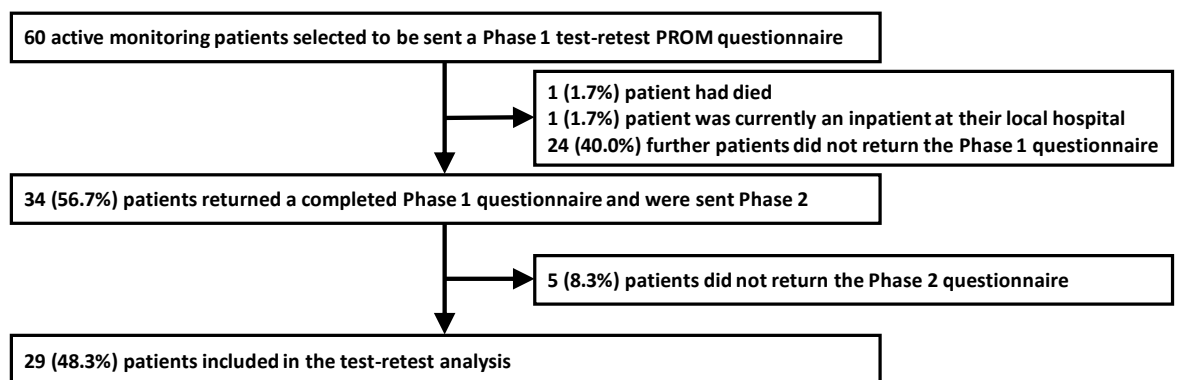


Figure 5.9: Cohort diagram showing test-retest response rates and attrition categories

Table 5.8 shows the test-retest ICC values for the four PROM domains.

Table 5.8: Test-retest ICC scores for PROM domains

	Symptoms	Physical	Psychosocial	Cognitive
Item 1	1.000	0.907	0.910	0.785
Item 2	0.840	0.960	0.696	0.934
Item 3	0.966	0.874	0.974	0.856
Item 4	0.790	0.721	0.899	
Item 5	0.726	0.711	0.747	
Item 6	0.573	0.922	0.935	
Item 7	0.956	0.811	0.931	
Item 8	0.937	0.841	0.890	
Item 9	0.801	1.000	0.628	
Item 10	0.973		0.812	
Item 11	0.875		0.857	
Item 12	0.671		0.609	
Item 13			0.838	
Item 14			0.664	
Item 15			0.799	
Item 16			0.926	
Overall	0.919	0.910	0.875	0.923

Most items and all domains show a good or excellent ICC result, indicating useful reproducibility and agreement. Item 6 in the Symptom domain (“Feeling unsteady or uncoordinated”) is notably low however, suggesting only a moderate degree of measurement agreement with other items in that domain (Koo, 2016). Further work with greater sample sizes would be required to confirm that finding.

5.4.7 Responsiveness

Box and whisker charts showing the responsiveness in the reported PROM scores between the baseline Q1 measurement and both the 6 week Q2 measurement and the 3 month Q3 measurement are presented below.

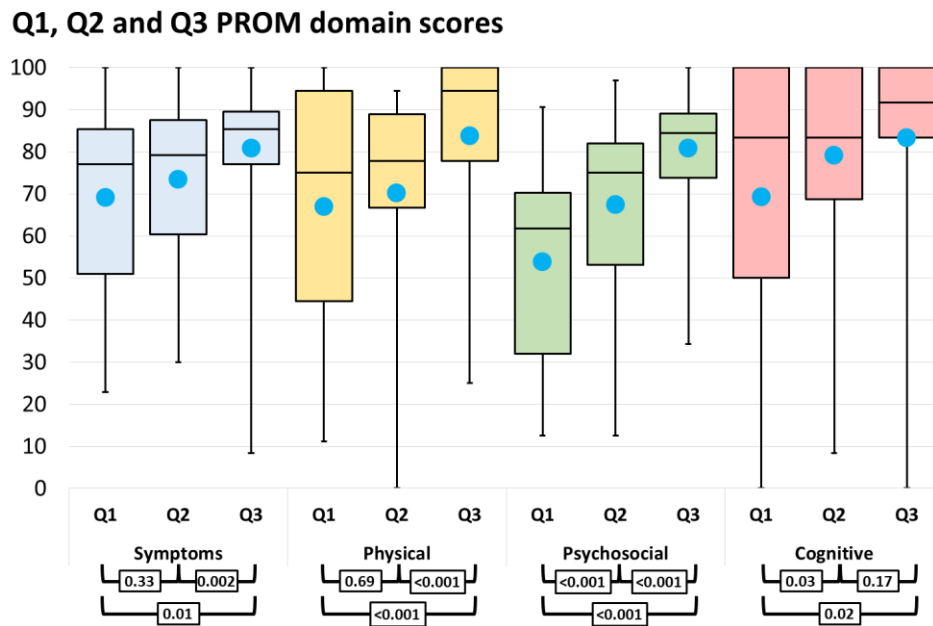


Figure 5.10: Box plot showing PROM domain scores and statistical comparisons at Q1, Q2 and Q3

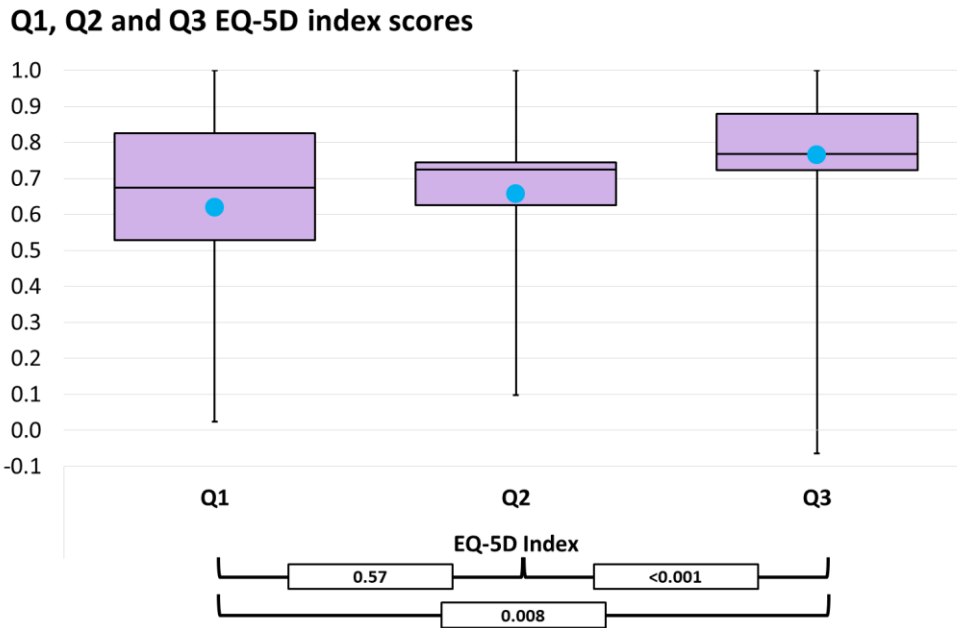


Figure 5.11: Box plot showing EQ-5D index scores and statistical comparisons at Q1, Q2 and Q3

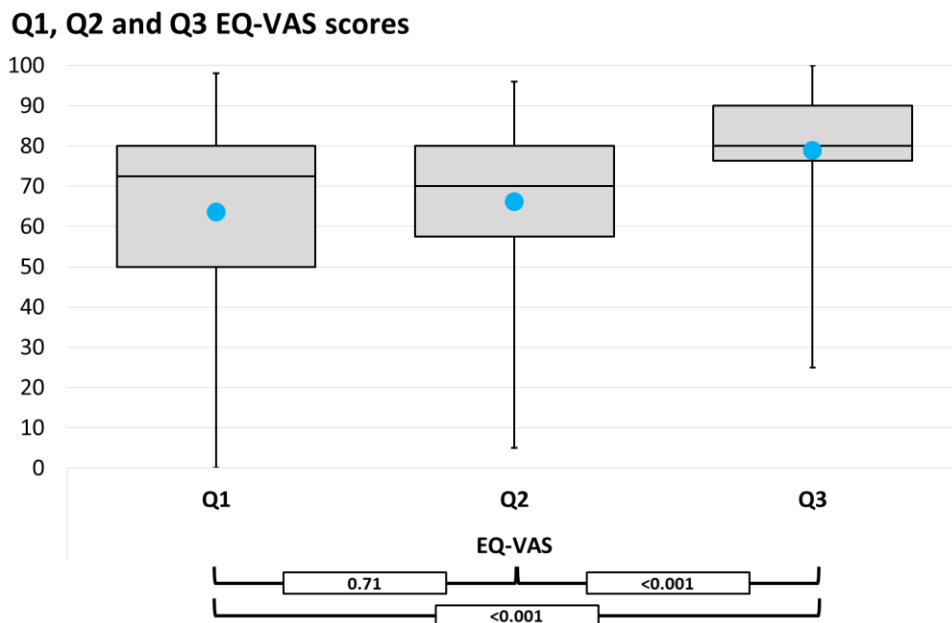


Figure 5.12: Box plot showing EQ-VAS scores and statistical comparisons at Q1, Q2 and Q3

5.4.7.1 Alternative presentation of results

Perhaps a more relevant way of presenting the data is by plotting the differences between baseline Q1 scores and scores at Q2 and Q3. The following charts retain the box plot presentation but report the values as differences from Q1.

