Explorations into the unique issues and challenges facing older men with haemophilia

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ABSTRACT

For the first time people who have haemophilia are facing the same aging issues as the general population due to longer life expectancy. This brings new and unique challenges for this group, and adds further complexity to their treatment, care, and support. The existing literature on this topic is dominated by a medical perspective that focuses on treatments and haematological management of haemophilia and common co-morbidities. Very little investigation into the wider effects of the challenges of growing older with haemophilia on an individual's holistic wellbeing has occurred. Accordingly, this research investigates the wider psychosocial experiences of older men with haemophilia in New Zealand using an exploratory sequential design that included a literature review, focus group, and national questionnaire. This mixed-method study identifies the unique issues and challenges faced by older men with haemophilia in Aotearoa, New Zealand, and their perceptions of the support services available to them. The results indicate that there are substantial challenges for older people with haemophilia, some of which have been identified in previous research and some which are presented in this thesis for the first time. Results also show that older people with haemophilia in New Zealand have very effective supports and services available to them. This research provides a starting point for in-depth conversations about older people with haemophilia and offers recommendations that can ultimately improve and enhance the lives and wellbeing of older men with haemophilia in New Zealand.

DEDICATION

To all the people with bleeding disorders around the world; your stories and resilience have inspired me and my practice - this thesis is for you. A special thank you to The Haemophilia Foundation of New Zealand for their support, and particularly the staff I had the privilege of working with over the last seven years.

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TABLE OF CONTENTS

LIST OF TABLES

LIST OF FIGURES

CHAPTER ONE – INTRODUCTION

It is now widely accepted that people with haemophilia (PWH) are living much longer lives due to improved treatment and care. A Swedish study, conducted from 1831 to 1902, investigating the life expectancy of people with severe haemophilia found that the median life expectancy for this population was 11 years old (Larsson, 1985), and data from Finland suggested the mean age of life expectancy was 7.8 years between 1902 and 1939 (Ikkala et al., 1982). Over time, due to numerous factors, this rate has slowly increased and life expectancy for PWH is now approaching that of the general male population (Mannucci, Schutgens, Sant'agostino, & Mauser-Bunschoten, 2009).

PWH are likely to have benefited from the general contributors to health improvement around the world such as medical advances, control of infectious diseases, recognition of hazards like tobacco and unsafe transportation, and improved sanitisation, drinking water and food handling (Centres for Disease Control and Prevention [CDC], 1999). More specifically this population has also benefited in recent years from advances in haemophilia care including the availability of safe, effective factor concentrate (explained later in this chapter), the development of comprehensive care programmes, new approaches to overall patient management, novel technologies, clinical trials, and therapeutic modalities such as home treatment and prophylaxis (Dolan & Gerry, 2010; Oldenburg, Dolan, & Lemm, 2009). As a result, the haemophilia population is facing the same aging issues as the general population for the first time. This brings new and unique challenges for this group that have never been seen before, and add further complexity to their treatment, care and support.

It is of concern that the majority of the literature and discussion surrounding older people with haemophilia (OPWH) comes from a medical perspective that focuses on treatments and haematological management of haemophilia and common co-morbidities. There has been very little investigation into the wider impacts that aging with haemophilia has on an individual's holistic wellbeing and there are significant gaps in the broad knowledge-

base pertaining to the needs and experiences of OPWH (Young, 2012). Academics and service providers alike recognise that there are many new challenges arising for this population that have never been seen before, and that assessments of patients' holistic needs are urgently required (Dolatkhah et al., 2014; Franchini & Mannucci, 2010). An understanding of haemophilia is needed to appreciate the rationale for the research thus an overview of the disorder is presented next.

Haemophilia – A Royal Pain

Haemophilia was first known in the ancient world, having been described in the Talmud (a central text of Rabbinic Judaism) that presented an exception to ritual circumcision in families because two brothers had already died of blood loss following the procedure (Neuman, 2010). However, the first scientific and medical descriptions of haemophilia were not put forward until 1803 by Dr John Conrad Otto, when he described affected males as 'bleeders', a name which is still commonly used today to describe someone with haemophilia (Holden, 1995). Although a rare disease, haemophilia is well known to many due to its part in the history of Russian and European royal families, and is therefore sometimes referred to as a 'Royal Disease' (Oldenburg et al., 2009).

Haemophilia is a chronic lifelong condition that results in excessive internal or external bleeding, and sometimes in spontaneous bleeding. The disorder arises from the partial or total lack of an essential blood clotting factor in the blood, and though PWH do not bleed any faster than normal, they bleed for a longer time and it is harder for their bodies to form a clot (World Federation of Hemophilia [WFH], 2012). A lack of clotting factor VIII is known as haemophilia A, or 'classic' haemophilia, and occurs in almost 80% of all haemophilia cases. A factor IX deficiency is known as haemophilia B, or 'Christmas Disease' (named after the first patient described with the disease – Stephen Christmas), and is associated with the majority of the remaining cases (Neuman, 2010). There are three severities of haemophilia: having less than 1% of normal factor level is severe haemophilia

and is associated with spontaneous bleeds often requiring ongoing prophylactic treatment; moderate haemophilia is present when factor levels are between 1–5% and spontaneous bleeds are rare but minor trauma can cause serious bleeding; and people with mild haemophilia who have a factor level between 5–40% generally only have bleeding problems associated with a major injury or surgery (Bussing & Johnson, 1992).

Haemophilia is a hereditary (sex-linked, recessive) bleeding disorder, which is passed down genetically through family lines. Affected individuals are invariably male due to the hereditary nature of the condition (an X-linked chromosome disorder), while women, for the most part, are carriers of the disease. In approximately 30% of cases haemophilia occurs spontaneously where there is no family history present, but will then carry down through that family line (Lauzon, 2008). If a father has haemophilia and a mother is unaffected, no sons would have haemophilia but any daughter would be an obligate carrier of the gene. If a mother carries the gene and the father is unaffected there is a 50% chance at birth that a son will have haemophilia and there is a 50% chance that a daughter will carry the gene. Haemophilia is a relatively rare condition, only one in 10,000 people are born with haemophilia A, and one in 50,000 born with haemophilia B (WFH, 2012).

If someone with haemophilia suffers a superficial cut, mechanisms to stop this kind of bleeding typically remain. Where excessive bleeding most often occurs (and is of the most concern) is internally into the joints and muscles (Bussing & Johnson, 1992). Common physical signs and symptoms of haemophilia include: large deep bruises; bleeding into muscles, joints or organs; spontaneous bleeding; and bleeding for a long time after being cut, having teeth removed, surgery, or an accident. Internal bleeding can lead to pain, swelling, stiffness, limited joint mobility, disability, and in some cases a ‘bleed’ can be life threatening (Lauzon, 2008).

Treatment for haemophilia was originally conducted via transfusion of whole blood in London in 1840 (Lane, 1840); however, at this time, most people with symptoms typically relied on rest, ice, compression, elevation and immobilization to alleviate the symptoms. Fresh-frozen plasma was introduced in the 1950s in the hospital setting, and was only used for life-threatening haemorrhages, emergency, and surgery; however, this treatment was fraught with problems and mishaps such as the need for very large doses of treatment being infused over long periods of time in hospital (Mannucci, 2008). In 1964 Cryoprecipitate was developed, and this allowed for more effective treatment with smaller volumes of fluid to be transfused into the patient (Pool & Hershgold, 1964). Next came the commercial plasma-derived intermediate-purity factor concentrates which became available in the early 1970s and enabled patients and caregivers to treat themselves in the home setting, and to proactively treat early signs of bleeding (Mannucci, 2008).

Devastatingly, the first Acquired Immune Deficiency Syndrome (AIDS) cases in the haemophilia community were reported in 1982 (Oldenburg et al., 2009). Both AIDS and the Hepatitis C Virus (HCV) were contracted through the plasma derived clotting products, and this had a catastrophic impact on the bleeding disorder community. In New Zealand 29 people tested positively for the Human Immunodeficiency virus (HIV), and at least 188 contracted HCV (Lauzon, 2008). After the bleeding disorder community was hit with these harsh viruses, plasma derived concentrates began being prepared from larger pools of donors and safety was increased. The 1980s brought the development of recombinant factor VIII concentrates (a form of blood clotting agent which has been manufactured via recombinant DNA technology, based on animal cells, meaning it has little to no human product in it and therefore does not contain viruses). These recombinant treatments have demonstrated excellent efficacy with approximately 80% of bleeding episodes being controlled by a single dose of treatment, and with very high safety ratings (Lusher, Arkin, Abildgaard, & Schwartz, 1993). As can be seen, the older haemophilia population have been through many highs and

lows due to haemophilia treatments and changes over time, but the introduction of these recombinant clotting products has been instrumental in increasing life expectancy for PWH. It is important to note however, that four-fifths of the world's haemophilia population still have minimal or no access to treatments for haemophilia, and in areas where treatments are unavailable PWH seldom make it to adulthood (Chuansumrit, 2003; Mannucci, 2008).

Haemophilia is not curable, but the previously mentioned treatments can minimize symptoms and make it more manageable. The treatment (known as factor concentrate, blood products, or just 'factor') is given by an infusion into the bloodstream with a needle and raises the clotting activity of the blood to a sufficient level to diminish or stop bleeding (Rosendaal et al., 1990), ultimately preventing joint damage. In this modern age, some people use 'on-demand' therapy - where they have an infusion when they are having trouble forming a clot. Others are on prophylaxis, which prevents or reduces spontaneous bleeding by taking their treatment preventatively every few days (Lauzon, 2008). With the introduction of these effective treatment methods haemophilia has become a controllable disorder that no longer automatically results in severe levels of joint impairment, disablement, and early death (Rosendaal et al., 1990). In most countries, where treatment products are offered, there are also Haemophilia Treatment Centres (HTCs) which follow multidisciplinary care models to support people with haemophilia. Fortunately, in New Zealand clotting products and the support of multidisciplinary teams are provided free to residents through the public health system.

Beyond the strictly medical effects, individuals with haemophilia can face a number of psychosocial challenges which affect their total wellbeing. The following are some of the key psychosocial areas of concern which are widely accepted and discussed regularly in the literature for all ages and stages of PWH:

- Patients may be at an increased risk for problems associated with adjustment to their illness such as stress, high anxiety, low self-esteem, and high rates of depression (Ghanizadeh & Baligh-Jahromi, 2009)
- Parental over protection ('helicopter parenting' and 'bubble wrapping') due to parent's high anxiety about their child's haemophilia (Cassis, 2007)
- Less access to and participation in full time paid work (Plug, Peters, & Mauser-Bunschoten, 2008)
- Living with pain (Elander, Robinson, Mitchell, & Morris, 2009)
- A lack of understanding about haemophilia among the general public and many healthcare professionals, which can leave patients frustrated and with a lack of faith in others (Cassis, 2007)
- Significant time missed from school which can affect education (Barlow, Stapley, Ellard, & Gilchrist, 2007)
- Compliance issues with medical regimes, and ignoring or dismissing bleeding in order to fit in with the rest of society (Cassis, 2007)
- Problems around social participation and support due to time in hospital and physical inability to participate or attend (Triemstra et al., 1998)
- People hiding their condition and facing discrimination (Barlow et al., 2007)
- Hard decisions about marriage and reproduction due to the hereditary nature of the condition they had (Cassis, 2007).

Alongside the known psychosocial issues facing the general haemophilia population there are some unique, under-explored issues beginning to emerge specifically for OPWH, which have not been seen before. As those with chronic conditions live longer, many will experience the same age-related symptoms and conditions that the general population experiences; however, those with chronic conditions may be faced with some age-related

conditions sooner, or to a greater extent than individuals who are aging without disabilities (Leuven, 2012). Consequently, the effects on those facing haemophilia alongside old age can be substantial, for both physical health and mental and emotional wellbeing (Meijer & Van der meer, 2007). In addition, these effects add further complexity to the lives and care of OPWH (Bos, 2007). Therefore, more research is urgently needed into the area of OPWH to ensure their needs are recognised and that they are supported as well as possible into old age.

Research Aims and Objectives

As previously stated, people with haemophilia are now living much longer lives due to improved treatment and care. This brings new and unique challenges to the haemophilia community that have never been seen before. Unfortunately, research to date is very limited and does not comprehensively look at the experiences of OPWH from a psychosocial or holistic perspective.

Specifically, this thesis addresses two research questions: 1) what are the unique issues and challenges facing older men who are living with haemophilia in Aotearoa New Zealand? and 2) how do older men with haemophilia perceive the support services available to them in Aotearoa New Zealand? These questions will be addressed via the following research objectives:

- Identify the unique issues and challenges faced by these men
- Explore the prevalence and seriousness of these issues
- Describe the impact of these issues and challenges on their holistic well-being
- Identify which services and supports are being used
- Describe their perceptions of the effectiveness of these services and supports

To meet the aims and objectives this research followed mixed methodology using an exploratory sequential design, with each phase informing the subsequent phase. Phase I was a literature review, Phase II included a focus group and instrument development, and phase III

was a questionnaire. The conclusions and recommendations were then based on a triangulation of the results of all three phases of the study. The participants for all parts of the study were men with haemophilia who lived in New Zealand and were over the age of 45 years.

In addressing the above objectives, this thesis ultimately aims to produce new knowledge about an aging population that is under-researched in New Zealand and overseas. It provides insights into the wellbeing of OPWH that have implications for the assessment and improvement of current services. The thesis thus raises awareness of the experiences and needs of OPWH, and provides recommendations for the future. Specifically, this research provides OPWH, professionals working with OPWH, and support organisations with a starting point for conversations about aging with haemophilia in Aotearoa, New Zealand.

Overview of Sections to Come

This thesis runs in a linear chronological order from start to finish. Chapter Two focuses on a review of the current literature on aging with haemophilia. Chapter Three outlines the general methodological design of the research and the ethical considerations. The following two chapters present the methods and results of a preliminary focus group study, and the subsequent questionnaire study. The findings from the literature, focus group and questionnaire are then triangulated and discussed in Chapter Six, and the limitations, recommendations and concluding thoughts presented.

This chapter has provided background information to what haemophilia is, and why aging with haemophilia is a new, complex, and unique issue. It has indicated the usefulness and importance of the research project, and presented the questions and aims which will be explored throughout the thesis. The literature review in the following chapter will delve further into the challenges that OPWH face, and demonstrate that there are many unanswered questions within this field, further highlighting the importance of the research.

CHAPTER TWO – LITERATURE REVIEW

A literature review was conducted to see what research was currently available on OPWH both in New Zealand and abroad. It was important to understand and identify what was already known in this field, and where there were gaps in information, to ensure this research project built on the current knowledge base and would add new information to the field of OPWH. This review is structured in two key parts in accordance with the two research questions. Literature pertaining to the first research question is examined in the first half of the literature review and explores the unique challenges and issues for those living longer with haemophilia. Literature relevant to the second research question, relating to services and supports available to OPWH, is analysed throughout the literature review where a service is connected to a particular challenge or issue, then the latter half of the review is focused specifically on support.

Multiple themes were identified in the literature and these are depicted by subheadings throughout the review. Notably, there was minimal literature available on the older demographic with haemophilia, and what was available was rarely from a holistic or psychosocial perspective.

Challenges and Issues facing Older People with Haemophilia

The key areas where challenges were present for OPWH were: physical health, which encompassed medical issues, pain, sexual activity and physical limitations; specific age related changes such as falls, independence and transitioning to care homes; work and income and the challenges these presented to OPWH; and mental and emotional wellbeing, specifically related to depression and life satisfaction.

Physical Health

As a consequence of people living longer with haemophilia a growing number of OPWH develop common, age-related co-morbidities, and in some cases they may be at higher

risk of certain age-related issues (Franchini & Mannucci, 2010). Many studies exist on the complexity of comorbidities in OPWH and how to best manage these. In fact, this area is where the majority of all literature on OPWH is focused. The main comorbidities found to be affecting OPWH are blood borne viruses such as Hepatitis C (HCV) and Human Immunodeficiency Virus (HIV), hypertension, cancer, osteoporosis (Hermans, Moerloose & Dolan, 2014), arthritis (Konkle, 2011), renal disease (Franchini & Mannucci, 2010), diabetes (Mauser-Bunschoten, Fransen Van De Putte & Schutgens, 2009), and the development of inhibitors (Franchini & Mannucci, 2010). Miesbach, Alesci, Krekeler, & Seifried (2009) state that co-morbidities are being seen more regularly in those living longer with haemophilia and are becoming more challenging for both healthcare providers and the patient to manage. Due to these new challenges Khleif, Rodriguez, Brown & Escobar, (2011) argue that greater care is now required in managing OPWH's healthcare needs.