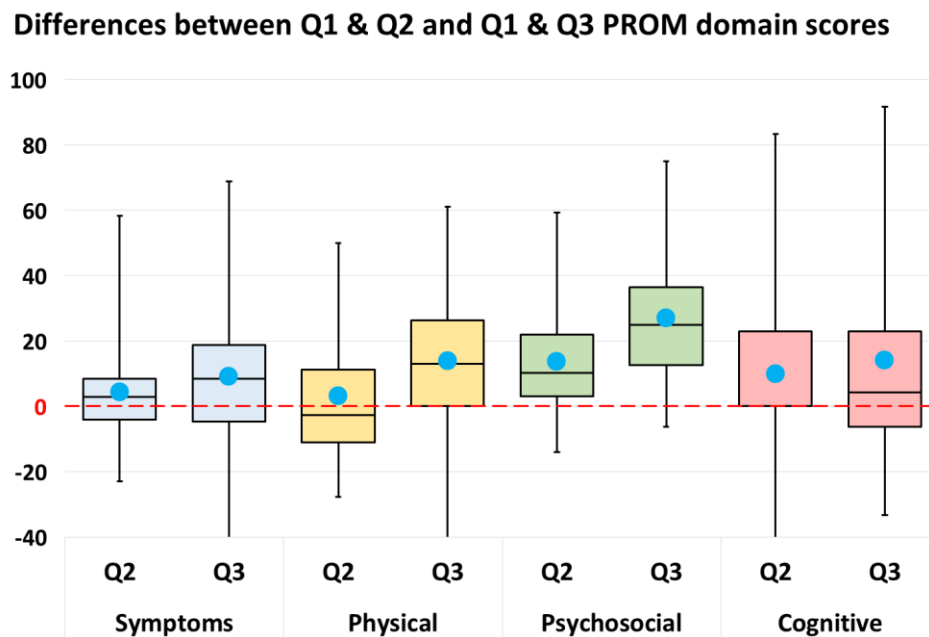


Figure 5.13: Box plot showing PROM domain score differences from Q1 at Q2 and Q3

Differences between Q1 & Q2 and Q1 & Q3 EQ-5D index scores

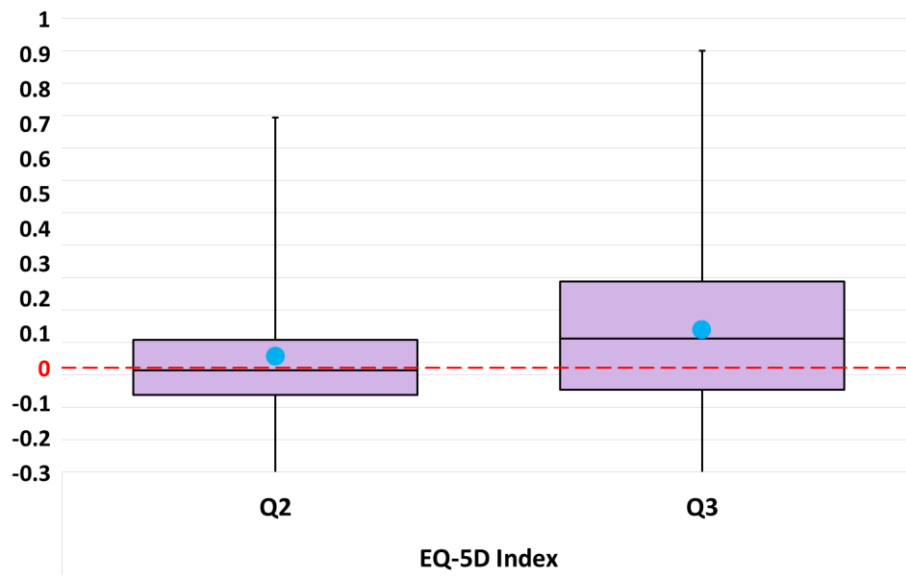


Figure 5.14: Box plot showing EQ-5D Index score differences from Q1 at Q2 and Q3

Differences between Q1 & Q2 and Q1 & Q3 EQ-VAS scores

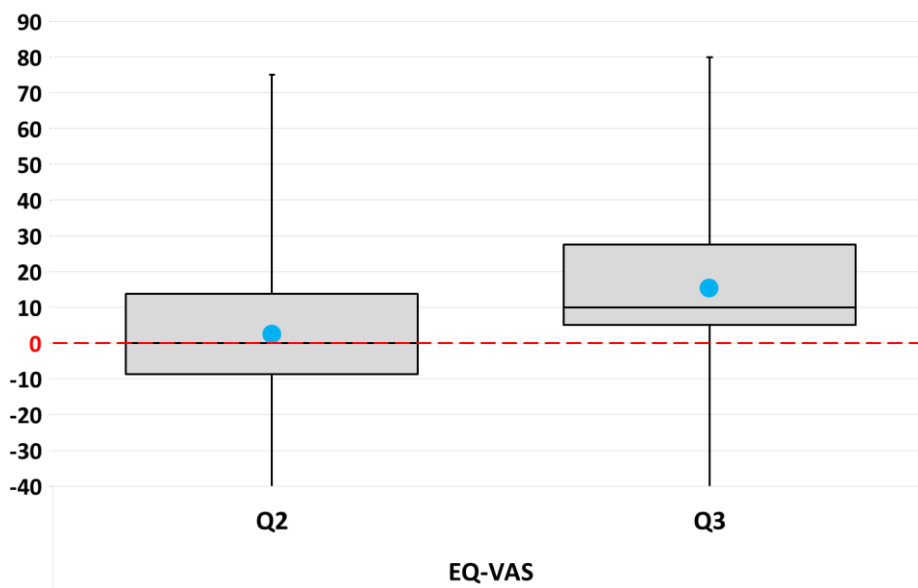


Figure 5.15: Box plot showing EQ-VAS score differences from Q1 at Q2 and Q3

For the NHS PROM program (see <https://digital.nhs.uk/data-and-information/data-tools-and-services/data-services/patient-reported-outcome-measures-proms>), the results have been presented in the following way, which simplifies the output into patients who reported HRQoL improvements, patients who reported a deterioration in HRQoL and patients who stayed the same. The first chart analysis specified that any increase or decrease in domain or EQ-5D score indicated an improvement or deterioration, the second utilised the MIC₉₀ boundaries described in Section 5.3.3.

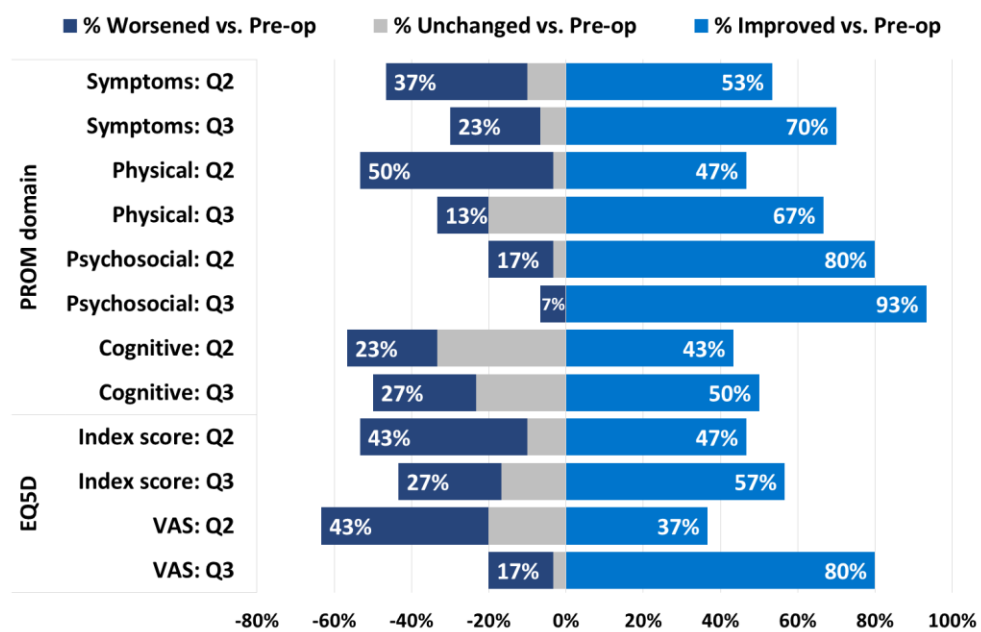


Figure 5.16: PROM results, NHS format (all differences considered relevant)

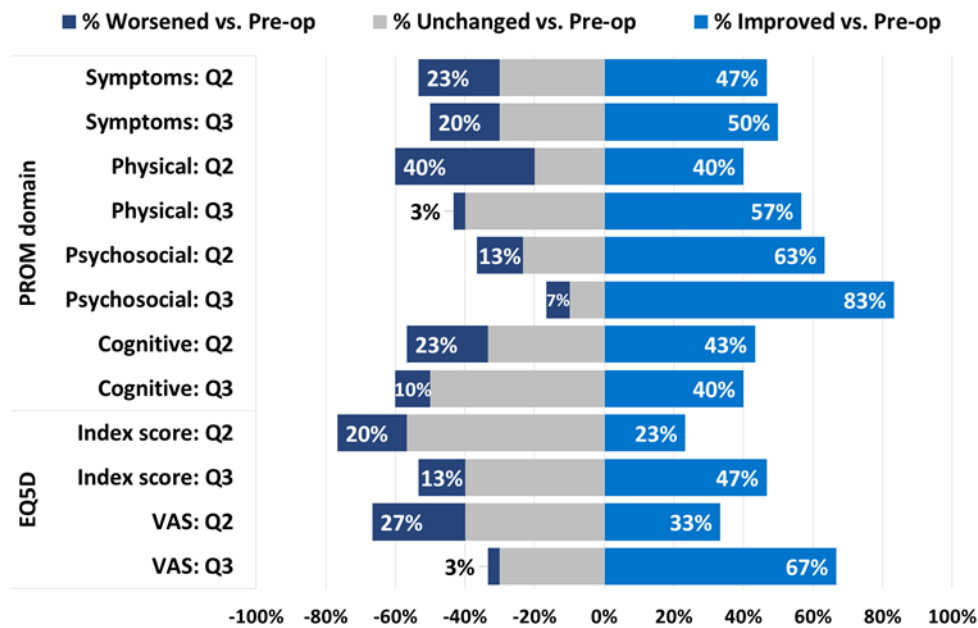


Figure 5.17: PROM results, NHS format (MIC₉₀ differences considered relevant)

5.5 Summary

In summary, the quantitative findings presented in in this chapter suggest that the new proximal TAA PROM tool could be a practical and scientifically valid measure for patient based HRQoL reporting. Although the sample size in the pilot was relatively small, the results show the PROM instrument returning reliable, interpretable and responsive data in this patient population.

The questionnaire is broadly acceptable to patients. Although response rates in this pilot did not reach those seen in the national Knee and Hip Replacement PROM studies, they are comparable to those reported in both Groin Hernia and Varicose Vein procedures – that comparable response rate occurred despite the inclusion of an additional follow-up questionnaire.

Reliability was shown through the high level of internal consistency (with Cronbach’s alpha results for the PROM domains ranging from 0.855 to 0.955), and result replication (with ICC results for the PROM domains

ranging from 0.875 to 0.923). These high levels of reliability compare well with those reported in the CROQ validation (Schroter, 2004), where every domain had a Cronbach's alpha score and an ICC score > 0.80 (item level results were not published).

Interpretability was demonstrated by estimating the minimally important clinical differences at the 90% level, for each domain at both 6 weeks and 3 months. This approach facilitates a more inferential analysis by clinical staff, and perhaps helps to identify subgroups of patients who would benefit from additional resources – for example surgical prehabilitation or psychological counselling.

Responsiveness was shown using three different approaches:

- The first approach (Figures 5.10, 5.11 and 5.12) uses box and whisker plots to show the range of results and any statistically significant differences between the baseline PROM score and the follow-up scores at 6 weeks and 3 months.
- The second approach (Figures 5.13, 5.14 and 5.15) again uses box and whisker plots, but the unit of measurement in this analysis is the difference between the two sets of follow-up scores and the baseline score. Therefore only the differences at 6 weeks and 3 months need to be shown.
- The third approach (Figures 5.16 and 5.17) is the one currently used by the NHS, which is a simplified view on a single scale. The unit of measurement is again the differences between results at 6 weeks and 3 months when compared to the baseline.

These different approaches each have strengths and weaknesses, and the intention was to give a range of visualisations that may find favour with different audiences. The first approach may appeal to academics or clinicians, as it uses p-values and attempts to paint a more complete picture of results. However it may not be suitable for the layperson as p-values and

box and whisker plots would require specialist knowledge to interpret. The second approach is a simplification of the first, concentrating on the follow-up results as compared to baseline. This removes the p-values and includes a “0” line to indicate the pre-operative scores, which is perhaps more intuitive. However, the box and whisker plots still require some specialist knowledge to decipher upon first glance. The third approach is the most simple, and has the appeal of presenting all the results on a single chart. It is similar to the second approach in that it shows the differences from baseline rather than all the scores, but this shows a percentage difference rather than using the units of measurement which are particular to each domain. Deciding on the appropriate way to present PROM results for particular audiences could be explored in future studies.

To conclude, these results give encouragement that the TAA PROM tool is a useful instrument in this population. Furthermore, they offer some initial, novel evidence that the majority of patients may experience an improvement in their physical and psychosocial HRQoL at 3 months post-operatively, based on pilot data MIC₉₀ calculations. However, further quantitative analysis with a larger sample size and including Rasch modelling (see Section 5.3.3) and psychometric work will be needed to give robust confirmation of the precision and validity of the instrument. Also, careful evaluation of population and procedural differences would have to be made before this PROM was used in other settings.

Chapter 6

Discussion and conclusions

6.1 Introduction

The following final thesis chapter presents a summary of the research findings. The implications and limitations of the work are discussed, along with the scope and generalisability of the findings. Finally, possible directions for future research projects are outlined.

6.2 Overview of findings

This thesis began by reviewing the history, development and contemporary understanding of aortic aneurysms, HRQoL and PROMs (Chapter 2). This review provided evidence of a knowledge gap around suitable PROM tools that could be used for patients being surgically treated for proximal TAA. Several areas for improvement were identified, including the lack of a robust conceptual model or theoretical framework that reflects these patients' lived experience of TAA, a derived item set of suitable questions or a feasible, evaluated PROM tool which could be routinely used to measure HRQoL in these individuals.

The most appropriate methodology for answering these questions was presented in Chapter 3. This included a summary of the underlying pragmatic philosophical approach, a discussion of PROM development techniques and a rationale for including both qualitative and quantitative results in an exploratory sequential mixed methods research strategy.

Chapter 4 included the design, analysis and results of the qualitative phase of the study. This comprised a short appraisal of the current literature regarding qualitative investigations into this patient population, then a

discussion of the recruitment strategy, descriptive statistics for the study participants and a justification of sample size. The interview and data collection plans were also described, along with an exploration of Thematic Analysis – the chosen approach to qualitative data investigation. A detailed presentation of the key results from the qualitative analysis was then included which provided in-depth information about patients' thoughts and feelings concerning their experiences with TAA and the healthcare services, these themes were then collected into the established physical, psychological and social HRQoL domains along with the emerging concepts of how TAA affects HRQoL: the effect of disease and treatment, attitudes of and towards friends and family and the continued diagnostic monitoring for aneurysmal disease. These were then summarised in a conceptual model, showing the interconnectedness of TAA disease, diagnosis, treatment and HRQoL. Item identification, questionnaire construction and findings from the focus group evaluation were also presented.

The results of the TAA PROM pilot were shown in Chapter 5. These included the approach to recruitment, delivery and data collection along with descriptions of how representative of the overall population the respondent cohort was. Response rates and missing data values were also presented. The standard PROM evaluation criteria were explored, including tool feasibility, interpretability, precision, reliability, validity and responsiveness. Minimally important changes were estimated for each domain, along with Cronbach's alpha and intraclass correlation coefficients for each questionnaire item. Post-operative questionnaire results within each separate domain were compared to their pre-operative baseline measures using paired statistical tests to identify any significant differences.

6.3 Implications of this research

6.3.1 Contribution to knowledge

This thesis has contributed to the currently published knowledge as follows:

1. By establishing the need for a TAA PROM by means of an in-depth literature review
2. By generating a conceptual model of patient HRQoL in patients who suffer from TAA disease
3. By developing and piloting a PROM for patients who undergo surgical treatment for TAA disease
4. By demonstrating the feasibility of delivering a PROM tool to this patient population, at 2 separate post-operative time intervals
5. By presenting the differences that surgical treatment of proximal TAA disease made to each PROM domain

It is hoped that these results may be of some potential use to future patients who are scheduled to undergo surgery for proximal TAA disease, either by supporting their understanding of what to expect from their treatment or allowing them to express opinions that may otherwise go unobserved. It is further possible that the PROM results could influence the delivery of healthcare, especially for patient information documentation, post-operative rehab strategies, or pre-operative support clinics for 'watch-and-wait' patients.

6.3.2 Generalisability of findings

The main barrier to how generalisable the findings of this study are is the small sample size, especially in the quantitative phase of the study. Small sample sizes in this cohort are explained somewhat by the relatively low incidence of surgical treatment for proximal TAA disease, especially when compared to the nationally mandated hip and knee replacement PROM populations, or the revascularisation population targeted in the recent pilot study (see Section 2.5.6.1).

6.4 Limitations

This study contains several limitations. There is an element of selection bias, as all patients were recruited from a single tertiary hospital in the North West of England. This bias would extend to environmental factors, treatment similarities and demographic characteristics such as ethnicity.

The lack of qualitative research experience that the primary investigator had should also be acknowledged as a limitation. It is possible that a more experienced qualitative researcher would have been able to elicit richer, more detailed results by using the skills and expertise which would be developed when carrying out multiple qualitative studies.

The primary researcher's employment at the hospital which hosted the study should also be considered. There would exist the potential for an unconscious degree of impartiality or a level of preconception about hospital care and treatment that should be taken into account when assessing the results of this research.

The TA approach to the qualitative phase of the study was considered to be appropriate based on TA output being conducive to PROM construction and the straightforward, introductory techniques of the method appealing to the novice qualitative researchers. It may be however, that a qualitative approach which produces a narrative output, such as an ethnography or phenomenology, could be used to extract a richer set of results from the data. Ethnographies are a more time consuming approach to qualitative research and they can be limited in how generalisable the findings are and in how subjective the analysis can be (Goodson, 2011). But in general, a deeper, thicker descriptive understanding of the research question under consideration can be achieved. Nanton (2016) used an ethnographic method to construct a narrative around how patients' personal identities can be compromised when suffering from serious or advanced illnesses, while Perry (2006) developed a feminist ethnography to report the experiences of how families respond to caregiving, with particular reference to the management of vulnerability.

It is possible that a phenomenological approach could also provide more involved results. Phenomenologies recognise that an individual's lived experience involves emotion, memory, thought and perception and each of these contain an 'intentionality' as the individual focuses on a specific entity or event (Rodriguez, 2018). Phenomenologies have been used in healthcare with some success, Kirkengen (2007) presented an integrated perspective on complex diseases such as cancer and autoimmune disorders which attempted to go beyond biomedical models such as aetiology, treatment and prognosis. Angner (2009) studied the relationship between health and happiness in 383 older adults living in community accommodation, the findings suggested that subjective measures of health may be better predictors of happiness than objective measures, and Finley (2003) published a phenomenological analysis of a single individual recently diagnosed with multiple sclerosis, focussing on diminished possibilities, the relationship between disease, activities and relationships, and the change in relationship the patient experienced with their body.

6.5 Future research

There are several possible avenues for future research studies in this area of proximal TAA PROM development. These are reviewed in the following sections.

6.5.1 Further PROM testing and development

The current proximal TAA PROM instrument requires more extensive psychometric testing. Greater patient numbers from other high-activity aortic surgery units are required to perform accurate Rasch tests and to allow a more robust evaluation of the questionnaire items, including any redundancy.

Future plans for PROM collection should include detailed exploration into alternative methods of delivery. Campbell (2015) offers encouragement to

the more widespread use of electronic PROM versions as his studies have found these methods of delivery to be equivalent to paper versions. However these innovations would have to be carefully planned and resourced before they could be put into practice.

Translation of the tool into different languages is another development that has the potential to be valuable. Wild (2005) and Sousa (2011) offer recommendations as to how this may be achieved, but caution that PROM translation is time-consuming and requires both intense planning and a rigorous methodological approach.

The questions asked at 6 weeks and 3 months in the current pilot are identical. Now that this study has shown some feasibility for the strategy of collecting results at 2 stages of the post-operative period, further work could be done to refine the questions asked at these two time points. For example, the question referring to post-operative attendance at a rehabilitation course may only need asking at the 3 month stage.

Some recognised layout / question construction issues need to be resolved. For example, the direction of the responses to the FFT question in the Q2 and Q3 instruments follow the national template and are presented from positive to negative, whereas all the previous domain question responses are presented from negative to positive. This switch in direction could confuse patients who have become accustomed to the previous layout, or result in unintended responses if they are ticking boxes 'on autopilot'. This conditioning effect could be particularly evident as the FFT question appears towards the end of the PROM.

The most acceptable way to account for the nature of aortic disease should be determined. Theoretically, the approach was taken that proximal TAA disease was distinct from problems with the aortic valve. In practice however, the overwhelming majority (94.4%) of these patients present with aortic valves which require surgical replacement. The causes and effects of valvular disease and aneurysmal disease are likely to be closely linked, the treatment the patients typically receive is a single operation for both

disorders, so it follows that any QoL questionnaire should be worded in a way that will account for this disease duality. The current questionnaires (Appendices F2, G2 and H2) are worded in keeping with treating TAA disease as a separate entity. It may be that patient comprehension and overall accuracy could be improved by using an approach which combined the two disease types.