Liver cancer is of particular concern as it is the leading cause of death in persons with haemophilia (Kupfer, Ruf, & Matz, 2005). This is due to the 'bad blood' scandal in the 1980s, when more than 90% of haemophilia patients in western countries who had received plasma derived clotting factor concentrates became infected with HCV (Dolan et al., 2009). Not only are some OPWH burdened with the hepatitis C virus and its specific health impacts, they also have to deal with the many serious long-term consequences that can accompany HCV. One of these impacts is that approximately one quarter of those chronically affected with HCV will go on to develop end-stage liver disease (Mauser-Bunschoten, Posthouwer, & Knegt, 2007), which can lead to death, as unfortunately the curative option of liver transplantation is made especially difficult for PWH due to bleeding problems (Posthouwer & Dirk, 2007). HIV also affected this population during the 'bad blood' scandal and therefore HIV related cancers are also more common in this group (Tagliaferri et al., 2002). General cancers are also a common cause of death in OPWH. A study conducted in the Netherlands from 2000–2007, found that malignancies, (other than liver cancer), were the primary cause of death for a PWH over 40

years old (Mauser Bunschoten et al., 2007). The prevalence across different types of cancers differed according to different studies; nevertheless, in all cases the prevalence is increased in OPWH compared to age-matched populations (even after excluding HCV and HIV related cancers) (Liu et al., 2014). Another of the more common and serious comorbidities for OPWH is kidney (renal) disease, which has a higher presentation rate compared to the general population, as PWH have additional risk factors for the disease, including HIV infection, factor inhibitors, and bleeding in their kidneys (Konkle et al., 2009).

Regardless of the type of co-morbidity it is well recognised that they all bring further complications for PWH, specifically in terms of conflicting medications and treatment options, new specialists being involved in their care, issues associated with undergoing surgery, and risks of bleeding. Therefore, as Dolan (2010) indicates, it is important that OPWH are aware of all age related diseases and obtain the necessary checks. The complications associated with co-morbidities are unique in that they have never been seen before in this population, as PWH have rarely lived to an age where they develop common age-related conditions. So, much is to be learned both from a medical standpoint and about how these comorbidities will affect the lives and wellbeing of OPWH.

Pain was another significant area of concern present throughout the international literature. Discussions focused on chronic and acute pain as well as the management of pain. Pain can have serious consequences on an individual's quality of life (QOL) (Riley, Witkop, Hellman, & Akins, 2011) therefore a clearer understanding of the nature of OPWH's pain, and how to best mange it, is needed. The Haemophilia Experiences, Results and Opportunities study (HERO study) was an international, multifaceted, multidisciplinary initiative, and the first large scale international research investigating psychosocial issues for PWH. It included 1,236 people across 10 countries, and was a collaborative study with many different researchers involved. The HERO study showed that of those surveyed, 50% of the adults with haemophilia reported having constant pain, and of those, only one in 10 said they had not

experienced any pain in the last four weeks (Garrido, Ramirez, Forsyth, & Lorio, 2012). It has also been found that pain has many associated mental and emotional consequences. This can be seen in the survey of 71 adults in Utrecht, the Netherlands, with severe haemophilia where 85% of participants reported negative mood due to pain (Genderen et al., 2006). Further, Elander et al.'s (2009) study of 209 men with haemophilia from the United Kingdom, concluded that the intensity of pain was the primary influence on physical QOL and negative emotions.

Despite apparent high rates of pain occurring in PWH, Elander (2014) recognises that pain specialists are rarely consulted, and that there was often ineffective management of pain issues by medical professionals. HTC staff are of particular concern, as they have had little specific training in, or understanding of, what relieves chronic pain. One U.S. study of pain experienced by 1,147 adults with a bleeding disorder identified that 39% of participants reported that pain was not well treated by their HTC (Witkop et al., 2012). In another study of adults with severe haemophilia, 50% of participantsreported constant and untreated pain (Riley et al., 2011). Numerous researchers strongly express concerns at the management of pain in OPWH, and recognise that there are large gaps in research concerning what is known in this area and how best to deal with it. These researchers also agree that pain has a large impact on QOL and daily functioning, and that it needs to be addressed (Holstein et al., 2012; Mauser-Buschoten et al., 2009; Philipp, 2010; Witkop et al., 2012 & Young, Tachdiina, Baumann, & Panopoulos, 2014). Because pain is badly managed and can severely affect QOL, Riley et al. (2011) recognise the importance of ongoing psychosocial assessment, and the need to standardise the management of pain treatment, approaches, and protocols to help reduce the effect of pain on people's lives.

There are also some problems with patients self-treating, and using drugs, both illicit and prescribed, as a way to manage pain. PWH are at least as vulnerable as other chronic pain populations to opioid-related adverse events, and to developing abusive behaviours and

addictions (Mannucci, Schutgens, Sant'agostino, & Mauser-Bunschoten, 2009). As stated by one participant in Rolstad's (2014) research, "Pain relief is a huge challenge, you know, because you go on narcotics and then you get addicted to them, which creates another set of challenges" (p.490). Thus, pain is a unique challenge for PWH and, unfortunately, there is little specific information in terms of OPWH and their experiences of pain, needs around pain support, and the complexities of medical management of pain. Fortunately, however, authors recognise this gap and realise pain is an under-investigated issue for OPWH, and one that would benefit from further research (Riley et al., 2011).

Another major issue experienced by OPWH is joint disease and its associated problems. Hemarthrosis (or joint bleeding) is the most common complication seen in OPWH despite advancements in treatment and care. Consequently, it is seen as a major concern for both healthcare providers and OPWH themselves (Mulder & Llinas, 2004; Valintino, 2010). Hemarthrosis most commonly affects the knees, elbows, and ankles, and recurrent bleeding in the same joint leads to development of a 'target joint' (a joint that bleeds repeatedly and ends up damaged), and often to arthritis (Madhok, York, & Sturrock, 1991). Many adults advancing into older age, who have severe haemophilia, were not treated with prophylaxis as children and therefore now have established joint disease and the associated burden of joint deformity, muscle weakness, and impaired proprioception (problems with body movement and positioning) (Konkle & Barbara, 2011). Such conditions can affect QOL, and as recognised by Chen et al. (2015) may cause difficulty with physical functioning, vitality, social functioning, pain, increased risk of falls, and can lead to emotional problems. Nearly two thirds of adults with haemophilia in the HERO study reported having limited mobility and almost half experienced limitations with usual activities (Garrido et al., 2012).

Issues with mobility also affect OPWH's abilities to participate in physical activity. Participation in exercise is often reduced because of mobility problems and also because people with haemophilia have often grown up unable to participate in athletic activities,

especially contact sports, due to concern that it might lead to bleeding problems. They are therefore less likely to have the motivation for, or appreciation of, exercise (Rolstad, 2014; Goto et al., 2014). Numerous studies showed that reduced mobility is a significant issue for OPWH, as is their ability to participate in physical activity. However, there is little discussion about the psychosocial effects associated with this reduced ability to move and participate, or ways that it might be managed or addressed.

Engaging in sexual activity contributes to wellbeing and QOL, however this issue was reported by multiple authors as a concern for OPWH. The main reasons for this include: musculoskeletal problems that limit activities and participation; sexual desire being disturbed by side effects of medications; fatigue; low testosterone; pain; arthropathy; iliopsoas muscle bleeding (bleeding into the muscle by the pelvis); and the fear of HCV or HIV transmission (Gianotten & Heijnen, 2009). Despite plentiful discussion about the inability of OPWH to fully engage in sexual activity, the HERO study found that 78% of adults with haemophilia were satisfied with the overall quality of their sex lives (Cassis, Querol, Forsyth, & Lorio, 2012). Numerous authors indicate it is important that OPWH receive support and referrals to specialists for sexual wellbeing and that OPWH are taught how to adapt and explore other options for comfort and satisfaction. Routine discussions with healthcare professionals are also encouraged (Hermans et al., 2014; Lambing & Kachalsky, 2009; Mauser-Bunschoten et al., 2009). While there is much discussion about sexual engagement in PWH by academics, it is hard to gather a full picture on this topic from the voice of those affected with haemophilia themselves, perhaps due to the sensitive nature of the topic.

Age Related Changes

It seems that the most common cause of injury in the general population of those aged 65 or more occurs by suffering a fall. Approximately 35% of community-dwelling older adults fall each year due to issues with circulatory changes, sensory perception, and balance, all of which increase the potential for falls (Forsyth, Quon & Konkle, 2011). This is further

increased in PWH as they have moderate impairments in balance, stability, gait, and mobility compared to the general population. Falls for PWH can have very serious consequences and can lead to head or internal bleeding, which can be life threatening or result in long hospital stays (Mauser-Bunschoten et al., 2009). A recent Dutch study of 74 PWH, aged 40 years or older, found that 32% had fallen in the past year (Sammels, Vandesande, Vlaeyen, Peerlinck & Milsen, 2014), and a study of 20 PWH from the Alfred Hospital in Australia, showed that 50% had fallen in the last year. Falling and balance are areas of growing concern for OPWH due to the serious consequences they can have on an individual (Fearn et al., 2010 & Dolan, 2010).

The complications experienced by OPWH have resulted in challenges for a person's ability to self-infuse their haemophilia treatment. Lambing and Kachalsky (2009) indicate that joint arthropathy and reduced dexterity can be aggravated with aging, which limits the ability to use a needle and syringe. Scar tissue from years of using the same vein can occur and age related vision problems may also make it difficult for someone with haemophilia to find a vein and self-infuse. Therefore, the loss of independence that comes with being unable to medicate themselves can also contribute to waning mental health, distress, and frustration as they can no longer complete what was once an easy task (Mauser-Bunschoten et al., 2007). This was a topic scarcely mentioned in the literature, despite being an issue which has the potential to have tremendous negative effect on a person's wellbeing.

OPWH will face an increasing need for care outside the home, hospital, and clinic as they live longer into middle and old age (Allen & Kachalsky, 2010). For some, their support networks may shrink and they may need to give up independent life and move into a nursing home. For haemophilia patients, who have fought for self-determination for many years and manage their own condition, this could be very traumatic and cause additional stress and negative emotions (Mauser-Bunschoten et al., 2009). Care for these patients in settings outside their home or hospital is hampered by the lack of factor products (and whether they

are allowed in the nursing home or not) and concerns about who will administer factor infusions, as well as who will educate the nursing homes about haemophilia (Allen & Kachalsky, 2010). De Knecht-Van Eekelen (2007) interviewed older men with haemophilia and her findings showed that some participants were very worried about going into nursing homes where the employees knew nothing about haemophilia care.

Work and Income

OPWH are still less involved in fulltime paid work than people without haemophilia and most have historically been dependent upon disability schemes and allowances in order to survive (Rolstad, 2014). PWH suffer more from occupational disability than men from the general population, and they experience regular sick-leave due to bleeds, pain, missing work for medical appointments, disability and mobility issues, and a lack of motivation (Bos, 2007). There is also often fear of discrimination in the workplace and being treated differently because of their disorder (Rolstad, 2014). Due to their physical limitations, PWH have significantly fewer job options than men without haemophilia, and sometimes have fewer opportunities for advancement or switching to different employers (Rolstad, 2014). The HERO study showed more than half of the respondents reported a negative impact of haemophilia on their working life, and some believed they had lost a job because of haemophilia (Forsyth et al., 2012). All of these issues, as well as the need for early retirement, may affect finances and increase psychosocial problems, including depression (Iannone, Pennick, Tom, Cui, Gilbert, Weihs, & Stopeck, 2012). On the contrary, Mauser-Bunschoten et al. (2007) and De Knecht-Van Eekelen (2007) claim there has been an increasing number of persons with haemophilia entering the work force, and many are so determined to work and live normal lives that they do a better job than the average employee and have held jobs for much longer periods. To add to this, the HERO study shows that people with haemophilia tend to be educated, and that the majority were employed (Forsyth, Gregory, & Lorio, 2014).

Financial problems were mentioned throughout the literature in relation to private insurance. For instance, Rolstad (2014) found that the single greatest challenge for men with haemophilia was obtaining either private or public insurance that would cover the cost of their medical treatment. It is important to note, however, that this is not the case in New Zealand, as treatment is publicly funded. The 'satisfaction with income' study conducted with adults with haemophilia found that 43% of the participants were unhappy with their income, 12% were satisfied and 17% were without income. This study also used a QOL questionnaire and found that there was a significant correlation between QOL and level of income (Dolatkhah et al., 2014). In contrast, a study conducted in Italy on 210 OPWH found that these participants had no problems with money management (Siboni, 2009).

Mental and Emotional Wellbeing

According to the literature, OPWH are disposed to mental health and emotional problems, and, in fact, may be more vulnerable to chronic self-esteem issues, stress, depression, and anxiety than the average age-comparable population (Canclini et al., 2003). Depression may stem from a range of things, such as: having a history of frequent medical problems; multiple hospital visits; painful joint disease; unemployment; a lack of social support; and, perhaps, from poor self-esteem that began during childhood when physical limitations and perceived difference from others established a foothold on their lives (Standish, 2001). This is concerning, as depression can be seen to cause or accelerate other health-related diseases. It can also lead to: reduced social or mental functioning, dangerous behaviour such as drug or alcohol abuse, and poor adherence to medical treatments; and have a major detrimental impact on one's general QOL (Raison & Miller, 2001). One study of 41 adult patients with haemophilia suggested that approximately one-third met the criteria for depression (Iannone et al., 2012). Another found 37% of the adult patients treated at the Arizona HTC reported they were suffering from some depressive symptoms, and almost 20% reported moderate to severe depressive symptoms. Forsyth et al. (2014) also showed that 47%

of adults with haemophilia in their study experience psychological and psychiatric conditions due to the impact of their haemophilia. Although many of the studies were not focused specifically on OPWH, it is believed that mental and emotional issues such as anxiety, depression, pain, and discomfort are increased for OPWH compared to younger PWH (Niu et al., 2014). Thus it is important that depression screening be incorporated as part of comprehensive care for OPWH, as this would be useful for psychosocial evaluation and engagement with patients (Iannone et al., 2012).

There is very little literature available on the satisfaction with life of PWH. What is available is mixed; some research indicates that PWH experience lower quality and satisfaction with life than non-affected individuals (Siboni et al., 2009) while others show that PWH in general have high life satisfaction (Beeton, Neal & Lee, 2005). The latter perspective is thought to be based on PWH's build-up of resilience over time, their ability to advocate for themselves and to ask for what they need, and because they may be less likely to complain about the unimportant issues in their lives (Rosendaal et al., 1990). It may also be that their internal locus of control (belief about their ability to influence events and outcomes) reduces the perceived seriousness of their haemophilia (Triemstra et al., 1998). De Knecht-Van Eekelen (2007) conducted interviews with OPWH and found that some had good life satisfaction and felt very grateful (and showed very positive mind-sets and resilience), whereas others had a very low life satisfaction.

The literature demonstrates that physical health is the biggest challenge for OPWH, particularly co-morbidities, pain, sexual activity, and physical limitations. Unfortunately, there was very little research pertaining to specific age-related changes such as falls, independence, and transitioning to care homes. Work presented challenges to many OPWH, as did mental and emotional wellbeing, with many studies showing depression being elevated in this population. Leading authors in the field of PWH, including Franchini & Mannucci (2010), Young (2012), Bos (2007) and Dolatkhah et al. (2014), all recognise that OPWH are

facing many new challenges and that assessments of patients 'needs' are urgently required. Due to the wide variety of challenges for this population, it is important to see what services and supports are available to address and deal with these issues.

Services and Supports for Older People with Haemophilia

The second part of the literature review was conducted to investigate what services and supports were available to OPWH. For this section, the term 'services' refers to those groups or individuals who can provide assistance, support, aid, and education to OPWH. The key services mentioned throughout the literature were comprehensive care teams, including haemophilia treatment centres (HTC) and physiotherapists. Mentioned far less frequently in the literature, but very important to acknowledge when considering holistic wellbeing, were psychosocial professionals, the National Membership Organisations (NMOs), and the role of informal social support in the lives of OPWH.