6.5.2 Value of QoL information in high risk operations

Additional investigation into how patients perceive the value of QoL information in respect to more life-threatening diagnoses and treatments should be undertaken. While PROMs for life-threatening conditions have been encouraged (Insight & Feedback Team, 2017) if the condition is 'life or death' rather than functional, such as hip and knee replacements, then PROM results are likely to be of secondary interest at best. Patients who are candidates for surgical treatments understand that if their TAA continues to grow it will inevitably rupture and cause catastrophic injury. In a similar way, cancer patients recognise that if they do not receive treatment then the prognosis is bleak. So what value are HRQoL or PROM results to patients who have a high likelihood of death if their condition goes untreated? As PROM agendas extend into more complex pathologies, this type of question will become more relevant. It may be that the resources which are used for PROM construction, validation and administration would be better distributed on improving vital outcomes rather than on questionnaires.

Kotronoulas (2014) performed a systematic review into the value of cancer PROMs, and found some positive associations including an increase in the frequency of discussions regarding patient outcomes. Some studies included in the review also reported an increase in supportive care measures, patient satisfaction and improved symptom control. However, Kotronoulas concluded that more research would be required to support the cost-benefit of PROM implementation, including additional resources to handle the administrative

burden as well as additional support for clinicians to respond to patient concerns and issues.

6.5.3 Finding the most effective way to present PROM results

Testing and assessment of the most effective and informative way to present PROM results to both patients and clinicians is important. Research into the most informative, valuable way of delivering the results to stakeholders should include ease of interpretability, time required to assess performance and recommended levels of detail.

6.5.4 Utilise full extent of EQ-5D information

With results from more patients in a larger trial of the PROM, the EQ-5D results could be used to estimate QALY's (Gusi, 2010; Ogden, 2017), which could be used to inform national guidelines on the costs and benefits of proximal TAA disease and treatment.

6.5.5 QoL in surveillance patients, including possible interventions

Some future consideration should be given to the situation of individuals who have been diagnosed with an aortic aneurysm, but do not yet meet the size criteria for surgery – “watch-and-wait”, or surveillance patients. As can be seen from the results of the pilot study, the most significant health improvements for this patient population appear to be within the psychosocial domain. It follows that patients who are under indefinite surveillance for a potentially life-threatening disease may be suffering a greater burden of stress, anxiety or social preclusion than the general population. Some recent

work focussing on AAA's has been published around this subject, with thoughtful conclusions made about the difficulties in balancing the advantages and disadvantages of a surveillance program (Ericsson, 2019). UK AAA screening programs will in future have a QoL component for patients, with data collected online via the Screening Management and Referrals Tracking (SMaRT) IT system (Meecham, 2016; Public Health England, 2017). Earnshaw (2019) recognises that small QoL changes may seem inconsequential in light of the mortality risk that large aneurysms carry, but stresses that an understanding of the impact surveillance has upon patients is essential to how screening programs are designed and funded.

Similar work, directed towards the proximal TAA patient population, could be carried out in the future. Initiatives such as counselling or psychological prehabilitation (as Tsimopoulou (2015) describes regarding patients awaiting cancer treatment) may have a positive effect, but an appreciation of the costs and benefits of such proposals needs to be fully comprehended before recommendations can be made.

6.6 Conclusions

Following on from the stated aims of this study, information about the experiences of patients suffering from proximal TAA disease has been gathered, with particular reference to subsequent surgical treatment. This took the form of both in-depth patient interviews and the delivery of PROM questionnaires. The findings substantiated some of the conclusions already suggested by the existing literature in this area, regarding the improvements that surgical treatment can have on these patients' HRQoL. They also shed new light on specific issues that this patient population face. The information gathered from the qualitative study phase was used to develop a PROM tool that was designed to assess HRQoL and health status in these individuals. The evaluation properties of this new instrument were found to be acceptable, which unlocks the potential for this PROM to be used within routine clinical practice as part of patient support and outcomes assessment.

It is hoped that the findings described here will improve the experiences of these patients, for although they are not the largest pathological group, the effects of TAA disease upon HRQoL can be significant. The patient's family, friends and healthcare professionals may also find the results useful in order to better understand, or improve upon, the day-to-day experiences and medical management of this population.

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Appendix A. Letter of ethical approval



Health Research Authority

NRES Committee West Midlands - Solihull

The Old Chapel
Royal Standard Place
Nottingham
NG1 6FS

Tel: 0115 8839436

03 March 2015

Mr Matthew Shaw
Research Unit
Liverpool Heart and Chest Hospital
Thomas Drive
L14 3PE

Dear Mr Shaw

Study title:	THORACIC AORTIC ANEURYSM- A HEALTH ECONOMIC EVALUATION AND QUALITY OF LIFE ASSESSMENT: A PROSPECTIVE PATIENT QUALITY OF LIFE ASSESSMENT
REC reference:	13/WM/0456
Amendment number:	Substantial Amendment 1
Amendment date:	16 February 2015
IRAS project ID:	138112

The above amendment was reviewed by the Sub-Committee in correspondence.

Ethical opinion

The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

Approved documents

The documents reviewed and approved at the meeting were:

Document	Version	Date
Notice of Substantial Amendment (non-CTIMP)		16 February 2015
Other [Updated REC Form]		16 February 2015
Participant consent form	3	10 February 2015
Participant information sheet (PIS)	3	10 February 2015
Research protocol or project proposal	3	16 February 2015
Summary CV for Chief Investigator (CI) [Mr Matthew Shaw]		

Membership of the Committee

The members of the Committee who took part in the review are listed on the attached sheet.

R&D approval

All investigators and research collaborators in the NHS should notify the R&D office for the relevant NHS care organisation of this amendment and check whether it affects R&D approval of the research.

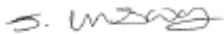
Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at <http://www.hra.nhs.uk/hra-training/>

13/WM/0456:	Please quote this number on all correspondence
--------------------	---

Yours sincerely

pp. 

Dr Rex J Polson
Chair

E-mail: nrescommittee.westmidlands-solihull@nhs.net

Enclosures: List of names and professions of members who took part in the review

Copy to: Margarita Perez-Casal, Liverpool Heart and Chest Hospital

NRES Committee West Midlands - Solihull

Attendance at Sub-Committee of the REC meeting on 25 February 2015

Committee Members:

<i>Name</i>	<i>Profession</i>	<i>Present</i>	<i>Notes</i>
Dr Rex J Polson	Consultant Physician - Chair	Yes	
Ms Gill Tomlinson	Head of Radiology, Solihull Hospital	Yes	

Also in attendance:

<i>Name</i>	<i>Position (or reason for attending)</i>
Miss Joanne Unsworth	REC Assistant

Appendix B. Recruitment letter, interviews

Mr/s P Patient

Addr 1

Addr 2

Addr 3

PP1 1XX

1st January 20XX

Dear Mr/s Patient,

I am writing to invite you to take part in a research study we are undertaking at Liverpool Heart and Chest Hospital which will look at quality of life in aortic aneurysm patients. You are being contacted because you have had aortic surgery in the past, or are due to have surgery soon.

The study involves a single one-to-one interview with a researcher at Liverpool Heart and Chest Hospital. The interview will last about one hour and take place in a private room within the hospital. All information given will be kept confidential. If you decide to participate, all of your travelling expenses and car parking charges will be reimbursed by the research team (up to a maximum of £50).

Your participation in the study would not alter your treatment in any way.

Please find enclosed a copy of the Patient Information Sheet which gives all the information regarding this study, I would be grateful if you would take the time to read it and decide whether you would be willing to be interviewed. It may be useful to discuss it with your family and friends or your GP or the Patient and Family Support Team at Liverpool Heart and Chest Hospital. The Patient and Family Support Team can be contacted on 0151 600 1517.

A member of the research team will call you in a few days. If you decide to participate we will discuss what will happen next. If you decide not to participate, the quality of your care at Liverpool Heart and Chest Hospital will not be affected in any way. In the meantime, if you have any questions or would like any further information then please contact Matthew Shaw, a member of the research team on 0151 600 1487.

Yours sincerely,

A handwritten signature in black ink, appearing to read 'M L Field', written in a cursive style.

Mr Mark L Field
Consultant Cardiac Surgeon
Liverpool Heart and Chest Hospital
Thomas Drive
Liverpool
L14 3PE

Appendix C. Patient information sheet, interviews

Liverpool Heart and Chest Hospital



NHS Foundation Trust

Information Sheet for Participants

01/01/20XX

Development and validation of a Patient Reported Outcome Measure (PROM) for patients undergoing Aortic Surgery

Dear Mr/s Patient,

You are being invited to take part in a research study which will examine the experiences of patients undergoing Aortic Surgery by means of an interview. This research is part of a PhD thesis being undertaken at Liverpool University. Before you make a decision, it is important for you to understand why this research is being done and what it will involve. Please take time to read the following information carefully and discuss it with your friends, relatives and your GP if you wish. Please do not hesitate to ask if there is anything that is not clear or if you would like more information. You may contact the study investigators whose details are given below. Take time to decide whether or not you wish to take part.

What is the purpose of the study?

During hospital visits many patients are often asked to complete questionnaires to assess their overall health and wellbeing. This information is useful for informing the hospital about the quality of services it provides. However, recent research has shown that many of them may not be acceptable to some people because some of the questions may be inappropriate. For example, the questionnaire may not mention some issues that are important to the patient.

We are inviting patients to participate in interviews to discuss and give their opinions on their aortic aneurysm disease before and after surgery. We would record the discussion and, with your permission, we may quote your comments but they would be anonymised. Information from the interviews

will inform the design of the next generation of questionnaires that we hope will be more meaningful to future patients.

Why have I been chosen?

You have been contacted because you have an aortic condition. You are due to have an aortic operation/intervention, or have had an aortic operation in the past, and are therefore eligible to take part in this study.

Do I have to take part?

You do not have to take part in this study. If you do decide to take part, you will be asked to sign a consent form and we will give you a copy of this information sheet and the consent form to keep. If after deciding to take part in the study you change your mind you are free to withdraw at any time, without giving a reason, and without your medical care or legal rights being affected.

What will happen to me if I take part?

If you agree, we will ask you to take part in a one to one interview with a student researcher in our hospital research unit. This interview will take place after your forthcoming outpatient appointment. We expect the interview to take no longer than 1 hour. All travelling expenses and any additional fees (e.g. car parking) up to a maximum of £50 will be covered by the hospital.

Will my taking part in this study be kept confidential?

Yes. If you decide to take part, all information that is collected about you during the course of the study will be kept strictly confidential and will only be accessed by members of the research team. Names will not be written on any of the transcripts, or reports of this research study. You will not be able to be identified from any report that is published from this study. With your permission, your GP will be informed that you are taking part in the study.

What will happen to the results of the interviews?

The results of this study will not be known for some time after the last person taking part in the study has completed their interview. At the end of the study a report on the findings of the interviews will be produced in collaboration with the patient's representatives/Service User's Research Endeavour (SURE) group at the Liverpool Heart and Chest Hospital. The results may also be published in medical journals and presented at research seminars and conferences. If you would like us to send you a copy of any published papers, please let us know.