Comprehensive Care

The aforementioned section on increasing comorbidities for OPWH shows they are contending with a growing number of new conditions. Consequently, OPWH require intervention from a variety of different specialists, services, and people. Raabe (2008) states that haemophilia is often not well understood by medical providers who do not specialise in it because it is such a rare condition. It is often difficult for PWH to find doctors and medical providers who are either knowledgeable about haemophilia and its management or who are willing to listen to them about their treatment needs, both of which can cause a lot of frustration for PWH (Rolstad, 2014). In many cases OPWH may never have consulted primary care physicians due to the rarity and complexity of their haemophilia, and therefore generally rely on their HTC to take responsibility for the management of all their health problems (Mauser-Bunschoten et al., 2009). However, with the rising number of age-related co-morbidities in OPWH, and the complexity of treatment, care, and management, it is essential for OPWH to engage with appropriate medical specialists. It is also imperative that

OPWH receive general health checks and are connected with general practitioners to manage their overall health. It is vital that there is a smooth flow of information and communication between the HTC and other specialists and GP's involved in the care of OPWH (Hermans et al., 2014).

The role of HTCs is to provide optimal care for all people with bleeding disorders. HTCs have made critical contributions toward improving QOL for individuals with haemophilia, and teaching and empowering patients to participate in their own care (Evatt, Black, Batorova, Street & Srivastava, 2004). HTC staff often develop on-going relationships and rapport with patients, and provide multiple services for not only physical symptoms but also psychosocial issues (Allen & Kachalsky, 2010). There is compelling data to suggest that HTCs have a significant effect on reducing the mortality rates in patients with haemophilia and that the overall survival of PWH may heavily depend on the level of expertise within, and their experience of, their HTC (Soucie et al., 2000).

While the majority of literature praises HTCs, the wider multidisciplinary teams, and the support they provide, there is some controversy over the support provided by HTCs specifically for the older demographic with haemophilia. De Bruin et al. (2012) argue HTCs often "fail to address the integral healthcare demand, including psychosocial issues, of patients with multiple chronic conditions. As a result, multimorbid patients are prone to receive fragmented, incomplete, inefficient, and ineffective care" (p. 109). However, it is acknowledged by others that HTCs have limited experience with the wide scope of issues the older haemophilia population is facing. Rolstad (2014) found PWH had mixed reactions about the services provided by their local HTC. On the one hand, PWH would like to be able to spend more face time with staff members in their HTC, exchange more information, and have mutual (rather than one-sided) conversations. On the other hand, PWH do not want their annual visits to consume a great deal of time, or to participate in lengthy comprehensive visits with a whole series of services providers. Several authors argue that individuals working with

older people with chronic conditions need to be upskilled, and they need to be more proactive instead of reactive (García-Goñi, Hernández-Quevedo, Nuño-Solinís, & Paolucci, 2012). It is clear that HTCs are very beneficial, however, because aging with haemophilia is a new phenomenon, they currently lack specific knowledge of the needs of OPWH and how to best deal with them.

On the whole haemophilia physiotherapy is seen to be a very effective service, which is paramount in helping PWH prevent, manage, and recover optimally from bleeds (Wittmeier & Mulder, 2007). Specialist physiotherapists help with pain management, education, home programs, bleed rehabilitation, post-operative rehabilitation, joint and muscle assessments and evaluations, and a range of other treatment options (Beeton et al. 2010). They can also put in place specialised and individualised care and physical training programs (Hilberg, Herbsleb, Puta, Gabriel, & Schramm, 2003). Very little information pertaining to physiotherapy and OPWH (other than in terms of falls and balance) exists in the current literature, and according to Dolan (2010) a radical review of the available physiotherapy services for OPWH is required to improve future comprehensive care for this age group. So, although there is positive discussion of the value of physiotherapy for PWH, more targeted research is needed to ensure physiotherapy services meet the specific needs of OPWH.

It is commonly understood that psychosocial factors have a significant impact on QOL for OPWH (Cassis et al., 2012a). It is noted by some that with an elevated level of distress (i.e., anxiety, depression, and anger) among OPWH, counselling and social support is essential (Posthouwer et al., 2007). Despite the need for psychosocial support, Rolstad (2014) found that the men in his study were reluctant to receive psychosocial support and unreceptive to therapeutic interventions. The role of social workers in the lives of OPWH (as reported in the majority of the literature) focuses primarily on organising insurance for treatment and care, however, this is not relevant in New Zealand as treatment is publicly funded. The social work role is also acknowledged in relation to the HIV and HCV infections prevalent in PWH

in the 1980s. At this time social workers played a large role in supporting PWH and their wider families in many ways (Allen & Kachalsky, 2010). Social workers helped to educate people about the virus. They helped PWH to work through emotional issues connected to contracting the virus, and understanding the ramifications for their future. Social workers ran events and workshops and connected PWH to others who had also contracted the viruses for social support, and they practically assisted PWH with advocacy. There is some general and basic information on the role of the social worker in the lives of those with haemophilia, such as to educate, advocate for, empower, and support PWH. However, the social work role and its importance is predominantly described in relation to newly diagnosed families, young children, and teenagers. There is scant literature on the social work role in relation to the changes or improvements they need to make in their practice to adequately deal with the new needs of OPWH. There seems to be very little available in the literature that adequately describes or encourages assistance from professionals such as psychologists, counsellours, social workers, psychiatrists, and other psychological interventions that could provide support to OPWH.

Social Support

The Haemophilia Foundation of New Zealand (HFNZ) is the National Membership Organisation (NMO) for people with bleeding disorders throughout New Zealand. 'Still Standing,' a book of their history suggests HFNZ has always been dedicated to providing excellent education, support, care, and advocacy for all people with bleeding disorders in New Zealand (Lauzon, 2008). Basic information is available about NMOs around the world in terms of their function and histories, but there is little academic information on their contribution to OPWH. An exception is a recent study conducted by Rolstad (2014) in which the participants said they did not feel that they personally needed much in the way of support from the bleeding disorder community. In fact, several men observed that the same set of topics are addressed over and over again at community events. This created a disincentive for

men to participate in activities. Although there is little research on NMOs, and specifically NMOs and OPWH, Mauser-bunschoten et al. (2007) recommend that people living longer with haemophilia should become members of these organisations to obtain more information, education and support. However, NMOs need to be aware of the issues experienced by OPWH and develop 'wellness' programs, practices, policies, and supports which are directed at preventative strategies to reduce the physical and psychological impacts of aging (Street, Hill, Sussex, Warner, & Scully 2006). Currently, it is hard for NMOs to do so when so little information about the psychosocial effects of living longer with haemophilia exists.

It is well known in general that social relations and interaction with other people is an important part of the everyday life and well-being of older people (Pynnonen, 2012). According to Bos (2007), and Iannone et al. (2012) a lot more research is needed in the area of wellness programmes and peer meetings for senior haemophilia patients, as these interventions may help to prevent the physical and psychological impacts of aging. The importance of social connectivity is widely accepted, yet there is so little information available pertaining to its specific importance for PWH, let alone OPWH.

Partners and significant others have important roles in assisting with care, treatment, and support of their older person with haemophilia. Partners can take on a lot of the ‘weight’ and extra responsibility, and often OPWH are overly dependent on their partners (Standish, 2001). The reliance on a partner is problematic for those without them, and historically PWH are less likely to be married compared to those without haemophilia (Siboni, et al., 2009). The recent HERO study, however, reported most PWH are married or in long-term relationships and are satisfied with the support from their partners and family (Cassis, et al., 2014). Nevertheless, those OPWH who do have a spouse may face substantial grief and challenges if their partner passes away before them, but previously this was rarely seen due to their much shorter life-expectancy (Mauser – Bunschoten et al., 2007).

It has been indicated that a lack of perceived social support in OPWH is a significant risk factor for depression (Iannone et al., 2012) and that, in fact, social support acts as a mediator or a buffer in coping with illness or stress and promoting wellbeing (Bos, 2007). It is interesting that other than an article by Bos (2007), no other mention of the important role of family (other than partner), extended family, or friends has been made. Very little information was found about the role of family, friends, social, cultural, and spiritual groups or supports and their importance in helping or increasing wellbeing in OPWH.

It was found that, for the most part, the medical comprehensive care team were effective supports for PWH. However, they have little specific expertise with the older population with haemophilia, and there is some concern about the coordination between different medical services and specialists. The importance of psychosocial professionals, the NMOs, and the role of social support in the lives of OPWH were rarely mentioned. Leading authors in the field of PWH, including Franchini & Mannucci (2010), Young (2012), Bos (2007) and Dolatkhah et al. (2014), all recognise the need for new tools, recommendations and processes to address how services can effectively work with and care for OPWH. There are significant gaps in the literature, very few studies exist that provide evidence of the holistic wellbeing or psychosocial experiences of OPWH, and very few studies have been conducted using the voice of OPWH themselves. This is likely due to the fact that the existence of an elderly haemophilia population has only been realised relatively recently (Bos, 2007). The emerging literature that is available is typically framed from a medical point of view focusing on comorbidities and the haematological management of these. Throughout the review only two references from New Zealand could be sourced on PWH in general, the rest of the literature was from an international perspective. This shows that much more needs to be learned about the OPWH population in Aotearoa, New Zealand, as well as around the world. In response to these identified gaps in the literature, this thesis aims to provide an in-depth exploration of the unique issues and challenges facing older men with haemophilia and their

perceptions of support services in Aotearoa, New Zealand. The following chapter explains the procedure for the project, including the design, methodology and ethical considerations that took place.

CHAPTER THREE – DESIGN

As previously highlighted in the literature review, OPWH are facing new and unique challenges that have never been seen before. Thus, to address these gaps and concerns, a research project was carefully considered and developed. Mixed methodology was chosen in order to allow integration of both qualitative and quantitative data. This methodology was implemented using a three phase design consisting of an examination of the literature, a focus group, followed by a national questionnaire. These methodological and design choices are explained in detail below along with the ethical considerations which took place, but first the researcher's position within the research is explained.

Position as a Researcher

The researcher has worked for the HFNZ as an outreach worker for seven years. In this role she has observed older men with haemophilia reach 'normal' old age only to be faced with additional, but different, challenges to those experienced by men with haemophilia before them. A key driver for the research was the desire to better understand, and critically examine, this emerging situation from a psychosocial and holistic perspective. In doing so it was found that there were few answers from service providers, people with haemophilia, or from the academic literature and this led to great concern that more research and action was needed in this area. HFNZ supported this research project through partial funding, and therefore had some input into the research topic due to their desire to see the research undertaken and to see how they could better meet the needs of OPWH in New Zealand. With this support came the researcher's need to carefully balance her innate social worker perspective and desire to empower, listen to, and understand the voices of aging men with haemophilia via qualitative methods, with the needs of the HFNZ to obtain quantitative data and numbers and statistics which they could use to contribute to service development.

The researcher is positioned as a critical realist. According to this epistemological position, the researcher's role is to recognise the reality of the natural order, and the events and discourses of the social world. The critical realist acknowledges that to understand and change the social world we must identify the structures at work that generate those events and discourses (Bhaskar, 1989). Knowledge of this reality is socially constructed and conceptualised by the subjective views of the researchers and participants (Bryman, 2008). Consequently, mixed-methods research is well-matched with critical realism as it allows the researcher to integrate qualitative and quantitative research methods in such a way as to capture subjective knowledge and provide rigorous analysis of it (Patton, 2002). Insider verses outsider theory was also considered, by looking at whose definition of reality has more importance, the individuals involved in the relationship (insiders) or those who externally observe those individuals (outsiders). Olson (1977) argues that both perspectives give different insight on either objective or subjective reality and both of these views are important to gain a cohesive picture. In this case the researcher positions herself somewhere in between the two realities (insider and outsider). Although not an older man with haemophilia, I have worked alongside OPWH for the last seven years so do have an exceptional understanding of their lives and situations. I acknowledge that my personal experiences may influence my analysis and interpretation of the data, and therefore particular consideration was given to the research design, which is detailed below.

Three Phase Design

This mixed methods study followed an exploratory sequential design, with three phases. As is typical in this type of design, each phase informed the design and process of the subsequent phase, and the final inferences were based on the results of all phases of the study (Tashakorri & Teddie, 2000). By using a variety of research techniques and triangulating the results of each phase, the design was intended to comprehensively explore the experience of participants from many angles, as depicted in figure 1.

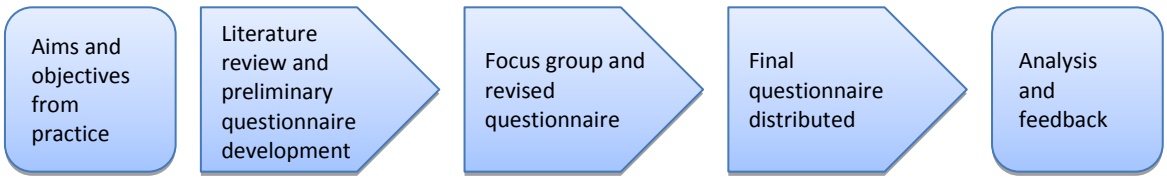


Figure 1. Research design phases.

Phase I – Literature Review and Preliminary Questionnaire

Phase one consisted of conducting an extensive review of the literature to identify what research had already been carried out and what the findings were. The review of literature was also completed to see what methodologies and designs had been previously used in research in this area, to identify controversy between authors and viewpoints, and to identify any gaps in the literature. The information obtained from the literature review (presented in Chapter Two) was used to inform the development of a preliminary questionnaire called the Older People With Haemophilia Questionnaire (OPWHQ). This was done by identifying key areas that leading researchers recognised as possible issues and challenges for this population, then putting these into question format and into a preliminary questionnaire.

Phase II – Focus group

A focus group was chosen in order to consult with a select group of aging men affected by haemophilia in Aotearoa, New Zealand. Focus groups allow researchers to listen to debate and gain a deeper understanding of viewpoints and experiences of participants (Walter, 2006). These elements were identified as being important for this research as the experience of older men with haemophilia in New Zealand had never been heard before. Therefore, it was important to create an environment where OPWH could openly and freely talk and share with one another and where the researcher could observe all of their interactions and opinions. The best way to achieve this was via a focus group. The data generated from the focus group was examined using thematic analysis (as described in Chapter Four), and was later triangulated with the questionnaire results and literature review in the discussion chapter (Chapter Six).

The focus group was 90 minutes long, and included five male participants aged 45 years or older, who had haemophilia, selected via purposive sampling (as later explained in Chapter Four). The purpose of the focus group was to facilitate discussion on the topic of older men with haemophilia in New Zealand, and to discover the participants' opinions, perceptions and experiences. A second aim was to read through the preliminary OPWHQ and obtain the participants' comments and feedback on it.

Intermediate phase – instrument development.

The focus group was useful in ensuring the OPWHQ aligned with the men's experiences and asked questions that were significant, relevant, and comprehensible to them (and the New Zealand context). All feedback from the focus group was used to inform a revised OPWHQ which was then emailed to focus group participants for one final round of feedback and suggestions. A variety of changes were made to the OPWHQ based on the focus group, including alterations to structure, wording, format, response categories, and some topics and questions being added or removed, as explained in detail in Chapter Four.

Phase III – Questionnaire

Questionnaires are known to be useful because they allow participants to complete them when and where convenient to them, therefore reducing burden on participants. Questionnaires also allow honest answers, especially to private behaviours and they are a cost effective means to gather data (Davidson & Tolich, 2003). Therefore, a questionnaire was chosen as a way to collect a significant amount of data from a wide range of people who lived large distances from one another in a short period of time.

The OPWHQ is an anonymous self-report questionnaire that was distributed to all men on the HFNZ database with haemophilia who were 45 years or over (n = 91). Follow up reminders were sent one month later, with a total return of n = 40. This OPWHQ focused on seven key areas over 64 questions: demographics; comprehensive care; physical health; work, income, and transport; social support; mental, emotional, and spiritual wellbeing; and age

related changes. A variety of response categories were used, including scales, open ended and closed questions (the questionnaire is described in detail in Chapter Five).