Who is organising and funding this study?

The principle investigator for this study is Mr Matthew Shaw. The study is being supervised by Mr Mark Field of the Aortic Surgery Team at Liverpool Heart and Chest Hospital and Dr Alan Haycox of the Management School department at the University of Liverpool. The study is funded by the Aortic Surgery Team at Liverpool Heart and Chest Hospital. Researchers are not receiving any extra payments other than their usual salaries.

Who has reviewed this study?

The study was given a favourable ethical opinion for conduct in the NHS by the **Solihull Research Ethics Committee – West Midlands**. In addition, a patient representative of the Liverpool Heart and Chest Hospital Service Users Research Endeavour (SURE) group and the local research group have given their approval.

Contact for further information

If you have any other questions about the study, please contact **Matthew Shaw** at:

Research Unit
Liverpool Heart and Chest Hospital NHS Foundation Trust
Thomas Lane
Liverpool
L14 3PE

Tel: 0151 600 1487

E-mail: matthew.shaw@lhch.nhs.uk

What do I do now?

A member of the research team will get in touch via telephone in the next few days. In the meantime, please think about whether you would like to participate in the study or not.

What if there is a problem?

If you have a concern about any aspect of this study, you should ask to speak with any of the principal researchers listed above who will do their best to answer your questions. If you have concerns about any aspect of the way you have been approached or treated during the course of the study, you may wish to contact the hospital's Patient Advice and Liaison Service (PALS) now known as "Customer Care Team" on telephone number: 0151 600 1275 or 0151 600 1517

If you wish to make a formal complaint, please write to:

Chief Executive
Liverpool Heart and Chest Hospital NHS Trust
Thomas Drive,
Liverpool,
L14 3PE

or telephone the **Patient Complaints Manager** on 0151 600 1257

**Thank you very much for considering taking part in our
research.**

**Please discuss this information with your family and friends
if you wish.**

Appendix D. Patient consent form, interviews

Patient Consent Form

Patient Identification Number for this study: ___ ___ ___

Principal Investigator: Matthew Shaw

LHCH Research Study No: 1023

Title of Project: Development and validation of a Patient Reported Outcome Measure (PROM) for patients undergoing Aortic Surgery

Please initial boxes and sign at the bottom of the sheet

- 1 I have read and understand the patient information sheet provided to me and I have been given a copy to keep. I have had all my questions answered to my satisfaction, and I voluntarily agree to participate in this study.
- 2 I understand that my participation in this research project is voluntary. I understand that I am free to withdraw myself or my information from the research project at any time, without giving a reason and without my medical treatment or legal rights being affected.
- 3 I understand that participation involves being interviewed by a researcher from Liverpool Heart and Chest Hospital. The interview will last approximately 1 hour. The interview will be recorded.
- 4 I understand that the researcher will not identify me in any reports using information obtained from this interview, and that my confidentiality as a participant in this study will remain secure. Subsequent uses of records and data will be subject to standard data use policies which protect the anonymity of individuals and institutions.
- 5 I understand that information from this study may be used in scientific publications. My identity will be protected at all times.
- 6 I have been given a copy of this consent form.

.....
.....
Name of patient Date Signature

.....
.....
Name of researcher Date Signature

Appendix E. Interview guides

E.1 Pre-operative interview guide

Semi-structured interview questions (Pre-op)

1. Demographics: age, gender, educational level, employment or former employment, post code
2. Can you explain the details of how you were diagnosed with the aortic aneurysm?
3. Did/do you have any other health problems?
4. What was your quality of life like before being diagnosed? Any restrictions on your activities? Did you feel unwell?
5. How did you feel when you were diagnosed? (surprised, worried, anxious, scared?)
6. Did you restrict any of your daily activities once you were diagnosed?
7. How did your friends and family react?
8. How long did you have to wait until your operation?
9. What are/were your thoughts about how the surgery would affect your quality of life e.g. independence, mobility, ability to work, pain, time in hospital, complications?
10. Has the diagnosis affected intimacy in any way?
11. Anything else to do with your condition that you'd like to comment on?

E.2 Post-operative interview guide

Semi-structured interview questions

1. Demographics: educational level, employment or former employment
2. Can you explain the details of how you were diagnosed with the aortic aneurysm?
3. Did/do you have any other health problems?
4. What was your quality of life like before being diagnosed? Any restrictions on your activities? Did you feel unwell?
5. How did you feel when you were diagnosed? (surprised, worried, anxious, scared?)
6. Did you restrict any of your daily activities once you were diagnosed?
7. How did your friends and family react?
8. How long did you have to wait until your operation?
9. Did you think that the information you were given by clinicians was consistent?
10. What are/were your thoughts about how the surgery would affect your quality of life e.g. independence, mobility, ability to work, pain, time in hospital, complications?
11. How did the surgery go?

12. Were there any post-operative complications?
13. Where there any immediate or permanent side effects to the surgery?
14. Were there any other post-operative health problems?
15. How long do you feel it took to recuperate after the surgery?
16. Has your lifestyle/daily activities been any different since recovering after the surgery? In what way? What can't you do now that you could before the operation?
17. Has the surgery affected intimacy in any way?
18. Despite any infirmities / side effects of the operation, how do you feel in yourself now?
19. Anything else to do with your condition that you'd like to comment on?

Appendix F. Pre-operative patient mailing

F.1 Pre-operative PROM letter

Liverpool Heart and Chest Hospital 
NHS Foundation Trust

January 20XX

Dear Xx Xxxxxx,

Patient reported outcome measures for elective surgery

We would like your help with a survey that aims to evaluate the outcomes of heart surgery at Liverpool Heart and Chest Hospital, where you are scheduled to undergo an operation.

We will use the results of the survey to see which areas of our care are good, or which need to improve. This questionnaire is also being sent to other people who are having similar operations, to ask them about their experiences too.

Your feedback is very important to help us understand your health both before and after the operation.

It should only take about 15 minutes to complete the survey. You can do this on your own or ask a friend or somebody in your family to help. The doctors or nurses who treat you will not know that you have taken part in the survey and all your answers are **completely confidential**. This survey is voluntary, so if you don't want to take part, this will not affect your care and you don't need to give a reason if you choose not to be involved. You also have the right to withdraw from the survey at any point in the future without giving a reason.

If you do decide to take part, please complete and return the questionnaire in the **FREEPOST** envelope enclosed (you do not need a stamp). You will then be sent a second survey about 6 weeks after your operation, and a third survey about 3 months after your operation. The responses from these follow-up surveys will help us understand the how the operation has affected your health.

If you do not want to take part, you do not need to take any further action.

This survey is part of a research study run by Liverpool Heart and Chest Hospital. If you would like more information about the survey or need help to answer the questions, please call our Project Co-ordinator, Matthew Shaw on 0151 600 1487 and he will do his best to help.

Thank you

Yours sincerely,



Mr ML Field

**Consultant Cardiac Surgeon & Clinical Lead for Aortic Surgery,
Liverpool Heart and Chest Hospital**

Some questions and answers

What is the 'patient reported outcome measures' study about?

This study aims to assess how effective the operation you have is at improving your health. During hospital visits many patients are often asked to complete questionnaires to assess their overall health and wellbeing. This information is useful for informing the hospital about the quality of services it provides. However, recent research has shown that many of them may not be so useful as some of the questions asked may be inappropriate. For example, the questionnaire may not mention some issues that are important to the patient. The enclosed questionnaire has been designed to be more focussed on specific aspects of health that are related to aortic conditions.

Why have I been chosen?

You have been sent this letter because you have an aortic condition. You are due to have an aortic operation and are therefore eligible to take part in this study.

Do I have to take part?

You do not have to take part in this survey. If after deciding to take part you change your mind you are free to withdraw at any time, without giving a reason, and without your medical care or legal rights being affected.

How are my details being used?

If you decide to take part, all information that is collected about you will be kept strictly confidential and will only be accessed by members of the research team. Names or addresses will not be written on any of the transcripts or reports resulting from this research study. You will not be able to be identified from any report or analysis that is published from this study.

Can a relative or friend of the patient complete this questionnaire for them?

Yes, but the answers to the questions should be the views of the person who the questionnaire was sent to.

I can't answer one of the questions – what should I do?

If you can't answer a question just leave it blank and move onto the next one.

Why is the NHS spending money on a survey?

It is important for the NHS to ask people what they think about its services, as their views help to improve care. This survey has been specially developed to make sure that it asks questions about issues that really matter to people.

What will happen to the results of the questionnaires?

At the end of the study a report on the findings of the questionnaires will be produced in collaboration with the patient's representatives / Service User's Research Endeavour (SURE) group at the Liverpool Heart and Chest Hospital. The results may also be published in medical journals and presented at research seminars and conferences. Names or addresses will not be made public at any point. If you would like us to send you a copy of any published papers, please let us know.

Who do I contact for further information?

If you have any other questions about the study, please contact Matthew Shaw at:

Research Unit
Liverpool Heart and Chest Hospital NHS Foundation Trust
Thomas Lane
Liverpool
L14 3PE

Tel: 0151 600 1487

E-mail: matthew.shaw@lhch.nhs.uk

F.2 Pre-operative PROM questionnaire (Q1)

Aortic Aneurysm Surgery Questionnaire



Before your operation

Completing the questionnaire

For each question please tick clearly inside the box that is closest to your views using a black or blue pen. Don't worry if you make a mistake; simply cross out the mistake and put a tick in the correct box.

IMPORTANT INFORMATION

The purpose of this questionnaire is to collect information about the quality of healthcare services. The information collected will be used to produce statistics about the quality of healthcare services offered by different healthcare providers (hospitals) across the NHS. These statistics will be used to measure and improve the quality of healthcare services.

With your permission, the personal details that you provide and other information held about you in other NHS databases will be used to analyse and interpret the information collected.

By completing this questionnaire you are giving your consent for the information provided to be used for the purposes set out above. You are agreeing that:

- Your personal details and other relevant health information related to this operation will be held and used by Liverpool Heart and Chest Hospital, including relevant information held about you by the Personal Demographics Service, the Demographics Batch Service, the Secondary Uses Service, the Central Cardiac Audit Database and other NHS databases.
- Your personal details can be used to send you related follow-up questionnaires in the future.

Your personal information will be handled securely and it will be anonymised after analysis and before any publication. Liverpool Heart and Chest Hospital will not release your personal information unless required by law.

Your participation is voluntary. If you do not want to take part, do not fill in the questionnaire. You may withdraw the information you give the NHS in this questionnaire upon request, up to the point at which data are analysed, results are published and personal details have been removed.