Mixed Methodology

Qualitative data was gained from the focus group and the open ended questions in the OPWHQ, and quantitative data was collected from the closed questions and scales in the OPWHQ. Creswell & Clarke (2007) state that by mutually illuminating both qualitative and quantitative data, a researcher can gain a broader and better understanding of the topic than could be achieved with either approach alone. Triangulation was then used to combine both qualitative and quantitative data collected as part of the research along with the literature so they could be mutually corroborated. Bryman (2008) states that triangulation is a good way to cross check each source of data against the other, to reinforce or reveal any gaps, and add greater validity to the research. Mixed methodology is a pragmatic approach that is responsive to the aims and objectives identified for this research project and aligns with the researcher's epistemological position.

Mixed methods are known to be useful to investigate a topic from many different angles, unpack its complexity, and gain deeper explorations into the topic (Tashakkori & Teddlie, 2009). Using mixed methodology allowed the researcher to gain an understanding of OPWH, and to ensure the inferences fit with the New Zealand context and experience. In terms of depth, a Masters' thesis is limited in time and scope. Therefore, mixed-methods analysis was considered to be the most appropriate for working within these restrictions, capturing primary qualitative data for depth, which allowed space for participants' stories and experiences to be heard, and gave richness and illustration to the research. This was balanced with a quantitative component to add a breadth of information across multiple perspectives and from a wide range of people.

The haemophilia community involves people from multiple disciplines, all with different needs: medical professionals, scientists, and NMOs often require data of a quantitative nature, while social workers and care professionals often greatly value qualitative work. The HFNZ supported this research and contributed to it financially with a requirement that quantitative data be generated that the people within the orginisation could use to make positive changes to their service delivery. Therefore, using mixed methodology was useful in reconciling the needs of all stakeholders and ensuring the research was useful for a wide range of groups and individuals. There was a fine balance between gaining acceptance with those who appreciate quantitative information in the form of statistics and figures, whilst still being able to engage in qualitative research which explores people's experiences and stories.

Another reason mixed methodology was selected was because it is a renowned approach to use when producing tools that do not already exist, thus ensuring their credibility (Creswell & Clarke, 2007). There was no other existing tool available which was deemed appropriate to investigate OPWH from a holistic perspective; hence the development of a new instrument was necessary. The Older Person With Haemophilia Questionnaire (OPWHQ) was created through this research, using multiple phases. The preliminary questionnaire was based on the findings from the literature review. Then changes and revision to the questionnaire were made based on the focus group discussion and subsequent email feedback from focus group participants. A mixed methodology was therefore considered to be the 'best fit' for this research in terms of addressing the aims and objectives. However as with all research there were many ethical considerations to take into account.

Ethical Considerations

This mixed methods, three-phase research design was carefully considered at the outset and throughout to ensure it followed ethical practice. Ethics approval was sought and gained from the University of Auckland Human Participants Ethics Committee (UAHPEC)

prior to the commencement of this study. There were some areas that provided more ethical challenges than others, as explained below.

Conflict of Interest and Bias

Some participants involved in the research were the researcher's clients, as she was the Northern outreach worker (social worker) for HFNZ. Therefore, a power differential could have existed between her employed work role, and her role as an independent academic researcher. To address this issue, participation in the focus group was sought only from individuals based outside the region where the researcher worked as an outreach worker (to ensure they would not be her clients). The OPWHQ was anonymous, which ensured the researcher did not know who had responded, and if it had been completed (or not completed) by one of her clients.

HFNZ provided partial funding for the study, so had some input into the research topic due to their desire to see the research undertaken. The study participants were aware of HFNZ's input into the research, as this was stated clearly on the focus group and questionnaire participant information sheets (PIS) (see Appendix A and C). Participants were made aware that HFNZ, (or any other organisation or individual), would not have access to the hard data or individual questionnaires but would have access to the findings.

Voluntary Participation and Informed Consent

The PIS for both the focus group and questionnaire stated that participation was completely voluntary and there would be no repercussions for choosing to participate or not participate in the research. Participants could ask questions at any time in order to find out more about the research process, and telephone numbers and emails for the researcher, her supervisors, and Head of School were provided to participants. At the beginning of the focus group all participants signed a consent form and these were collected and securely stored. For the questionnaire, participants were considered to have consented by filling in and returning their questionnaire (as explained in the PIS).

Anonymity and Confidentiality

For the focus group, participants were asked to respect the privacy of all members and not to disclose or discuss the other participants' identities or information. The OPWHQ was anonymous and participants were asked not to put any identifying details on it. In both cases, for the analysis, participants' information was de-identified and they were assigned a study code. Confidentiality was unable to be completely guaranteed in the focus group or questionnaire (as stated in respective PIS and consent forms), as there was a chance that people reading the findings may recognise some of the comments if they represented a unique or identifiable experience or point of view.

Harm to Participants

There were no risks or harm anticipated to occur for the participants of this research. However, it was recognised that some questions could have the potential to cause some discomfort, anxiety, or embarrassment to the participants. The PIS for both the focus group and questionnaire directed participants to contact their HFNZ outreach worker if they wanted to discuss or work through any of the issues raised by the research questions. In the case where the researcher was a participant's social worker the participants were directed to her manager.

Data Collection and Storage

Responses from the hardcopy questionnaire were entered into an electronic file and these, along with the questionnaire data, entered online. The audio recording files and the written transcription of the audio recording are, and will be, kept on the researcher's password-protected computer indefinitely. The original hard copies of the completed questionnaires along with the focus group consent forms are securely stored in a locked filing cabinet in the researcher's main supervisor's office at the University of Auckland's Epsom campus for six years, after which point they will be destroyed in compliance with the University of Auckland's secure destruction of research data procedures.

Use of Information

Participants were informed that the information gathered would be used as part of a Master of Social Work thesis being conducted through the University of Auckland. They were informed that the de-identified data and findings may be used to improve service delivery, and may be published in academic journals or discussed in presentations or at conferences. HFNZ (and other organisations and individuals) would not have access to the hard data or individual questionnaires but would have access to the findings.

This chapter has given an overview of the methodology chosen for the research, including details of the design structure. It has also outlined some of the key ethical considerations and dilemmas which were explored and addressed in the initial stages of the research. Chapter Four addresses Phase II – the focus group. It explores in detail the participant selection and the procedure of the focus group, and then move onto the data analysis and the findings of the focus group.

CHAPTER FOUR – FOCUS GROUP

A focus group was chosen as the preferred method for consulting with a select group of aging men affected by haemophilia in New Zealand. Walter (2006), indicates that focus groups allow researchers to listen to debate, gain a deeper understanding of viewpoints and experiences, and to quietly observe group dynamics, emotional responses, and both what is said and not said. In this research, the focus group was used in order to do just that, gain an understanding of participants' views, experiences, and perceptions. It was also used to get feedback on the preliminary OPWHQ, which was developed from the literature review, and once revised and finalised with help of the focus group, was used in the subsequent stage of the research project.

Methodology

Participants

Participation was solicited from Wellington-based men living with haemophilia who were members of HFNZ and over the age of 45. Wellington was chosen as the region from which the focus group participants would be sourced as it ensured that the participants were not the researcher's own clients, thus reducing risks associated with this conflict of interest. Focus group participants were identified with assistance from the HFNZ Manager of Outreach Services using purposive sampling. In purposive sampling, participants are selected because of their relevance with regards to understanding a social phenomenon (Bryman, 2008), and with the intention to recruit people with experiences that may be typical for other cases (Davidson & Tolich, 2003). The HFNZ Manager of Outreach Services was advised by the researcher to identify people who met the selection criteria and who represented a variety of ages, ethnicities, bleeding disorder severities, and experiences. The HFNZ distributed the PIS to the 14 people they selected, and five people chose to attend the focus group. Although five is a small number of participants for a focus group, Morgan (1998) indicates that small groups are suitable when participants are likely to have a lot to say on a research topic, if they are

very involved with the topic, or participants are emotionally charged by the topic, as was anticipated in this group.

Of the attendees the ages ranged from 45–72 years. One participant identified as Māori, one as Māori/NZ European, and three as NZ European. Four of the participants had severe haemophilia and one had moderate haemophilia. Three participants lived within a one-hour drive of an HTC, and the other lived further away.

Procedure

The five participants arrived at the hotel in Wellington for a dinner. A dinner was offered and paid for by HFNZ as an incentive for participation and to encourage casual introductions, discussion, and to get to know one another in a relaxed environment before the focus group began. Rapport building allowed for more open and extensive discussions in the focus group which immediately followed the dinner, as participants felt at ease and comfortable with one another. The dinner lasted one hour and the focus group lasted 90 minutes.

Participants were fully informed that their participation was voluntary, they could leave at any time, and they did not need to answer any question they did not want to. Participants were also informed about the reasons for audio recorders, and that their personal information would be kept confidential, although confidentiality could not be guaranteed between participants. Consent forms were then signed and collected and the focus group discussion commenced. At the conclusion of the focus group, participants were thanked, informed of the next steps, and a koha ($20 grocery voucher) was given to each participant as a token of appreciation.

Interview Schedule

There were three main aims for the focus group which aligned with the research question

1. To gather general opinions, thoughts and ideas from the participants about the issues and challenges they, as older men with haemophilia in New Zealand, faced
2. To gather general opinions, thoughts and ideas from the participants of the services and supports available to them in New Zealand
3. To read through the preliminary OPWHQ and obtain feedback on its relevance, length and accessibility.

The semi-structured interview schedule was important to ensure that each of these sections had equal time for deliberation and consideration. It also allowed the researcher to interject when necessary to ask follow up questions or to gain further clarity. Questions relating to the first two aims (above) were asked at the beginning of the focus group so the participants were not influenced by the findings from the literature or the preliminary questionnaire (which were later presented). These initial two questions (based on the initial two aims) enabled the researcher to get a real sense of the participants' views that were most prevalent or important to them. The first two aims were also important to highlight any necessary changes needed for the phase III OPWHQ, and to ensure it would be relevant to men with haemophilia in New Zealand. For the third aim, gathering participants' feedback on the preliminary OPWHQ, participants reviewed each question in the draft questionnaire and deliberated on the relevance, importance, wording, structure, response categories, and were then given the opportunity to provide additional comments.

All participants agreed to be contacted one month later via email to review the revised questionnaire and to provide one last round of feedback on it. This was done to ensure participants were given sufficient time, space, and multiple opportunities to evaluate and contribute to the OPWHQ. Following this, the researcher made the final changes, thus ending the feedback cycle.

Data Analysis

During the focus group a thematic analysis of the discussion was conducted. Thematic analysis is a widely recognised method for analysing qualitative data. It essentially involves the extraction of key ideas or themes from data (Braun & Clarke 2006; Bryman 2008). The researcher took note of the key themes identified in the men's narratives on poster paper during the focus group. Themes pertaining to each broad question (first issues and challenges, and then services and supports) were then reviewed collectively to ensure the ideas were accurately represented. Adjustments were made until the men mutually agreed that the themes were all correctly captured and reflected their viewpoints and the conversation that had taken place. For the first question, participants were also asked to individually choose the *one* unique challenge or theme they believed was the *most* significant to their life to enable the researcher to get a better understanding of the prevalence and seriousness of the issues (these are indicated in Figure 2). The focus group was audio recorded using two devices: a voice recorder application on the researcher's mobile phone and an audio digital recorder. This was to ensure one of the recorders would still capture the audio of the focus group if there were any technology glitches. The audio recording was later manually transcribed by the researcher into a Microsoft Word document.

Another level of thematic analysis was conducted on the data following the transcription of the focus group to enable a more in-depth exploration. This followed Braun and Clarke's (2006) six-step methodology as it complimented this research because of its theoretical flexibility and organic coding process. The steps of this methodology include;

1. Familiarizing yourself with your data
2. Generating initial codes
3. Searching for themes
4. Reviewing themes
5. Defining and naming the themes
6. Producing the report

After becoming familiar with the data, codes were identified, and these were then reviewed to identify themes. The number of times a particular issue was discussed (by the same or different participants) was also taken into account, as was the time taken to discuss an issue, the emotion generated by an issue, and if an issue was agreed upon by all participants or not. Braun and Clarke (2006) suggest that there are many ways to analyse data, and that researcher judgement is necessary to determine what a theme is and is not. They state that the 'key-ness' of a theme is not necessarily dependent on quantifiable measures, but whether it captures something important in relation to the overall research question. For example, 'physical health – decreased mobility and the negative impact on physical activity' was only briefly discussed in the focus group, yet all participants stated that it was a challenge in their lives and the researcher observed that it generated a lot of emotion. Three of the five participants later identified that physical health theme as the single biggest issue in their lives. This demonstrated the importance of taking all things into consideration when looking at themes.

The coding and themes were then reviewed by the researchers' supervisors for internal homogeneity within thematic categories - the extent to which the data that belong in a certain category holds together in a meaningful way. Also for external heterogeneity across categories – to assess the extent to which differences among categories are bold and clear (Patton, 2002). Thematic maps were created to visually depict the central and sub themes [see Figures 2 and 3].

Findings

Challenges and Issues Facing Older People with Haemophilia

The key themes which were identified in the focus group as the unique issues and challenges faced by older men with haemophilia in Aotearoa, New Zealand were: physical health declining, specifically decreased mobility and negative impact on physical activity; facing new medical problems due to comorbidities and the complexity this added to their

treatment and care; the high cost of their haemophilia treatment to the healthcare system, and the fear that this would prevent them getting the service, care, operations, and treatments they would need in the future; losing independence due to no longer being able to care for themselves and consequently spending more time in hospital. In Figures X below, *n* represents the number of participants who indicated that was the single *most* significant issue or challenge for them. Losing independence was a mutually agreed major theme however no one said it was the single most important issue for them.

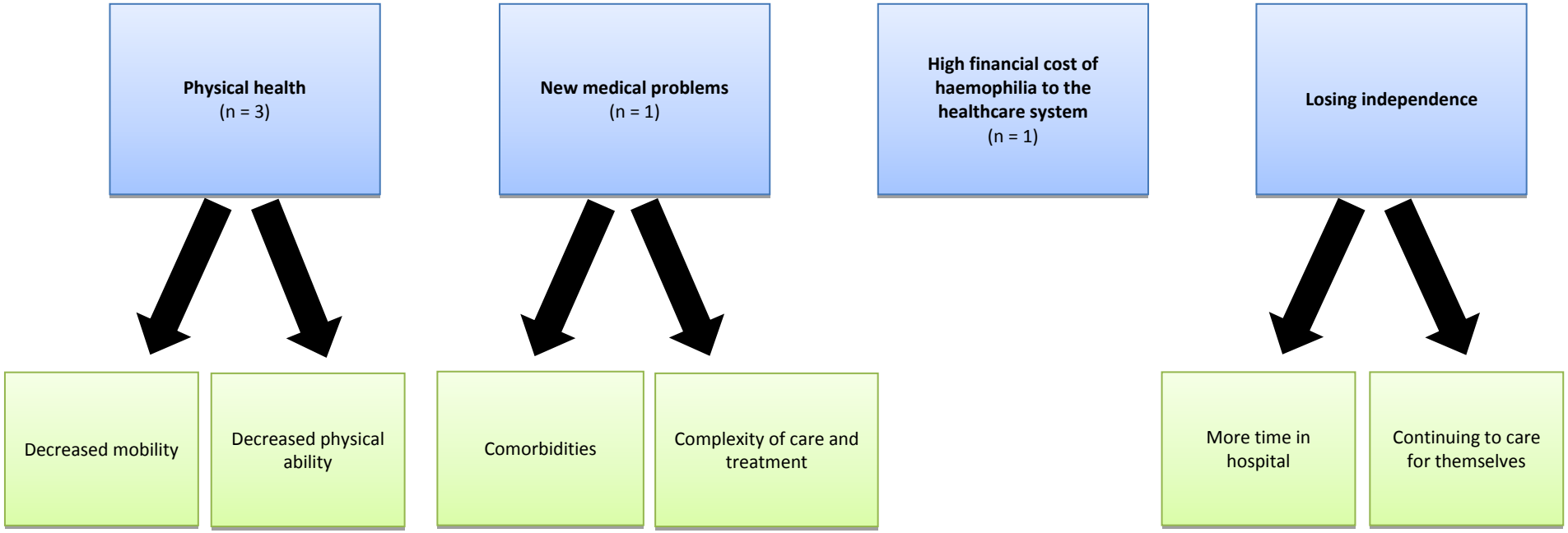


Figure 2. Thematic map of unique issues and challenges faced by OPWH as discussed in focus group.

Physical Health

Participants of the focus group described physical health, specifically decreased mobility and decreased physical ability as the most prominent issues for them. Three out of five participants said this area was their greatest challenge – making it the most prevalent problem overall. However, they spent very little time discussing this issue, perhaps because they all agreed and understood the significant impact of decreased mobility and physical ability in their lives.

Participants reported that their mobility had decreased, which had caused their movement to slow, so they were no longer able to participate in sports and activities, which they had once enjoyed. They expressed frustration with regards to knowing that exercise is good for them, with one claiming "it's good for my knee and good for my ankle", but often they were physically unable to participate in activity. For instance, another argued, "I want to get back to walking [down golf fairway]. I used to do it all the time, but I can't, not even with my walker." They reminisced and laughed about their "golf swing getting worse and worse" and how much their bodies had deteriorated over time, "[mobility will] continuously slide away to a point where we are immobile." All agreed it had negatively affected their wellbeing in a very serious way. However, there was also positive discussion around the importance and usefulness of attending their group physiotherapy or hydrotherapy classes in relation to helping with mobility.

New Medical Problems

The participants described the ways in which their bodies were "breaking down" and noted they were getting comorbidities, other diseases on top of their haemophilia, which one man described as "all sorts of weird stuff". They noted it was sometimes hard enough to deal with haemophilia without the complication of managing new medical problems because of aging. One participant stated "it's all the other things that now come on top of the challenges that we have already had to face in life."

After the discussion about comorbidities, their focus turned from new illnesses to how these new conditions led to complexities for their care and treatment. They indicated that all of their medical needs were once fully managed by the HTCs and that they had rarely engaged with specialists or GPs, but this could no longer be the case with the onset of comorbidities. They also felt that haemophilia treatment once fixed everything for them, however, now they will need additional medications for their new 'old age' conditions. As stated by one participant "for everything we used to go and see the Haematology Department and get a shot factor, and that was the answer to every problem we ever had." They realised that with these new medical problems came the need for new medical treatments and that "you can't give yourself another shot of factor and fix the heart problem." They also said that with new medical problems came a higher risk for needing medical intervention and surgeries which are a great concern for older people with haemophilia. One participant explained it by saying "it's those complexities around all the other things; gall stones, heart problems, liver problems…where they are required to go inside you [operate] to fix it, that's where it gets complicated."

Coping with multiple comorbidities appeared to present many challenges for the participants, particularly as there was not much knowledge or experience of this within the haemophilia community. The group anticipated that things were going to get a lot harder for them medically as they continued to age, and that they would increasingly need new or more specialists involved in their care, along with medications, and surgeries. The participants worried that all of these factors may become very complex to manage alongside their rare bleeding disorder. Comorbidities and the complexity of managing these new medical problems was a mutually agreed area of importance by all participants, with one participant saying it was the greatest challenge for him.

High Financial Cost of Haemophilia to the Healthcare System

There was worry among the group that the very high cost of their haemophilia treatment to the healthcare system would prevent them getting the service, care, operations, and treatments they would need in the future. They recognised that DHBs are "tightening the screws" and constantly analysing the money people with haemophilia cost the system. One participant said "at some stage I would imagine that they are going to start doing the math and doing cost benefit analysis - if we fix this guy's ticker is it worth the cost?" There was nervousness amongst the group as the participants expressed concern that it would be them, the older men with haemophilia, who would miss out first unless they could strongly "prove their case on financial terms." One participant shared, "I know two cases where people [with haemophilia] have gone to the surgeons and the surgeon says: you need this operation or replacement but we don't have the funds for it." Another participant was turned away, he claimed, "I've tried that [to get needed operation] before, but they wouldn't do it."

Two participants also commented that clinicians appeared to be talking increasingly about the cost of their haemophilia treatment in front of them, and some had seen the costs of their treatment displayed on a price list at the blood bank when collecting their treatment. This issue clearly affected their mental and emotional wellbeing, with words such as "nervous," "fear", and "worry" being repeated throughout the dialogue. One participant felt this was the biggest issue for him, three others were also concerned about it; however, it is important to note that one man had never heard or thought about financial cost of haemophilia to the health care system as being a problem before. This was the area of discussion that took up the most time in the focus group.

Losing Independence

The participants alluded to their fears of losing independence subtly throughout the focus group. They were concerned about the likelihood of having to spend more time in hospital as they get older, due to things such as: longer recovery times, more hospital visits

and stays due to comorbidities, and an inability to treat themselves. One participant in particular talked of his bad hospital experiences saying, “I resent going to hospital , I hate hospital ,” and they all agreed they would much rather stay in their homes and try to be as independent as possible for as long as possible.

The group also mentioned issues about being able to continue to care for themselves, specifically to be able to find and access or inject their veins in order to give themselves their haemophilia treatment. They were worried that, as their eyesight or veins deteriorate with age, they will have to rely on medical professionals to do this for them. This was especially a concern for those with severe haemophilia who need to treat themselves more regularly. They also talked of how more frequent hospital visits would create practical, financial and logistical challenges for them. Some men said they are already starting to experience this loss of independence, with one participant indicating, “I can’t do that [treat at home]. I can’t give it to myself because I’ll miss the vein all the time.

Perception of Services and Supports for Older People with Haemophilia

The key themes identified by focus group participants about supports and services for older men with haemophilia in Aotearoa were: the complexity of having multiple specialists involved in their care and that there is a lack of knowledge about haemophilia by medical specialists (other than those in the HTC); the positive experience they have had with their HTC and the fear one of the haemophilia team members will leave their job; and the importance of social connection with others with haemophilia. The overwhelming response to the topic was that participants felt they have great services and supports in New Zealand. One participant asserted, “as a community we are very spoilt.”

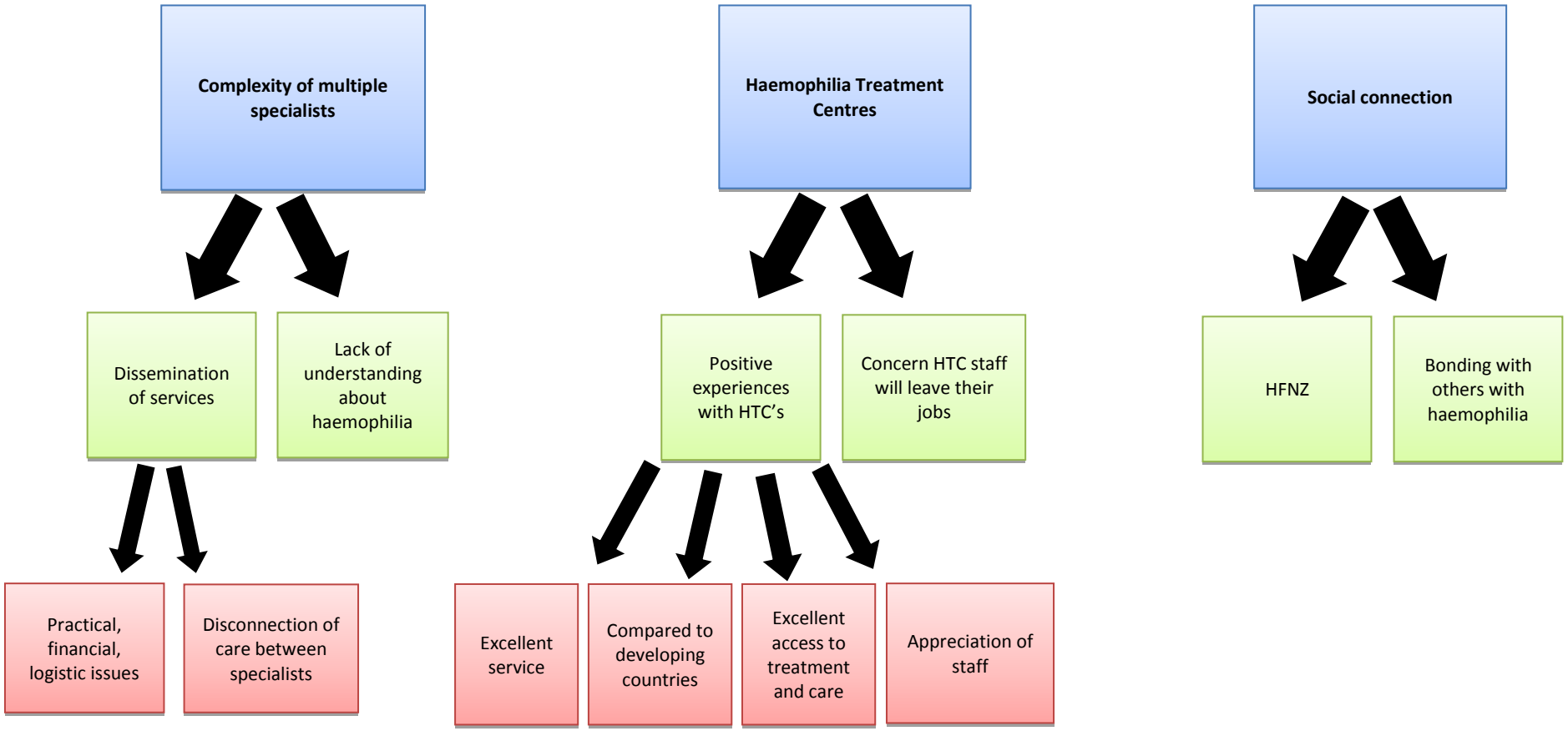


Figure 3. Thematic map of services and supports for OPWH as discussed in focus group.

Haemophilia Treatment Centres (HTCs)

All participants strongly agreed that they had great services, support, and a positive experience with their HTC. They conveyed very high praise for their specialist nurse and spoke fondly of their haematologists, physiotherapists, and surgeons. They compared the medical care they received in Aotearoa to developing countries such as Iraq and Cambodia, and were very grateful and appreciative for what they have here. "Compared to places like Cambodia we've got brilliant service. You look at the oldest people in Cambodia [with haemophilia] and their life expectancy is around 30" one participant remarked. They were also appreciative of the NZ comprehensive care model as another participant articulated,

> "I think we are very well looked after, I think that the combination of services that we have now - a Physiotherapist, in conjunction with a Haematologist, in conjunction with a Nurse Specialist. There are not many gaps in that …those people are committed to really looking after this community; it is really exceptional in Wellington."

All of the experiences that were shared in relation to their HTC were positive. Words such as "well-oiled machine", "caring" and "genuine" were used repeatedly. It was the access they had to their medical team that most impressed them, especially access to their nurses. One explained, "I had his [nurse] own private number and he'd just send me straight to the ward." Another participant said, "you ring [nurse] at any time of the day or night and he's virtually guaranteed to answer that phone within three rings. Three a.m. he's there." A third participant revealed, "when I came out of hospital with a bleed and I'd rung [nurse] and he came and picked me up and drove me to Kapiti, how amazing is that." This also extended to haematologists as expressed by another participant, "it used to be that I could call [haematologist] at home at three in the morning," and even an orthopaedic surgeon was spoken about fondly by one participant "you don't even make an appointment just turn up and I will see you now [orthopaedic surgeon]." The HTC was one of the first topics they discussed in the focus group (even though were being asked about issues and challenges, not services) and they continued praising their specialists throughout the discussion. All men

expressed similar feelings regarding their specialists; however, their experiences with emergency departments were not so positive, with three men saying it always took a very long time. “I never went to ED, you could go there and they’d just take so bloody long to see you” one said.

Their appreciation of medical staff was closely connected to another key theme: their fear and concern that HTC staff would leave their jobs, leaving gaps in services and people without specialist skills and knowledge. One participant stated, “if you take one of those things out of the equation…then we lose our point of contact.” They “rely on” and “need” their nurses and specialists involved closely in their care. They used words such as; “fear,” “nervousness,” and “risk” frequently in summing up their feelings on this topic with one sharing, “you live in fear that one of them will leave,” and another expressing, “I’m always really nervous that they are going to pull the plug on it, they are going to end it [physiotherapy service] or they will just quit and move on.”

Complexity of Multiple Specialists

There was some concern about decentralised services within the group (and for one member in particular). Participants said that because of their comorbidities and new medical problems they need to have other medical people involved in their care, with one participant stating “you’ve got to actually deal with different medical professionals and different departments.” This presents problems as the participants said that new medical professionals often lack understanding about haemophilia and the complexities of their rare bleeding disorder. One man shared his experience with the group, saying he had told his GP about having haemophilia and his GP said “oh I don’t know anything about that, what can you tell me about that?” They reminisced about times when they had to educate GPs, or specialists, about their condition. One participant had to do so at the emergency department at the age of eight. They were also worried that there may not be open and seamless communication between specialists, GPs, and their HTCs.

Although the group identified the issue as 'decentralized services', on further interpretation of the data, the majority of the conversation focused on the physical and practical implications of having services in different places whereas it "used to be a one stop shop - you'd just go to the hospital for everything." Now that services are in different locations it could mean more traveling, parking, stress and extra problems for people with mobility issues or who live in isolated places. One participant reflected that "it's hard if you are having mobility problems and you've got to travel all around the countryside to get the specialists." They were particularly worried for others with haemophilia who lived outside of the main centres of New Zealand and their access to quality care. "We hear horrible stories of guys in outlying areas that have been virtually forced to move to the cities because they just can't get the level of service or care," expressed one participant However, another said this concern was normal for any medical condition and not unique for the haemophilia population.

Social Connection

Throughout the focus group the men mentioned the importance of bonding and connecting with other people who have haemophilia. They talked about supporting and encouraging one another with things such as their group physiotherapy. They also mentioned the importance of their comradery and their ability to understand one another. They said it is often the "bonding and friendship" that helped them through the tough times. They worried about those who were not connected to others with haemophilia. One participant explained this by saying "[the] bunch of guys isolated out there on their own, doing their own thing, wondering why the world hates them." It seems that connecting with one another is often done through their own get-togethers, physical activity, physiotherapy initiatives, and sometimes through HFNZ.

Social connection facilitated through HFNZ was another discussion topic. The group recognised that HFNZ is "probably scratching their head" on how to best support the OPWH. Some participants felt HFNZ was not supplying the types of events that benefited and suited

older men, and that HFNZ may not understand how older men communicate and engage with one another. Participants' ideas included events specifically targeted to men's interests, and doing something alongside one another. One said "give us a beer and a fishing rod and we'll talk the day away." Other than suggestions for specific types of events and ways of engaging them, four out of five of the participants generally seemed to think HFNZ was supportive of them and their needs. One claimed it was the "best support we've ever had, and they are always looking at improving," and another said "services are bang on as far as I am concerned."

Preliminary Questionnaire

The final step of the focus group was to gather participants' feedback on the preliminary OPWHQ. Participants were given a copy of the preliminary questionnaire to work through, and then asked to deliberate on the relevance, importance, wording, structure, response categories of the questions and to provide any additional comments as mentioned earlier. There was debate about the questions on sexual activity, with one participant saying he would not be comfortable answering them; however, another participant said he would be very happy to answer them. The final consensus by the focus group was that the questions were important and should be included but that the researcher needed to be prepared that some people would not answer them. The participants were confused about the inclusion of spiritual questions; they didn't understand why they would be asked about spirituality or the importance of this area. These comments made the researcher think they had rarely been asked about spirituality or that they had never considered it a part of their wellbeing. Therefore, the questions were retained as it was seen by the researcher as an important area to explore further.

There was concern that only members of HFNZ would receive the questionnaire as that would exclude people who were unknown to HFNZ or who had asked to be removed from the HFNZ database or had chosen not to be members. This was recognised as a

limitation of the research and is discussed further in Chapter Six. The participants thought that, overall, the preliminary questionnaire read well and that OPWH would participate in answering it despite the length.

In total eleven questions were added to the questionnaire based on the focus group discussion and feedback on the preliminary OPWHQ. These were related to the limitations they experience due to mobility issues, whether they were involved with medical specialists other than the HTC, the coordination of care between HTC and other specialists, the cost of haemophilia to the healthcare system and the effects of this on their care, fears, independence, emergency departments, government financial benefits and support to attain these, and the ability of residential care facilities to cope with people with haemophilia. Two questions were also amended, and alterations were made to the structure and format based on the focus group participants' suggestions (i.e wording size, less open ended questions). All suggested changes were made and the adapted OPWHQ was emailed back to the focus group participants for final suggestions; however, there were none thus this concluded the feedback cycle.