If you have any queries about this questionnaire, please call the helpline on 0151 600 1487

Patient Details

Please write your name and address in CAPITAL LETTERS

First Name:

Surname:

House no/
Name:

Street/Road:

City/Town:

County:

Post code:

Q1. What is your date of birth (Please ensure this is your date of birth **NOT** today's date)

D D M M Y Y Y Y

Q2. Is anyone helping you fill in this questionnaire?
(Please tick one box)

Yes No
 ₁ ₂

Q3. Are you?
(Please tick one box)

Male Female
 ₁ ₂

Q4. Which statement best describes your living arrangements?
(Please tick one box)

I live with partner/spouse/family/friends ₁
I live by myself ₂
I live in a nursing home, hospital or other long-term care home ₃
Other ₄

Q5. When were you first diagnosed with an aortic aneurysm?
(Please tick one box)

Less than 1 month ago	1 to 3 months ago	4 to 6 months ago	7 months to 1 year ago	More than 1 year ago
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q6. Have you had a previous operation for a heart condition or aortic aneurysm?
(Please tick one box)

Yes	No
<input type="checkbox"/> 1	<input type="checkbox"/> 2

Q7. During the past 4 weeks, how much were you troubled by each of the following problems related to your heart condition or aortic aneurysm?
(Please tick one box on each line)

	A lot	Quite a bit	Moderately	A little	Not at all
Pain or discomfort in your chest	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Pain or discomfort in your back	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Pain in your chest that radiates to other parts of your body (eg arms, shoulders, hands, neck, throat, jaw, back)	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Tiredness / lethargy	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling dizzy, lightheaded or faint	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling unsteady or uncoordinated	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Shortness of breath	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Palpitations (a strong or irregular heartbeat)	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
General physical weakness	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Changes to your voice (hoarseness, or differences in tone)	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Problems with eyesight (eg blurred or double vision)	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q8. During the past 4 weeks, how much trouble has your heart condition or aortic aneurysm caused you?
(Please tick only one box)

A lot	Quite a bit	Some	A little	None
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q9. This question asks about activities which you might do during a typical day. During the past 4 weeks, has your heart condition or aortic aneurysm limited you in your usual daily activities? Please indicate whether the activities listed below are limited a lot, a little, or not limited at all. (Please tick one box on each line)

	Yes, limited a lot	Yes, limited a little	No, not limited at all
Moderate activities, such as moving a table, pushing a vacuum cleaner, bowling or playing golf	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Lifting or carrying groceries	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Climbing one flight of stairs	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Climbing several flights of stairs	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Bending, kneeling or stooping	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Walking 100 yards	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Walking half a mile	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Bathing yourself	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Dressing yourself	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3

Q10. This question asks about the impact of your heart condition or aortic aneurysm on your family and friends and the extent to which it has interfered with your social activities. During the past 4 weeks, how often have you experienced the following as a result of your health? (Please tick one box on each line)

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
Family or friends being overprotective towards you?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling like a burden on others?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling restricted in your social activities (like visiting with friends, relatives, etc)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling worried about going too far from home?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling restricted in tactile gestures (like being touched or giving hugs)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q11. This question asks about your feelings about your heart condition or aortic aneurysm. During the past 4 weeks, how often have you felt...:
(Please tick one box on each line)

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
Worried about your heart or aneurysm?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Worried about doing too much or overdoing it?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Worried about dying?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Frightened by the pain or discomfort of your condition?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Uncertain about the future?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Depressed, or in a low mood?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Frustrated or impatient?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Anxious or upset?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
That your condition interfered with your enjoyment of life?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
That it was difficult to keep a positive outlook about your health?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
That it was difficult to plan ahead (eg holidays, social events, etc.)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q12. This question asks about problems related to your heart condition or aortic aneurysm. During the past 4 weeks, how much of the time did you...:
(Please tick one box on each line)

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
Have difficulty reasoning and solving problems (eg making plans, making decisions, learning new things)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Forget things (eg events that happened recently, where you put something, appointments)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Have difficulty doing activities involving concentration and thinking?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Under each heading, please tick the **ONE** box that best describes your health **TODAY**

Q13. MOBILITY

- I have no problems in walking about 1
- I have slight problems in walking about 2
- I have moderate problems in walking about 3
- I have severe problems in walking about 4
- I am unable to walk about 5

Q14. SELF-CARE

- I have no problems washing or dressing myself 1
- I have slight problems washing or dressing myself 2
- I have moderate problems washing or dressing myself 3
- I have severe problems washing or dressing myself 4
- I am unable to wash or dress myself 5

Q15. USUAL ACTIVITIES (e.g. work, study, housework, family or leisure activities)

- I have no problems doing my usual activities 1
- I have slight problems doing my usual activities 2
- I have moderate problems doing my usual activities 3
- I have severe problems doing my usual activities 4
- I am unable to do my usual activities 5

Q16. PAIN / DISCOMFORT

- I have no pain or discomfort 1
- I have slight pain or discomfort 2
- I have moderate pain or discomfort 3
- I have severe pain or discomfort 4
- I have extreme pain or discomfort 5

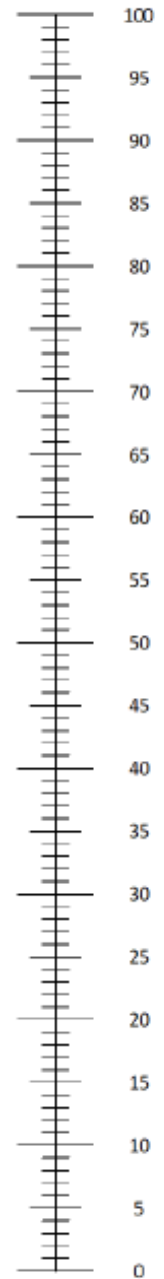
Q17. ANXIETY / DEPRESSION

- I am not anxious or depressed 1
- I am slightly anxious or depressed 2
- I am moderately anxious or depressed 3
- I am severely anxious or depressed 4
- I am extremely anxious or depressed 5

Q18. • We would like to know how good or bad your health is **TODAY**.

- This scale is numbered from **0** to **100**.
- **100** means the best health you can imagine.
0 means the worst health you can imagine.
- Mark an **X** on the scale to indicate how your health is **TODAY**.
- Now, please write the number you marked on the scale in the box below.

The best health
you can imagine



The worst health
you can imagine

YOUR HEALTH TODAY =

Q19. Have you been told by a doctor that you currently have any of the following?
(Please tick all that apply)

- | | | | | | |
|--|--------------------------|---|--|--------------------------|----|
| High blood pressure | <input type="checkbox"/> | 1 | Diseases of the nervous system (for example Parkinson's disease or multiple sclerosis) | <input type="checkbox"/> | 8 |
| Problems caused by stroke | <input type="checkbox"/> | 2 | Cancer (within the last 5 years) | <input type="checkbox"/> | 9 |
| Leg pain when walking due to poor circulation | <input type="checkbox"/> | 3 | Depression | <input type="checkbox"/> | 10 |
| Lung disease (for example asthma, chronic bronchitis or emphysema) | <input type="checkbox"/> | 4 | Arthritis | <input type="checkbox"/> | 11 |
| Diabetes | <input type="checkbox"/> | 5 | Coronary heart disease | <input type="checkbox"/> | 12 |
| Kidney disease | <input type="checkbox"/> | 6 | Problems with your heart valve(s) | <input type="checkbox"/> | 13 |
| Liver disease | <input type="checkbox"/> | 7 | Another long term health problem (please state): | | |

.....

Q20. Today's date (Please ensure this is today's date **NOT** your date of birth)

<input type="text"/>	<input type="text"/>	<input type="text"/>	<input type="text"/>	<input type="text"/>	<input type="text"/>	<input type="text"/>	<input type="text"/>
D	D	M	M	2	0		
				Y	Y	Y	Y

Q21. Do you consider yourself to have a disability?
(Please tick one box)

Yes	No
<input type="checkbox"/>	<input type="checkbox"/>
1	2

THANK YOU VERY MUCH FOR YOUR HELP

Appendix G. 6 week post-operative patient mailing

G.1 6 week post-operative PROM letter

Liverpool Heart and Chest Hospital 
NHS Foundation Trust

January 20XX

Dear Xx Xxxxxx,

Patient reported outcome measures for elective surgery

Before your operation about 6 weeks ago you completed a questionnaire for the patient reported outcome measure study. As you might remember, the study involves you completing questionnaires at three separate times. We are now sending you the second questionnaire. Completing the questionnaire three times allows us to find out how your health and quality of life has changed.

We hope you will be able to continue to participate, as your answers will help us to improve services for other patients.

Your feedback is very important to help us understand your health after the operation.

It should only take about 15 minutes to complete the survey. You can do this on your own or ask a friend or somebody in your family to help. The doctors or nurses who treat you will not know that you have taken part in the survey and all your answers are **completely confidential**. This survey is voluntary, so if you don't want to take part, this will not affect your care and you don't need to give a reason if you choose not to be involved. You also have the right to withdraw from the survey at any point in the future without giving a reason.

Please return the questionnaire in the **FREEPOST** envelope enclosed (you don't need a stamp). If you do not want to take part, please either return the blank questionnaire or call the helpline number below.

This survey is part of a research study run by Liverpool Heart and Chest Hospital. If you would like more information about the survey or need help to answer the questions, please call our Project Co-ordinator, Matthew Shaw on 0151 600 1487 and he will do his best to help.

Thank you

Yours sincerely,



Mr ML Field

**Consultant Cardiac Surgeon & Clinical Lead for Aortic Surgery,
Liverpool Heart and Chest Hospital**

Some questions and answers

What is the 'patient reported outcome measures' study about?

This study aims to assess how effective the operation you had is at improving your health. During hospital visits many patients are often asked to complete questionnaires to assess their overall health and wellbeing. This information is useful for informing the hospital about the quality of services it provides. However, recent research has shown that many of them may not be so useful as some of the questions may be inappropriate. For example, the questionnaire may not mention some issues that are important to the patient. The enclosed questionnaire has been designed to be more focussed on specific aspects of health that are related to aortic conditions.

Do I have to take part?

You do not have to take part in this survey. If after deciding to take part you change your mind you are free to withdraw at any time, without giving a reason, and without your medical care or legal rights being affected.

How are my details being used?

All information that is collected about you survey will be kept strictly confidential and will only be accessed by members of the research team. Names or addresses will not be written on any of the transcripts, or reports of this research study. You will not be able to be identified from any report or analysis that is published from this study.

Can a relative or friend of the patient complete this questionnaire for them?

Yes, but the answers to the questions should be the views of the person who the questionnaire was sent to.

I can't answer one of the questions – what should I do?

If you can't answer a question just leave it blank and move to the next.

Why is the NHS spending money on a survey?

It is important for the NHS to ask people what they think about its services, as their views help to improve care. This survey has been specially developed to make sure that it asks questions about issues that really matter to people.

What will happen to the results of the questionnaires?

At the end of the study a report on the findings of the questionnaires will be produced in collaboration with the patient's representatives / Service User's Research Endeavour (SURE) group at the Liverpool Heart and Chest Hospital. The results may also be published in medical journals and presented at research seminars and conferences. Names or addresses will not be made public at any point. If you would like us to send you a copy of any published papers, please let us know.

Who do I contact for further information?

If you have any other questions about the study, please contact Matthew Shaw at:

Research Unit

Liverpool Heart and Chest Hospital NHS Foundation Trust

Thomas Lane

Liverpool

L14 3PE

Tel: 0151 600 1487

E-mail: matthew.shaw@lhch.nhs.uk

G.2 6 week post-operative PROM questionnaire (Q2)

Aortic Aneurysm Surgery Questionnaire



After your operation

About six weeks ago you had surgery on your aorta. You may remember that you agreed that we could send you an *After your operation* questionnaire. Please can you fill in this questionnaire and return it using the provided pre-paid envelope. Thank you for your help.

Q1. Is anyone helping you fill in this questionnaire?
(Please tick one box)

Yes No

1 2

Q2. Today's date (Please ensure this is today's date **NOT** your date of birth)

D D M M Y Y Y Y

Q3. What is your date of birth (Please ensure this is your date of birth **NOT** today's date)

D D M M Y Y Y Y

Q4. Which statement best describes your living arrangements?
(Please tick one box)

I live with partner/spouse/family/friends 1

I live by myself 2

I live in a nursing home, hospital or other long-term care home 3

Other 4

Q5. Do you consider yourself to have a disability?
(Please tick one box)

Yes No

1 2

We are interested in finding out how you have been since your heart operation.
Please be sure to answer all questions.

**Q6. During the past week, how much were you troubled by each of the following problems related to your heart condition or aortic aneurysm?
(Please tick one box on each line)**

	A lot	Quite a bit	Moderately	A little	Not at all
Pain or discomfort in your chest	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Pain or discomfort in your back	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Pain in your chest that radiates to other parts of your body (eg arms, shoulders, hands, neck, throat, jaw, back)	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Tiredness / lethargy	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling dizzy, lightheaded or faint	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling unsteady or uncoordinated	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Shortness of breath	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Palpitations (a strong or irregular heartbeat)	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
General physical weakness	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Changes to your voice (hoarseness, or differences in tone)	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Problems with eyesight (eg blurred or double vision)	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

**Q7. During the past week, how much trouble has your heart condition or aortic aneurysm caused you?
(Please tick only one box)**

A lot	Quite a bit	Some	A little	None
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q8. This question asks about activities which you might do during a typical day. During the past week, has your heart condition or aortic aneurysm limited you in your usual daily activities? Please indicate whether the activities listed below are limited a lot, a little, or not limited at all. (Please tick one box on each line)

	Yes, limited a lot	Yes, limited a little	No, not limited at all
Moderate activities, such as moving a table, pushing a vacuum cleaner, bowling or playing golf	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Lifting or carrying groceries	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Climbing one flight of stairs	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Climbing several flights of stairs	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Bending, kneeling or stooping	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Walking 100 yards	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Walking half a mile	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Bathing yourself	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Dressing yourself	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3

Q9. This question asks about the impact of your heart condition or aortic aneurysm on your family and friends and the extent to which it has interfered with your social activities. During the past week, how often have you experienced the following as a result of your health? (Please tick one box on each line)

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
Family or friends being overprotective towards you?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling like a burden on others?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling restricted in your social activities (like visiting with friends, relatives, etc)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling worried about going too far from home?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling restricted in tactile gestures (like being touched or giving hugs)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q10. This question asks about your feelings about your heart condition or aortic aneurysm. During the past week, how often have you felt...:
(Please tick one box on each line)

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
Worried about your heart or aneurysm?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Worried about doing too much or overdoing it?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Worried about dying?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Frightened by the pain or discomfort of your condition?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Uncertain about the future?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Depressed, or in a low mood?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Frustrated or impatient?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Anxious or upset?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
That your condition interfered with your enjoyment of life?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
That it was difficult to keep a positive outlook about your health?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
That it was difficult to plan ahead (eg holidays, social events, etc.)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q11. This question asks about problems related to your heart condition or aortic aneurysm. During the past week, how much of the time did you...:
(Please tick one box on each line)

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
Have difficulty reasoning and solving problems (eg making plans, making decisions, learning new things)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Forget things (eg events that happened recently, where you put something, appointments)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Have difficulty doing activities involving concentration and thinking?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q12. This question asks about problems you might have had since your aortic aneurysm operation. During the past week, how much were you bothered by the following problems? If you did not have the problem, tick the last box 'Not at all'.
(Please tick one box on each line)

	A lot	Quite a bit	Moderately	A little	Not at all
Pain in your chest wound	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Infection in your chest wound	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Tenderness around your chest wound	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Numbness or tingling around your chest wound	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Bruising on your chest	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Swollen feet or ankles	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q13. This question asks about how satisfied you are with your aortic aneurysm operation.
How satisfied are you with the...:
(Please tick one box on each line)

	Very dissatisfied	Somewhat dissatisfied	Somewhat satisfied	Very satisfied
Results of your aneurysm operation?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4
Information you were given about your aneurysm operation?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4
Information you were given about how you might feel while recovering from your aneurysm operation?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4

Q14. This question asks about how satisfied you are with the post-operative hospital care.
How satisfied are you with...:
(Please tick one box on each line)

	Very dissatisfied	Somewhat dissatisfied	Somewhat satisfied	Very satisfied
The availability of hospital advice and support	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4
The regularity of outpatient clinic checkups?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4
The post-operative routine of regular follow up scans	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4
The way in which scan results are given to you?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4

Q15. Overall, how would you describe your attitude towards your aortic condition now compared to before you had your operation? (Please tick one box.)

Much worse	A little worse	About the same	A little better	Much better
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q16. Has your recovery from your aortic operation so far been: (Please tick one box.)

Slower than you expected	About what you expected	Faster than you expected	Did not know how long it would take
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4

Q17. Are the results from your aortic operation: (Please tick one box.)

Worse than you expected	About what you expected	Better than you expected
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3

Q18. Did you go on a rehabilitation course after you were discharged? (Please tick one box.)

No, I was not offered rehab	No, I chose not to attend	Yes, but I didn't complete the course	Yes, and I am still attending	Yes, and I completed the course
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q19. How likely are you to recommend this service to friends and family if they needed similar care or treatment?

Extremely likely	Likely	Neither likely nor unlikely	Unlikely	Extremely unlikely	Don't know
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5	<input type="checkbox"/> 6

Under each heading, please tick the **ONE** box that best describes your health **TODAY**

Q20. MOBILITY

- I have no problems in walking about 1
- I have slight problems in walking about 2
- I have moderate problems in walking about 3
- I have severe problems in walking about 4
- I am unable to walk about 5

Q21. SELF-CARE

- I have no problems washing or dressing myself 1
- I have slight problems washing or dressing myself 2
- I have moderate problems washing or dressing myself 3
- I have severe problems washing or dressing myself 4
- I am unable to wash or dress myself 5

Q22. USUAL ACTIVITIES (e.g. work, study, housework, family or leisure activities)

- I have no problems doing my usual activities 1
- I have slight problems doing my usual activities 2
- I have moderate problems doing my usual activities 3
- I have severe problems doing my usual activities 4
- I am unable to do my usual activities 5

Q23. PAIN / DISCOMFORT

- I have no pain or discomfort 1
- I have slight pain or discomfort 2
- I have moderate pain or discomfort 3
- I have severe pain or discomfort 4
- I have extreme pain or discomfort 5

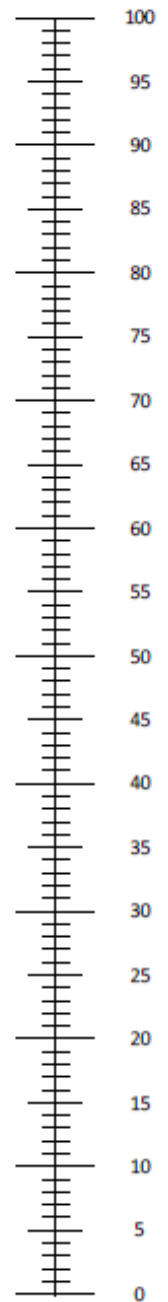
Q24. ANXIETY / DEPRESSION

- I am not anxious or depressed 1
- I am slightly anxious or depressed 2
- I am moderately anxious or depressed 3
- I am severely anxious or depressed 4
- I am extremely anxious or depressed 5

Q25. • We would like to know how good or bad your health is **TODAY**.

- This scale is numbered from **0** to **100**.
- **100** means the best health you can imagine.
0 means the worst health you can imagine.
- Mark an **X** on the scale to indicate how your health is **TODAY**.
- Now, please write the number you marked on the scale in the box below.

The best health
you can imagine



The worst health
you can imagine

YOUR HEALTH TODAY =

THANK YOU VERY MUCH FOR YOUR HELP

Appendix H. 3 month post-operative patient mailing

H.1 3 month post-operative PROM letter

Liverpool Heart and Chest Hospital 
NHS Foundation Trust

January 20XX

Dear Xx Xxxxxx,

Patient reported outcome measures for elective surgery

Before your operation about 3 months ago you completed a questionnaire for the patient reported outcome measure study. As you might remember, the study involves you completing questionnaires at three separate times. We are now sending you the third questionnaire. Completing the questionnaire three times allows us to find out how your health and quality of life has changed.

We hope you will be able to continue to participate, as your answers will help us to improve services for other patients.

Your feedback is very important to help us understand your health after the operation.

It should only take about 15 minutes to complete the survey. You can do this on your own or ask a friend or somebody in your family to help. The doctors or nurses who treat you will not know that you have taken part in the survey and all your answers are **completely confidential**. This survey is voluntary, so if you don't want to take part, this will not affect your care and you don't need to give a reason if you choose not to be involved. You also have the right to withdraw from the survey at any point in the future without giving a reason.

Please return the questionnaire in the **FREEPOST** envelope enclosed (you don't need a stamp). If you do not want to take part, please either return the blank questionnaire or call the helpline number below.