On the whole, participants were engaged with the focus group and involved and interested in the discussions. There were two main leaders within the group, one participant who mostly discussed his personal experiences and two participants who remained relatively quiet except in relation to a few topics; however, they were attentive and nodded along in agreement throughout the conversations. It is interesting to note that on most occasions when services or supports were mentioned, the participants primarily took it to mean the medical team around them, along with some social support from people in the haemophilia community. There was no discussion of spiritual support, support from family or friends, or wider (non-haemophilia focused) social support. Another surprising finding was that two of their greatest fears had, to date, not been mentioned in any literature on OPWH. Specifically, their worry that the high cost of their haemophilia treatment to the healthcare system would prevent them getting the service, care, operations and treatments they would need in the

future, and the fear that one of their haemophilia specialists would leave their jobs, leaving gaps in services and people without specialist skills and knowledge of their haemophilia. The overwhelming positivity about their medical specialists was another interesting and unanticipated finding.

The focus group was a very useful process in terms of gaining a much deeper understanding, contextualization, and insight into the lives and perceptions of older men with haemophilia in Aotearoa, New Zealand. The focus group discussion resulted in a variety of changes to the preliminary OPWHQ. Chapter Five explains the questionnaire in detail along with the participants, procedure, and results.

CHAPTER FIVE – QUESTIONNAIRE

The unique issues and challenges faced by older men with haemophilia and their perceptions of support services in New Zealand were not only examined through the focus group but also through the use of a questionnaire sent out to members across the country. As described in the methodology chapter, the OPWHQ is a standardised questionnaire designed specifically for this research. It focused on the experiences of an older population of people affected by haemophilia from a holistic perspective, which had not been previously investigated. This chapter presents the methods behind the questionnaire construction and administration, and the findings resulting from the analysis of both the qualitative and quantitative questionnaire data.

Methods

The OPWHQ is an anonymous, non-experimental, and self-administered questionnaire. It is a standardised questionnaire in that all participants received the exact same questions presented in the same way. By using a self-report questionnaire participants could choose to complete it when and where it was convenient to them, therefore reducing the burden on them. This was especially important for a population characterised by disabilities, as there were likely to be times when the respondents were in pain or in hospital and unable to complete all the questions at one time. The questionnaire included potentially sensitive questions, or questions which may have caused some discomfort (e.g. about engagement in sexual activity, dependency on pain medications, and depression), thus the decision was made to use a questionnaire that enabled respondents to remain anonymous. The OPWHQ was also a cost effective way to gather data from a large, geographically dispersed sample. Please refer to Appendix D for the complete questionnaire.

The length of the questionnaire was of particular concern, as it was relatively long (64 items in total); however, the consultation with the focus group members (described in Chapter

Four) revealed that participants believed a lot of the older men with haemophilia would happily answer the questionnaire, regardless of length, due to its importance. The layout, font and format were all carefully deliberated upon. Size 13 was used for the wording, tick boxes were large and all questions were well spaced to ensure questions were clear and easy to read and follow. The OPWHQ also adhered to multiple other questionnaire writing guidelines such as using an attractive layout, clear presentation, and keeping scales the same where possible. Double sided black and white printing was chosen to reduce the cost, without compromising the usability of the questionnaire.

Instrument

The focus group informed the refinement of a preliminary questionnaire, which was initially derived from the existing literature. As stated, the OPWHQ contains 64 questions organised into key categories including: demographic details (four questions); comprehensive care (15 questions); physical health (18 questions); work, income, and transport (six questions); social support (three questions); mental, emotional, and spiritual wellbeing (eight questions); age related changes (five questions); closing questions and thoughts (five questions). Questions are asked in multiple ways, but where possible the same formatting and wording is used to better enable comparisons. The question response options include: a dichotomous 'yes' or 'no' response (11 questions), for example, "Have you ever seen a haemophilia physiotherapist?"; categorical response options (three questions), for example, "What services do you use within HFNZ?" with multiple tick box options; and Likert scales with three-point scales (three), four-point scale (one), and five-point scales (31 questions). Many of the questions requiring responses on a Likert scale are asked in exactly the same way, for example eight questions ask "How effective is [particular service] in addressing your needs?" with five response options ranging from 'very ineffective' to 'very effective'. Three questions ask "How supported do you feel by [a particular service]?" with five response options ranging from 'very unsupported' to 'very supported', and eleven questions ask "to

what degree does [particular issue] affect you?" with five response options ranging from 'not at all' to 'severely'. Eight short answer open ended questions are also included. In some instances, these are presented in conjunction with another question, for example after asking if they have received professional support for a particular issue, then asking "Which service provided this support?"

The OPWHQ also includes three scales previously validated through psychometric analysis. These were for life satisfaction, depression, and social support. Exploratory factor analyses were conducted to assess the validity of each of the theoretical constructs with the current sample. The internal consistency (i.e. reliability) of each scale and subscale was also assessed. As suggested by Miller & Acton (2009), this is recommended to see if the measure produces the same psychometric results under different conditions. All analyses were conducted with IBM SPSS software Version 22 and the outline of the scales and the results of the preparatory analyses follow.

Diener, Emmons, Larsen & Griffin (1985), created the Satisfaction with Life Scale (SWLS) which is included in the OPWHQ. This short five-item instrument is designed to measure global cognitive judgements of a person's satisfaction with life and subjective wellbeing. Importantly, in the past it has been used specifically with people experiencing serious health concerns (Pavot & Diener, 2008). Multiple studies have been conducted to assess the psychometric properties of the scale, and there is considerable evidence for the unitary structure and convergence validity of the tool (Pavot, Diener, Colvin & Sandvik, 1991). It is shown to have high internal consistency, high temporal reliability, and scores correlated moderately to highly with other measures of subjective wellbeing (Deiner et al., 1985). Each question is measured on a scale from 'strongly disagree' to 'strongly agree' and each item is then scored from one to seven, with one being 'extremely dissatisfied' and seven being 'extremely satisfied'. The ratings for each question are then added together to calculate

a total score of a person's life satisfaction. The possible range of scores is from five, which is a low satisfaction with life, to 35, which is a high satisfaction with life.

To assess the construct validity of the SWLS with the current sample, a Principal Axis Factoring (PAF) with an oblimin rotation (which is the method that allows factors to be correlated) was conducted on the five items with the sample data (n = 40; respondents are described in the findings section of this chapter) to obtain eigenvalues for each component in the data. One component had eigenvalues over Kaiser's criterion of one, which explained 69.65% of the variance. The four other components present were all very low, ranging from 0.61 down to 0.19. Observation of the scree plot also indicated that there was one clear construct (as anticipated). The Kaiser-Meyer-Olkin (KMO) measure verified the sampling adequacy for the analysis (KMO = 0.876 for the total score), and all KMO values for individual items were greater than 0.720, which is well above the acceptable limit according to Field (2009). The Cronbach's alpha for the SWLS with the current sample was 0.89, which suggests high internal consistency. With both reliability and consistency being high, the scale could be used as intended.

The Major Depression Inventory (MDI) (Olsen, Jensen, Noerholm, Martiny & Bech, 2003) is another scale included in the OPWHQ. This tool was initially created through collaboration between the Psychiatric Research Unit for the World Health Organisation and the Centre for Mental Health. The MDI is a ten-item, self-report mood questionnaire, although 12 items appear to the participants because two questions are scored differently with a part 'a' and part 'b'. The MDI items measure frequencies of depression or low mood over the previous two weeks. Studies have shown that it is a useful tool to measure the severity of depressive states, has high construct validity, and significant correlations have been found between the MDI and other depression scales (Olsen et al., 2003). It has also been found to have acceptable sensitivity and specificity (Bech, Rasmussen, Olsen, Noerholm, & Abildgaard, 2001). Each item is scored on a six point Likert scale from 'at no time' (0) to 'all

the time' (5), and the depression rating is scored by adding the total sum of the 10 questions. The scale can be used to measure the severity of depression, where a higher score indicates more severe depression. It can also be scored differently and used as a diagnostic instrument to diagnosis mild, moderate, severe, or major depression. This is done by generating an ICD-10 diagnosis, which is an international statistical classification of disease and related health problems (World Health Organisation [WHO], 1992).

To assess the construct validity of the MDI with the current sample, a PAF was conducted on the 12 items using an oblimin rotation. The initial analysis was conducted to obtain eigenvalues for each factor in the data. Three factors had eigenvalues over Kaiser's criterion of one which was concerning. However, one factor was the strongest with a total eigenvalue of 6.73, explaining 56.04% of the variance, the other two totals were 1.43 and 1.17. An examination of the scree plot also confirmed there was only one major theoretically distinct construct present, suggesting the scale was measuring responses as intended. The Cronbach's alpha for the MDI with the current sample was 0.92, which suggests very high internal consistency.

Zimet, Dahlem, Zimet & Farley (1988), created the Multidimensional Scale of Perceived Social Support (MSPSS). It is a brief and easy to administer self-report questionnaire assessing both the perceived availability, and the adequacy of emotional and instrumental social support across three sub-scales of family, friends, and significant other. It has been demonstrated that the MSPSS is a psychometrically sound instrument with good internal and test-retest reliability as well as moderate construct validity (Zimet, Powell, Farley, Werkman & Berkoff, 1990). The MSPSS can be used to assess a person's total social support or to assess family, friends, and significant other support separately. It contains 12 items rated on a seven point Likert scale with scores ranging from 'very strongly disagree' (1), to 'very strongly agree' (7). The ratings are then added to obtain a total score which indicates high, moderate, or low levels of perceived social support.

The construct validity and internal consistency of the MSPSS was assessed with the current sample. An initial PAF analysis was conducted to obtain eigenvalues for each factor in the data. Oblimin rotation was used as it was assumed sub constructs would be correlated. Three factors had eigenvalues over Kaiser's criterion of one and in combination they explained 90.32% of the variance. As anticipated, the item loaded onto the three theoretically hypothesized factors: family support (12.87% of variance), significant other support (68.85% of variance), and friend support (8.59% of variance). These three constructs could also be clearly seen in the scree plot, therefore there was high construct validity with the current sample. The MSPSS also had very high internal consistency ratings with a Cronbach's α of 0.98 for perceived significant other support, 0.94 for perceived family support, and 0.95 for perceived friends support. The full MSPSS scale was also highly reliable with a Cronbach's α = 0.96; therefore, as the developers intended, the scale can be used to assess both overall perceived social support and the three specific sub constructs.

Another previously validated scale which was used in the OPWHQ was perceived general health. There is not yet full standardisation of the measurement of perceived health status across OECD countries. Therefore, the measurement tool used in this questionnaire is an exact replica of the scale used in the New Zealand Ministry of Health questionnaires over the last decade (retrieved from: www.health.govt.nz). This measure was chosen to enable comparisons with the general New Zealand population if required. The question asks "In general would you say your health is" with response options on a five point Likert scale from 'poor' to 'excellent'.

To conclude the OPWHQ, five open ended questions (on top of the eight short answer questions) are presented to obtain the respondents' attitudes, opinions, and experiences in their own words, for example "What are your biggest fears for the future?" and "What are you most grateful for in your life?"

Participants and Procedure

The full population of older men living with haemophilia in New Zealand was targeted for participation in this phase of the research. The specific sampling criteria were as follows: respondents needed to be male, 45 years or older, have haemophilia, and be living in New Zealand. People with haemophilia are invariably male due to the hereditary nature of the condition, therefore only men were used for this research to ensure the results were specific to their issues and needs. Age 45 was selected as the lowest age, as it was an agreed age to represent men moving into older age in a haemophilia population (as discussed and decided in collaboration with HFNZ). HFNZ's database was used to solicit participation from the target population. The only alternative was to access multiple hospital databases (as there is not one national bleeding disorder database in the health sector); however, this presented many ethical issues as well as time constraints (as is explained further in the limitations section in Chapter Six).

An HFNZ staff member identified everyone on the HFNZ database who fit the research criteria (n = 91) and posted out the OPWHQ and PIS to all relevant individuals with a freepost envelope included for the return of the completed questionnaire. A link to an online Survey Monkey version of the OPWHQ was included in the PIS for those who preferred to complete the questionnaire online. One month after the questionnaires had been posted, a reminder notice was emailed to all potential participants with email (n = 54), and posted to the remaining people without email (n = 36) by HFNZ, and another reminder was included in an issue of the HFNZ Bloodline Magazine (a triennial national magazine for people with bleeding disorders in NZ). These reminders encouraged potential participants to fill in their questionnaire or to make contact if they needed a new copy. The choice to use two formats (post and Survey Monkey) was made in order to enhance convenience and accessibility for the respondents. It did, however, raise some issues in terms of people potentially answering the OPWHQ twice (this is also addressed in the limitations section in Chapter Six). The

closing date for returned questionnaires was set one month after the reminders were sent out and in total 40 men with haemophilia responded to the OPWHQ. With all questionnaires submitted, the next phase involved cleaning and preparing the data for the analysis stage.

Preparing and Cleaning Data

Once all the OPWHQ were submitted, the data from the hardcopy questionnaires and entered into IBM SPSS version 22 on the researcher's password-protected computer. For questionnaires that were submitted online through Survey Monkey, the data were downloaded, printed and entered manually. In both cases each respondent was de-identified and assigned a study code number. All documents were carefully checked to ensure the data had been correctly inputted, and later, random questions from each questionnaire were selected and cross checked with their corresponding paper copy. Maximum and minimum values were also calculated in SPSS for all variables to ensure the responses were within the appropriate range. During the cleaning of data, problems were found in eight questionnaires. These included respondents who had ticked two answers (instead of one) or in-between answers, written notes beside answers (which did not require notes), or provided answers that were illegible. All of these discrepancies were discussed with the researcher's supervisors and resolved. This process is further explained in the limitation section in Chapter Six.

Data Analysis

Quantitative Data Analysis

With data collected and cleaned, the following step was to conduct both quantitative and qualitative analysis. As the aim of the study was to produce a comprehensive descriptive profile of the unique issues and challenges experienced by men over 45 years of age with haemophilia and their perceptions of supports and services, descriptive statistics in the form of means *(M)*, standard deviations *(SD)* and, in some cases, (valid) percentages were calculated on all standardised quantitative data. When amenable, the findings were illustrated in graphs and tables which were created both in Microsoft Excel and Microsoft Word.

Qualitative Data Analysis

Qualitative data as well as the previously mentioned quantitative data were obtained in the OPWHQ. The qualitative data were obtained through the open ended questions. The majority of the qualitative questions were asked at the end of the OPWHQ so the writing component would not deter respondents from filling in the scales first (as feared by the focus group participants). There were five open ended questions, each with a different focus: issues and challenges, fears, support systems, what they were grateful for, and coping strategies. There were also eight questions throughout the questionnaire asking for short answer responses. In most cases the open ended questions were answered with very short sentences, single words or bullet points (with a range of one to 41 word responses). The qualitative data was analysed using thematic analysis, which is seen by Braun & Clarke (2006) as a useful method for identifying, analysing, and reporting patterns and themes within data. A similar process to the thematic analysis used for the focus group data (see Chapter Four) was undertaken with the qualitative questionnaire data. This included the following steps: familiarisation with the data, coding data, finding themes, reviewing themes and then placing themes into thematic maps and graphs. It was found that respondents often mentioned many different themes within one answer thus these were coded accordingly. As a result, the number of responses associated with different themes in the thematic analysis were often greater than the number of people who responded to the questionnaire. The coding and associated themes were then reviewed by the researcher's supervisors for internal homogeneity prior to interpreting the findings.

Findings

The following questionnaire findings are outlined in accordance with the initial aims and objectives of the research. After presentation of the respondents' characteristics, the findings that identify the unique issues and challenges faced by older men with haemophilia are presented by subcategory. This is followed by the findings that identify the respondent's

perceptions of their supports and services in Aotearoa, New Zealand. In both sections the quantitative findings are presented first followed by the qualitative findings.

Respondent Characteristics

As indicated, a total of 40 people responded to the OPWHQ. Their demographic details are illustrated in Figures 4 to 7.