This survey is part of a research study run by Liverpool Heart and Chest Hospital. If you would like more information about the survey or need help to answer the questions, please call our Project Co-ordinator, Matthew Shaw on 0151 600 1487 and he will do his best to help.

Thank you

Yours sincerely,



Mr ML Field

**Consultant Cardiac Surgeon & Clinical Lead for Aortic Surgery,
Liverpool Heart and Chest Hospital**

Some questions and answers

What is the 'patient reported outcome measures' study about?

This study aims to assess how effective the operation you had is at improving your health. During hospital visits many patients are often asked to complete questionnaires to assess their overall health and wellbeing. This information is useful for informing the hospital about the quality of services it provides. However, recent research has shown that many of them may not be so useful as some of the questions may be inappropriate. For example, the questionnaire may not mention some issues that are important to the patient. The enclosed questionnaire has been designed to be more focussed on specific aspects of health that are related to aortic conditions.

Do I have to take part?

You do not have to take part in this survey. If after deciding to take part you change your mind you are free to withdraw at any time, without giving a reason, and without your medical care or legal rights being affected.

How are my details being used?

All information that is collected about you survey will be kept strictly confidential and will only be accessed by members of the research team. Names or addresses will not be written on any of the transcripts, or reports of this research study. You will not be able to be identified from any report or analysis that is published from this study.

Can a relative or friend of the patient complete this questionnaire for them?

Yes, but the answers to the questions should be the views of the person who the questionnaire was sent to.

I can't answer one of the questions – what should I do?

If you can't answer a question just leave it blank and move to the next.

Why is the NHS spending money on a survey?

It is important for the NHS to ask people what they think about its services, as their views help to improve care. This survey has been specially developed to make sure that it asks questions about issues that really matter to people.

What will happen to the results of the questionnaires?

At the end of the study a report on the findings of the questionnaires will be produced in collaboration with the patient's representatives / Service User's Research Endeavour (SURE) group at the Liverpool Heart and Chest Hospital. The results may also be published in medical journals and presented at research seminars and conferences. Names or addresses will not be made public at any point. If you would like us to send you a copy of any published papers, please let us know.

Who do I contact for further information?

If you have any other questions about the study, please contact Matthew Shaw at:

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H.2 3 month post-operative PROM questionnaire (Q3)

Aortic Aneurysm Surgery Questionnaire



After your operation

About 3 months ago you had surgery on your aorta. You may remember that you agreed that we could send you an *After your operation* questionnaire. Please can you fill in this questionnaire and return it using the provided pre-paid envelope. Thank you for your help.

Q1. Is anyone helping you fill in this questionnaire?
(Please tick one box)

Yes No

1 2

Q2. Today's date

(Please ensure this is today's date **NOT** your date of birth)

D D M M Y Y Y Y

Q3. What is your date of birth

(Please ensure this is your date of birth **NOT** today's date)

D D M M Y Y Y Y

Q4. Which statement best describes your living arrangements?
(Please tick one box)

I live with partner/spouse/family/friends 1

I live by myself 2

I live in a nursing home, hospital or other long-term care home 3

Other 4

Q5. Do you consider yourself to have a disability?
(Please tick one box)

Yes No

1 2

We are interested in finding out how you have been since your heart operation.
Please be sure to answer all questions.

**Q6. During the past week, how much were you troubled by each of the following problems related to your heart condition or aortic aneurysm?
(Please tick one box on each line)**

	A lot	Quite a bit	Moderately	A little	Not at all
Pain or discomfort in your chest	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Pain or discomfort in your back	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Pain in your chest that radiates to other parts of your body (eg arms, shoulders, hands, neck, throat, jaw, back)	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Tiredness / lethargy	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling dizzy, lightheaded or faint	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling unsteady or uncoordinated	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Shortness of breath	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Palpitations (a strong or irregular heartbeat)	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
General physical weakness	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Changes to your voice (hoarseness, or differences in tone)	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Problems with eyesight (eg blurred or double vision)	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

**Q7. During the past week, how much trouble has your heart condition or aortic aneurysm caused you?
(Please tick only one box)**

A lot	Quite a bit	Some	A little	None
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q8. This question asks about activities which you might do during a typical day. During the past week, has your heart condition or aortic aneurysm limited you in your usual daily activities? Please indicate whether the activities listed below are limited a lot, a little, or not limited at all. (Please tick one box on each line)

	Yes, limited a lot	Yes, limited a little	No, not limited at all
Moderate activities, such as moving a table, pushing a vacuum cleaner, bowling or playing golf	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Lifting or carrying groceries	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Climbing one flight of stairs	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Climbing several flights of stairs	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Bending, kneeling or stooping	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Walking 100 yards	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Walking half a mile	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Bathing yourself	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3
Dressing yourself	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3

Q9. This question asks about the impact of your heart condition or aortic aneurysm on your family and friends and the extent to which it has interfered with your social activities. During the past week, how often have you experienced the following as a result of your health? (Please tick one box on each line)

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
Family or friends being overprotective towards you?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling like a burden on others?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling restricted in your social activities (like visiting with friends, relatives, etc)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling worried about going too far from home?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Feeling restricted in tactile gestures (like being touched or giving hugs)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q10. This question asks about your feelings about your heart condition or aortic aneurysm. During the past week, how often have you felt...:
(Please tick one box on each line)

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
Worried about your heart or aneurysm?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Worried about doing too much or overdoing it?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Worried about dying?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Frightened by the pain or discomfort of your condition?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Uncertain about the future?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Depressed, or in a low mood?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Frustrated or impatient?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Anxious or upset?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
That your condition interfered with your enjoyment of life?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
That it was difficult to keep a positive outlook about your health?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
That it was difficult to plan ahead (eg holidays, social events, etc.)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q11. This question asks about problems related to your heart condition or aortic aneurysm. During the past week, how much of the time did you...:
(Please tick one box on each line)

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
Have difficulty reasoning and solving problems (eg making plans, making decisions, learning new things)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Forget things (eg events that happened recently, where you put something, appointments)?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Have difficulty doing activities involving concentration and thinking?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q12. This question asks about problems you might have had since your aortic aneurysm operation. During the past week, how much were you bothered by the following problems? If you did not have the problem, tick the last box 'Not at all'.
(Please tick one box on each line)

	A lot	Quite a bit	Moderately	A little	Not at all
Pain in your chest wound	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Infection in your chest wound	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Tenderness around your chest wound	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Numbness or tingling around your chest wound	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Bruising on your chest	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5
Swollen feet or ankles	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q13. This question asks about how satisfied you are with your aortic aneurysm operation.
How satisfied are you with the...:
(Please tick one box on each line)

	Very dissatisfied	Somewhat dissatisfied	Somewhat satisfied	Very satisfied
Results of your aneurysm operation?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4
Information you were given about your aneurysm operation?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4
Information you were given about how you might feel while recovering from your aneurysm operation?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4

Q14. This question asks about how satisfied you are with the post-operative hospital care.
How satisfied are you with...:
(Please tick one box on each line)

	Very dissatisfied	Somewhat dissatisfied	Somewhat satisfied	Very satisfied
The availability of hospital advice and support	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4
The regularity of outpatient clinic checkups?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4
The post-operative routine of regular follow up scans	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4
The way in which scan results are given to you?	<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4

Q15. Overall, how would you describe your attitude towards your aortic condition now compared to before you had your operation? (Please tick one box.)

Much worse	A little worse	About the same	A little better	Much better
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q16. Has your recovery from your aortic operation so far been: (Please tick one box.)

Slower than you expected	About what you expected	Faster than you expected	Did not know how long it would take
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4

Q17. Are the results from your aortic operation: (Please tick one box.)

Worse than you expected	About what you expected	Better than you expected
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3

Q18. Did you go on a rehabilitation course after you were discharged? (Please tick one box.)

No, I was not offered rehab	No, I chose not to attend	Yes, but I didn't complete the course	Yes, and I am still attending	Yes, and I completed the course
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5

Q19. How likely are you to recommend this service to friends and family if they needed similar care or treatment?

Extremely likely	Likely	Neither likely nor unlikely	Unlikely	Extremely unlikely	Don't know
<input type="checkbox"/> 1	<input type="checkbox"/> 2	<input type="checkbox"/> 3	<input type="checkbox"/> 4	<input type="checkbox"/> 5	<input type="checkbox"/> 6

Under each heading, please tick the **ONE** box that best describes your health **TODAY**

Q20. MOBILITY

- I have no problems in walking about 1
- I have slight problems in walking about 2
- I have moderate problems in walking about 3
- I have severe problems in walking about 4
- I am unable to walk about 5

Q21. SELF-CARE

- I have no problems washing or dressing myself 1
- I have slight problems washing or dressing myself 2
- I have moderate problems washing or dressing myself 3
- I have severe problems washing or dressing myself 4
- I am unable to wash or dress myself 5

Q22. USUAL ACTIVITIES (e.g. work, study, housework, family or leisure activities)

- I have no problems doing my usual activities 1
- I have slight problems doing my usual activities 2
- I have moderate problems doing my usual activities 3
- I have severe problems doing my usual activities 4
- I am unable to do my usual activities 5

Q23. PAIN / DISCOMFORT

- I have no pain or discomfort 1
- I have slight pain or discomfort 2
- I have moderate pain or discomfort 3
- I have severe pain or discomfort 4
- I have extreme pain or discomfort 5

Q24. ANXIETY / DEPRESSION

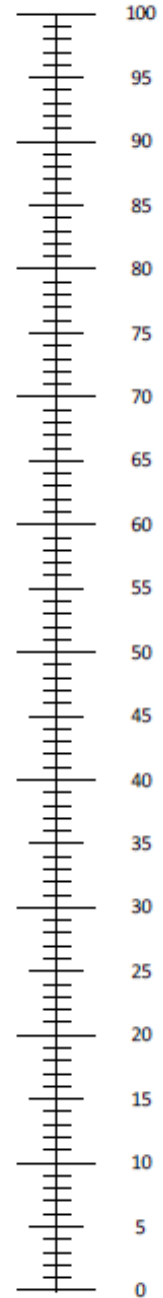
- I am not anxious or depressed 1
- I am slightly anxious or depressed 2
- I am moderately anxious or depressed 3
- I am severely anxious or depressed 4
- I am extremely anxious or depressed 5

Q25. • We would like to know how good or bad your health is **TODAY**.

- This scale is numbered from **0** to **100**.
- **100** means the best health you can imagine.
0 means the worst health you can imagine.
- Mark an **X** on the scale to indicate how your health is **TODAY**.
- Now, please write the number you marked on the scale in the box below.

YOUR HEALTH TODAY =

The best health
you can imagine



The worst health
you can imagine

THANK YOU VERY MUCH FOR YOUR HELP