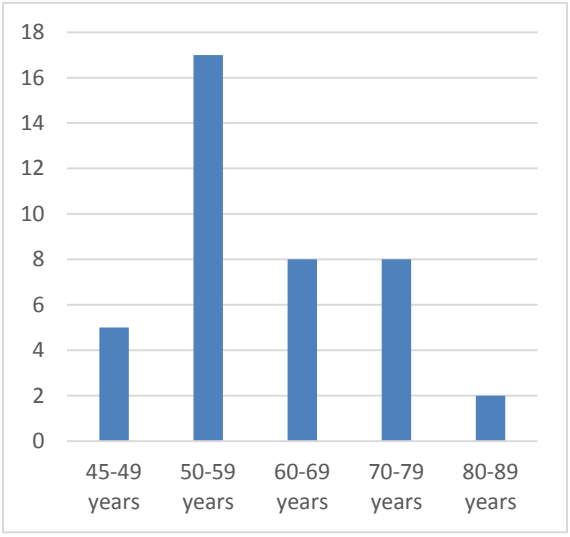


Figure 4. Age group of the respondents to the OPWHQ.

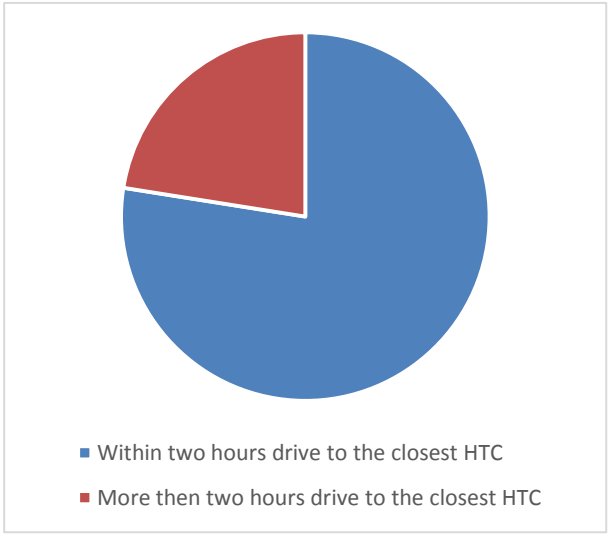


Figure 5. Living distance to the closest HTC for respondents to the OPWHQ.

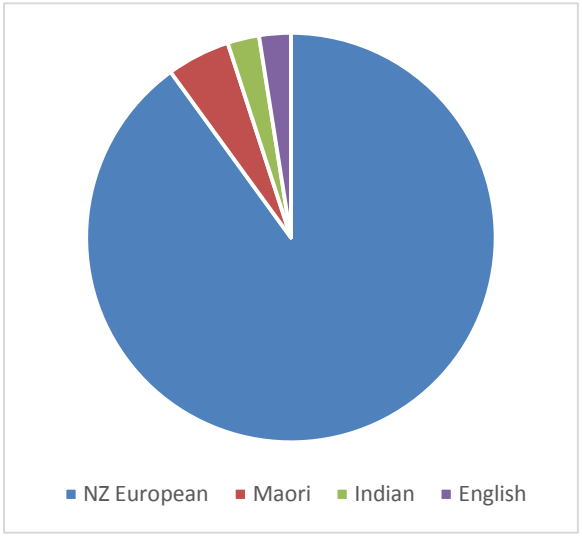


Figure 6. Ethnicity of the respondents to the OPWHQ.

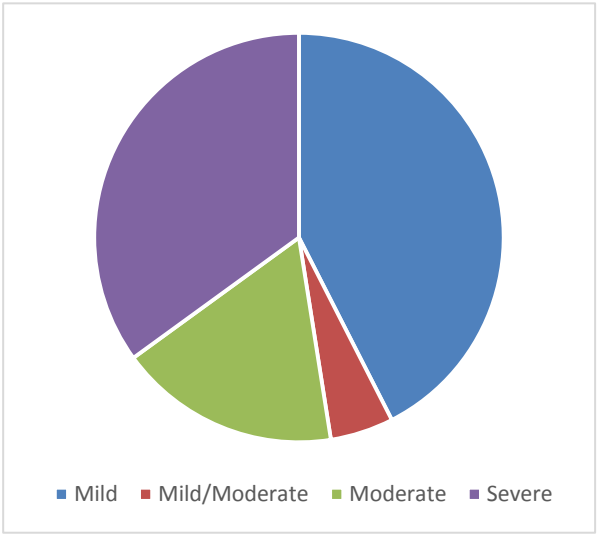


Figure 7. Haemophilia factor level of the respondents to the OPWHQ.

As can be seen, people from the age groups 45-49 up to 80-89 responded to the questionnaire. All severities of haemophilia were represented, but most experienced a mild level of severity. The vast majority (77.50%) lived within two hours of the nearest HTC, and

90% of respondents were NZ European, with only four people identifying with another ethnicity.

Unique Challenges and Issues facing Older People with Haemophilia: Quantitative findings

Physical Health

Mobility received the highest average rating with regards to the issue that had the greatest impact on the respondents' lives (as identified in the standardised question asking about the effect an issue has on a participant's life), this is shown in Table 1. On average respondents reported their mobility to be 'considerably' limited (30% participants). Decreasing physical ability was the second biggest issue for these men, shown in Table 1, and most respondents (32.50%) reported that haemophilia had a 'considerable' effect on their ability to engage in physical activity. The issues and challenges caused by limited mobility and declining physical ability are also seen in the thematic analysis of their greatest challenges (Figure 8) and biggest fears (9).

Table 1
Physical health issues faced by OPWH, and the prevalence and seriousness of these issues in their lives, from the OPWHQ findings

Physical health issues and challenges	*n*	Ratings of effect on life	
		M (out of 5*)	*SD*
Mobility limited	40	3.26	1.24
Physical activity limited	40	3.21	1.24
Experience of pain	40	2.83	1.13
Ability to engage in sexual activity	38	1.84	1.13
Dependency on pain medication	37	2.22	1.25

Note. *5 = the greatest impact on life.

Pain was another aspect that had a large effect on the lives of older men with haemophilia. On average, the degree of pain respondents felt was 'moderate', however there was a range of different experiences, from 15% reporting they experienced no pain to 5% who experienced severe pain. Respondents overall felt 'moderately' dependent on pain medication, although 43.20% stated they were not at all dependent, and no one indicated they were

severely dependent. Over half of the respondents (67.50%) had comorbidities (other medical conditions on top of their haemophilia) and of those, 58.30% indicated their other condition affected them more than their haemophilia. Just under half the respondents (44.70%) reported that haemophilia had affected their ability to engage in sexual activity in some way. On average it only had a 'slight effect' on their lives, and no one indicated their ability to engage in sexual activity was 'severely' affected.

Respondents rated their diet as healthy *(M = 3.61, SD = 0.72)* and only 2.60% indicated their diet was 'unhealthy' or 'very unhealthy'. Most respondents rated their weight as 'normal' (60%), the average was *(M = 3.24, SD = 0.627),* and no one indicated they were 'very underweight'. In total 42.50% of the respondent's perceived that their general health was on average 'good' *(M = 2.87, SD = 0.89).* However despite their general perception of their health, declining physical and medical health (especially into the future) was identified as a theme pertaining to both the second biggest challenge and second biggest fear (Figure 8 and 9 respectively).

Age Related Changes

The degree aging affected the independence of older men with haemophilia was low to moderate *(M = 2.35, SD = 1.27)* with the majority (32.50%) reporting it only had a 'slight' effect, but notably 10% indicated it had a 'severe' effect' on their life. Most respondents reported that their ability to self-infuse their own haemophilia factor treatment had not changed as they had gotten older (82.40%). However, of the small percentage who indicated it had impacted on their lives (17.60%), the effect on them was 'moderate' on average *(M = 2.91, SD = 1.14).* Of those it affected 36.40% indicated that changes in their ability to self-infuse their factor treatment had a 'considerable' effect on their life. Although the respondents did not specifically indicate aging was having a big effect on their independence, there were issues and fears closely related to this topic which did affect them and this was identified as a

theme in the open-ended questions (see Figure 9). Most of the respondents reported they had fallen since the age of 45 years-old which had led to a haemophilia bleeding episode (60%).

Work, Income and Transport

Throughout the OPWHQ the question was asked ‘to what degree does [this issue] affect your life?’, and 30% of respondents felt that haemophilia ‘moderately’ affected their working life as can be seen in Table 2. Overall, the extent to which haemophilia had negatively impacted on their working life was the issue that had the third most severe effect on participants, and was also mentioned as a challenge in open-ended responses (Figure 8).

Table 2
Work, income and transport issues faced by OPWH, and the prevalence and seriousness of these issues in their lives, from the OPWHQ findings

Issues and challenges	*n*	Ratings of effect on life	
		M (out of 5*)	*SD*
Work	40	2.85	1.21
Financial impact	40	2.18	1.20
Transport	40	1.53	0.93
Cost of H to the healthcare system	40	2.50	1.40

Note. *5 = the greatest impact on life.

The degree to which participants believed the cost of their haemophilia treatment to the healthcare system impacted on the medical service and care they received was ‘moderate’ on average (as shown in Table 2) however, there was a wide range of answers, and the most common was that this issue did not affect them at all (30%). The degree to which haemophilia impacted on them financially was also ‘moderate,’ however, in this case 40% also indicated it did not affect them at all. Respondents reported that they only had ‘slight’ transport difficulties to get to medical appointments. Notably, most respondents stated they had no, or only slight, transport difficulties (90%). Few respondents received a benefit other than superannuation (15%), and those who did stated that they felt ‘fairly supported’ by the government to obtain that benefit *(M = 2.64, SD = 0.809).* Finances were the third most

prevalent issue mentioned in response to the open-ended question asking about challenges in participants' lives (Figure 8).

Mental, Emotional and Spiritual Wellbeing

Extremely low levels of depression were found in older men with haemophilia in this study. On the six point MDI scale, the mean was 0.36 *(SD = 0.60).* Using the ICD-10 scoring (for diagnosis of depression) all of the respondents were classified as having lower than 'mild' depression (none of them achieved the threshold score to meet the criteria for diagnosis of depression). The average SWLS score was *4.83 (SD = 1.34)* indicating that most respondents were 'slightly' satisfied with their lives. There was a variety of personal coping strategies respondents had developed in their lives to increase their mental and emotional wellbeing, as can be seen in Figure 10.

Respondents felt moderately connected to their ethnic culture or heritage *(M = 2.78, SD = 1.19),* and spirituality, in general, was moderately important to them *(M = 2.43, SD = 1.48).* Notably, the largest portion said spirituality was not important to them at all (37.50%). The majority of respondents did not consider themselves to be religious (45%), followed by those who were unsure and answered 'sort of' (35%).

Unique Challenges and Issues Facing Older People with Haemophilia: Qualitative Findings

In this section some of the key themes that were identified through the thematic analysis of the open ended questions in the OPWHQ are presented. The graphs show the frequency of themes identified across all open-ended questions.

In the qualitative data, mobility was the most frequently reported challenge in the lives of older men with haemophilia in New Zealand (nearly double any other issue), as can be seen in Figure 8.

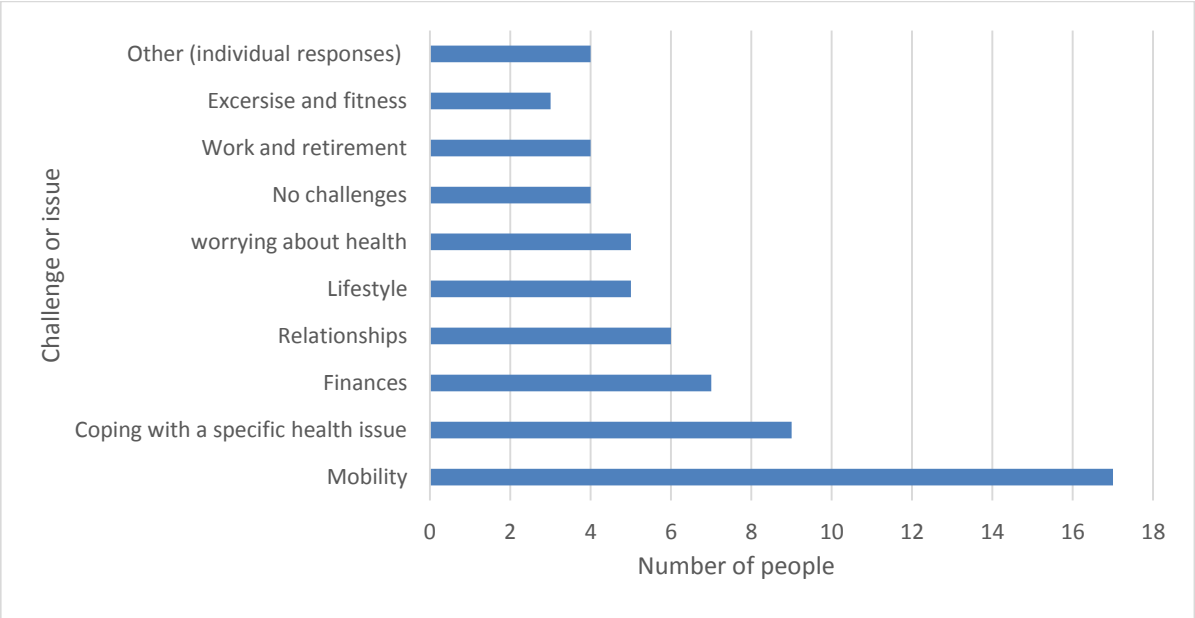


Figure 8. Biggest issues, challenges or concerns in the lives of OPWH, from the OPWHQ findings.

Some participants mentioned mobility in general, while others referred specifically to waning joints; for example, one man who wrote about his "crumbling hip" and another, his "deteriorating knee and hip joints". One participant also wondered "how much longer will I have good mobility without relying on 'aids', at what point will my knee or any other joint stiffen up?" The most prevalent fear of older men with haemophilia was also reduced mobility or complete immobility, as can be seen in Figure 9

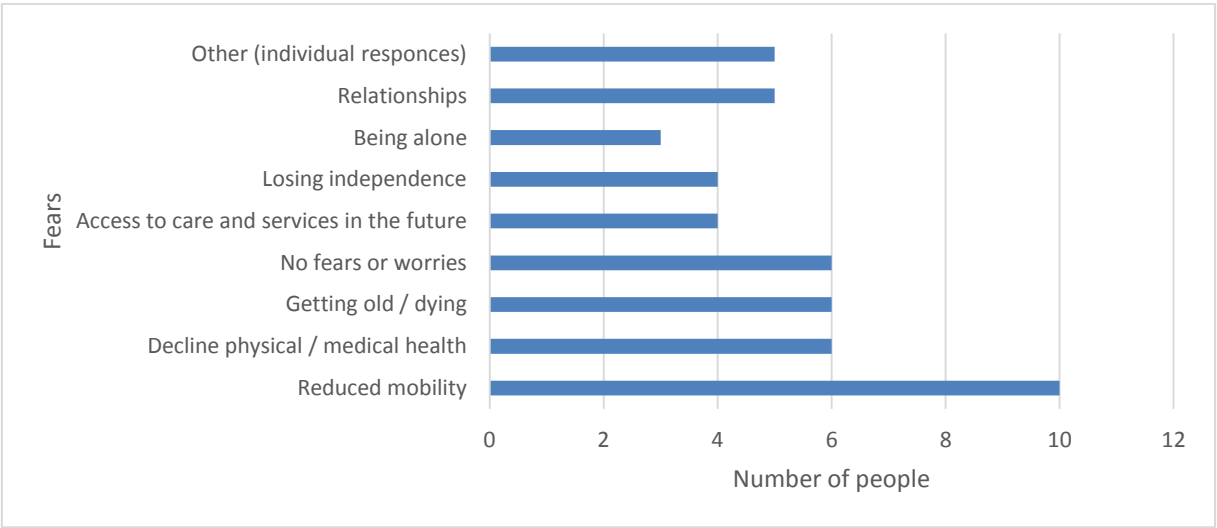


Figure 9. Biggest fears for the future, from the OPWHQ findings.

The following comments illustrate some of the fears participants had in terms of mobility: "losing mobility," "not being able to get around because of my hips and knees,"

"not physically being able to do anything," and the "possibility of someday being unable to walk." Interestingly (considering mobility issues and fears), physical activity, exercise, and sport were identified by many respondents as tools or personal coping strategies which help them in their lives (Figure 10). One respondent said it was "keeping active at all times," and another "'if you don't use it you lose it' is my motto."

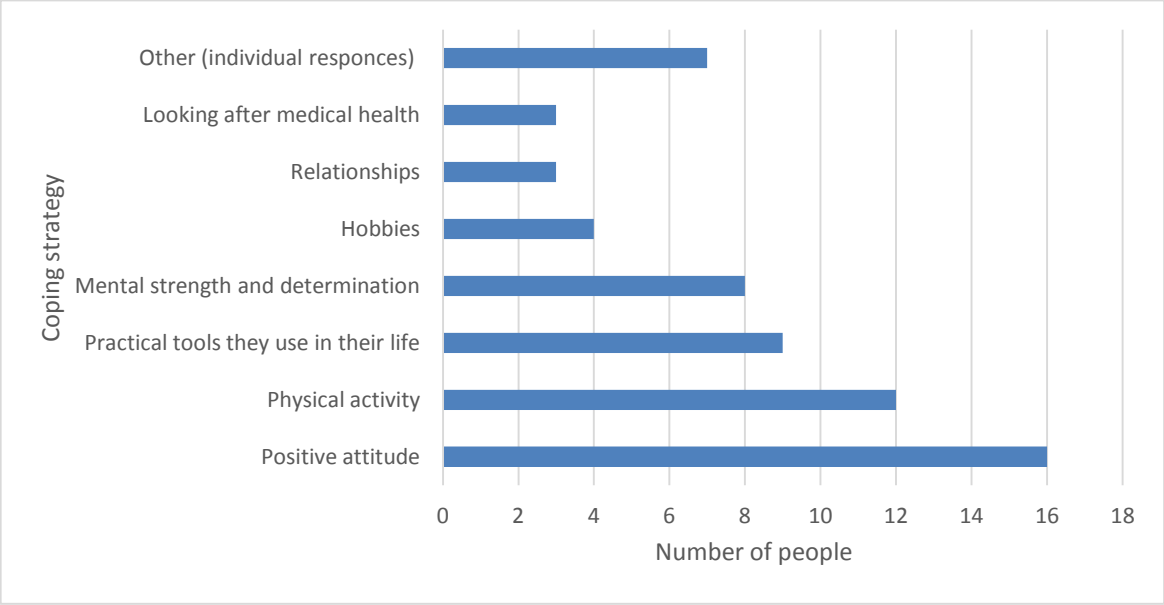


Figure 10. Personal coping strategies to keep OPWH going, from the OPWHQ findings.

Declining medical health was identified as a major challenge and issue, and also a major fear for respondents (see Figure 8 and 9). Many worried that their health would deteriorate or worsen prematurely, and that they would be unable to deal or cope with it. A few worried about proximity to hospital and medical services, and many about their ability to "maintain good health for the future." On the contrary, the analysis of responses to the question asking what they are most grateful for in their life revealed that medical health was the second most prevalent theme, with 14 people being grateful for their reasonable health, and some simply thankful for "being alive" (see Figure 13). Medical health was also briefly reported to be a positive coping strategy for some (see Figure 10).

The analysis of the open ended question asking about challenges in life revealed that finances were the third most prevalent issue. Seven people mentioned finances in terms of

“staying on top of bills”, ensuring financial stability, and “financial security into retirement” (Figure 9). On the contrary people were also very grateful for the material and financial assets they had (as shown in Figure 13). Some examples of this were; owning a home and being able to maintain it, having enough money to survive, and owning a car. Both sets of data show the importance of financial security, and the worry it presents for older men for haemophilia when it is not present.

The most frequently mentioned coping mechanism in terms of helping OPWH to maintain wellness was having a positive outlook and attitude to life, with 16 people mentioning it. Specific sentiments that were repeated within this category were: don't worry, keep positive, and, there are others worse off. Some quotes which showcase participants’ positive outlook on life include: “inspiration and vision and finding a purpose and creating a better life”, “after life’s setbacks and disappointments being able to pick yourself up and go on”, “appreciate what I have and not what I haven’t got”, and “just get on with it, don't feel sorry for yourself.” The third most frequently mentioned way to maintain wellness and reduce stress was the personal practical strategies that they had implemented in their lives. Every respondent mentioned different strategies they used ranging from community work, keeping busy, breathing techniques, driving a fast car, and pacing themselves.

Another prevalent coping mechanism to help OPWH to maintain wellness was relying on, and building, their mental strength (see Figure 10), which included independence (e.g., “trying to be as independent as possible”). Fear of losing independence was also mentioned in the question asking what their greatest fears were, with four people indicating that losing independence and “not being able to care for myself” in the future was a big fear (Figure 9). Closely aligned with this last theme was another major fear about the aging process, specifically, getting older, staying alive, and dying.

Perception of Services and Supports for Older People with Haemophilia: Quantitative Findings

The description of findings now moves from challenges and issues in OPWH lives, to identifying the services and supports being used by older men with haemophilia in New Zealand, along with their perceptions of the effectiveness of these services and supports.

Comprehensive Care

On the whole, respondents had a 'very positive' experience of coordination between care from different specialists (as shown in Table 3), and no one reported their experience of coordination between specialists was either 'very negative', or 'negative.' All respondents reported having a GP (100%), and on average respondents found their GP 'effective' in meeting their needs, with only 16.20% indicating their GP was 'very ineffective' or 'ineffective'. Approximately half of the respondents had been to A&E since they had turned 45 years of age (55%) and found the service 'effective' on average in meeting their needs. The respondents thought that old age residential care facilities on average would cope well with older people with haemophilia *(M = 3.28, SD = 1.30).* Interestingly, the second most common response was that residential care facilities would cope 'very well' with looking after them (23.10%).

Table 3
Use of comprehensive care support services, and the users' perceptions of the effectiveness of these services , from the OPWHQ findings

Support service	Service use (n)	Effectiveness rating	
		M (out of 5*)	*SD*
HFNZ	N/A	3.87	0.95
HTC	N/A	4.10	1.19
GP	40	3.46	1.04
Specialist physiotherapist	29	4.04	1.34
A&E (since age 45)	22	3.23	0.79
Coordination of care	20	4.25	0.79
Pain support	16	3.27	0.70
Emotional support	14	3.43	1.28
Mobility support	12	N/A	N/A
Sexual support	1	3.00	0.00

Note. Participants were not asked about service use with HFNZ and HTC as it was assumed they were all engaged with these services, and the effectiveness of mobility support was also not asked about.
*5 = the greatest impact on life.

On average, respondents felt their HTC was 'effective' in meeting their needs, with the majority (77.50%) finding their HTC 'effective' or 'very effective'. Most respondents also felt 'supported' by their HTC staff (M = 4.48, SD = 0.87), with only 5% reporting staff were either 'unsupportive' or 'very unsupportive'. Most respondents had seen a specialist physiotherapist (72.50%), and of those who had, most (52%) felt that the service was 'very effective' in meeting their needs. The thematic analysis of the open ended question asking what other conditions they had, on top of haemophilia, uncovered that the most common comorbidities were blood pressure problems (n = 5), HCV or the effects of it (n = 5), diabetes (n = 4), arthritis (n = 4), high cholesterol (n = 3), and cancer (n = 3). Eleven other individual conditions were also mentioned.

Some respondents had seen a professional for emotional concerns (35.90%), and of those who had, they felt the service on average was 'effective' in meeting their needs. Emotional support came from a wide variety of services including counsellors (n = 3), GPs (n = 3), church (n = 2), and other services which were mentioned on a one off basis included:

community, consultant, employer, ACC, and hospital. Over half of the respondents said they had not received professional help to deal with mobility issues (66.70%), which is surprising considering the prevalence and severity of their mobility issues (as presented in Table 3). Of those who had received support, the sources were: physiotherapists (*n* = 3), hospitals (*n* = 3), or orthotics centres (*n* = 2). Other individuals mentioned the disability resource centre, area health board, and orthopaedic specialists as services they had used. Just under half of the respondents (44.70%) reported that haemophilia had affected their ability to engage in sexual activity in some way, however only one person received professional support to help him cope with this issue, and that was provided through his GP. This was the least frequented support service both in terms of the amount of respondents that had used the service (one person), and the perceived effectiveness of the service. Less than half of those who experienced pain had been to see a specialist to help them deal with the pain (44.40%), and on average found the service was 'effective' in meeting their needs. Of those who had received professional help to deal with pain issues, the most common service used was the HTC (*n* = 7), GP (*n* = 4), pain team (*n* = 4), and other individuals wrote the hospital, liver transplant centre, ACC, and physiotherapist.

HFNZ

Most respondents felt supported by their HFNZ outreach worker (42.10%), (*M* = 4.05, *SD* = 0.99). Notably, only 7.90% felt 'unsupported' or 'very unsupported'. Below is a table outlining the percentage of people who used the services provided to them by HFNZ. Respondents could tick as many services as they used. As can be seen the Bloodline magazine and outreach worker support were used the most by the respondents and substantially more than all other services. In fact, ten of the sixteen services were used by less than half the respondents, and 'needs grants' were a service used by no one at all.

Table 4
Percentage of people who use the services provided to them by HFNZ, from the OPWHQ findings

HFNZ service	Service user rate
Bloodline magazine	73.00
Outreach worker support	70.30
Conferences	35.10
Connect socially with others	32.40
Workshops and camps	29.70
Panui (newsletter)	29.70
Masters group	24.30
Supportive footwear programme	24.30
Website	21.60
Exercise programme	21.60
Educational resources	16.20
Advocacy	10.80
MRG events	10.80
Defensive driving program	5.40
Piritoto	5.40
Needs grants	0.00
Other	0.00

Note. Piritoto (the HFNZ Māori group), is open only to Māori members (of which two identified as in the demographics). Therefore, there are only two possible participants, both of which use the service.

Respondents indicated that HFNZ was effective in addressing their needs as shown in Table 3, and only 5.10% found the service ineffective or very ineffective. When asked if HFNZ could make changes to improve their services the largest response was "maybe" (55.30%), and most of these respondents indicated all organisations can improve but provided no explanation of how this could be done. Some people (21.20%) indicated HFNZ could change or improve its service.

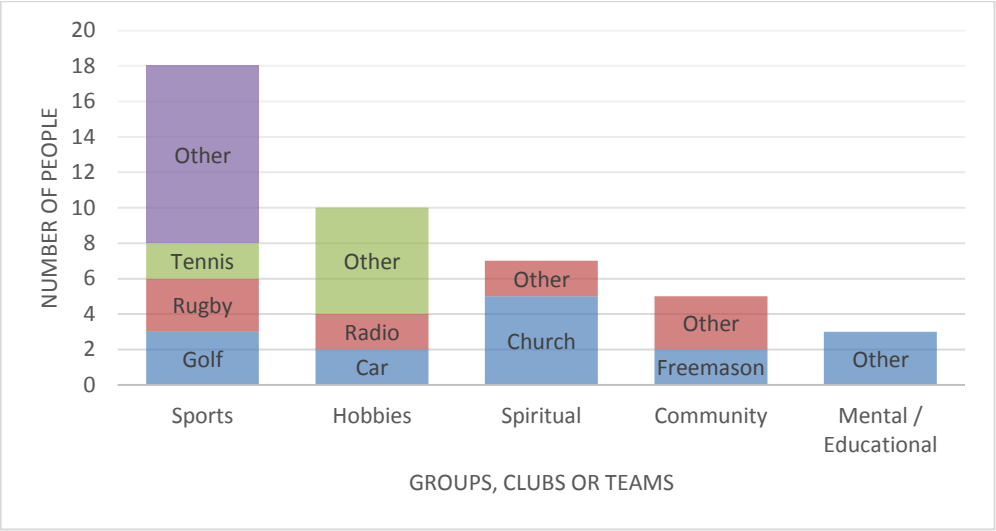
The main theme (mentioned by five participants) was that HFNZ needs to improve its communication through more contact and interaction, be more inclusive for all to access, and review different ways to engage with members. One man said HFNZ needed to provide "more interaction with older members," another said "while HFNZ has a wonderful service, it has got to be more open, and its services not only available to those members who attend all its

functions." The second theme (mentioned by three participants) was that HFNZ needed to review the services they provide, with one stating HFNZ need to "continue to evolve their service delivery model to ensure that it keeps up with the needs of an evolving community," and another saying it is "time for overall review of all services to find better ways to work." There were also a number of one-off suggestions by individuals.

Social Support

The responses to the validated seven point MSPSS which took into account support from family, friends, and one's significant other showed that on average most respondents strongly agreed that they had support from these people in their lives *(M = 5.71, SD = 1.23).* Individually, when the scale was broken into the three sub-categories and the mean for each separate category calculated, it was found that; 'significant other' offered the most support *(M = 6.06, SD = 1.48),* followed by 'family' *(M = 5.84, SD = 1.23)* and the lowest support was from 'friends'. Significant other and family were also strongly represented in the thematic analysis in terms of their most effective support system (see Figure 12).

In terms of social support, respondents were 'moderately involved' with social groups, clubs, or teams *(M = 2.85, SD = 1.46).* Altogether 43 groups were attended by 24 men (many of the people involved in groups attend more than one). The types of groups, clubs or teams they were involved in can be seen Figure 11. Respondents generally believed they participated in the same number of social groups as others their age *(M =2.49, SD = 1.00).*



Note. Other means there were individual answers in that category that were only mentioned by one person.

Figure11. The types and number of groups OPWH are involved with, from the OPWHQ.

Perception of Services and Supports for Older People with Haemophilia: Qualitative Findings

In this section some of the key themes which were identified through the thematic analysis of the open ended questions are presented. The graphs show the frequency of themes identified across all open-ended questions.

The thematic analysis of the open ended question asking about the most effective support system in participant's lives clearly showed that their biggest support was family, as shown in Figure 12.

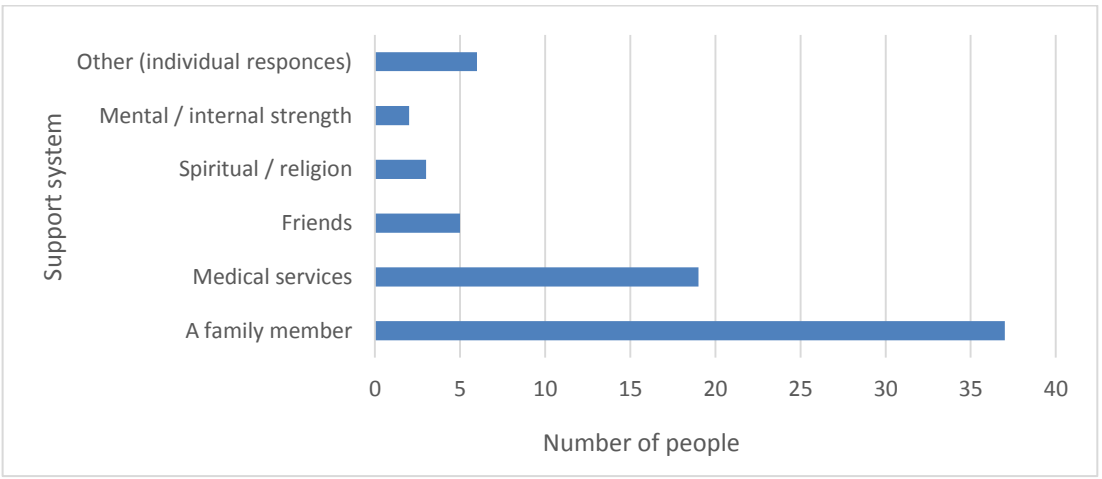


Figure 10. Most effective support systems identified by OPWH, from the OPWHQ findings.

Family was mentioned 37 times, which was nearly double the frequency of any of the other answers. The ‘family’ theme included 17 people saying their partner was their most valued support person, with one saying “my wife, without her I'm nothing,” and another suggesting "I think being in a loving relationship…is the most important as it transforms people’s lives, confidence, self-worth." Family in general was also mentioned regularly, two mentioned siblings and one mentioned their parents. The aspect participants were most grateful in their lives was also family (figure 13).

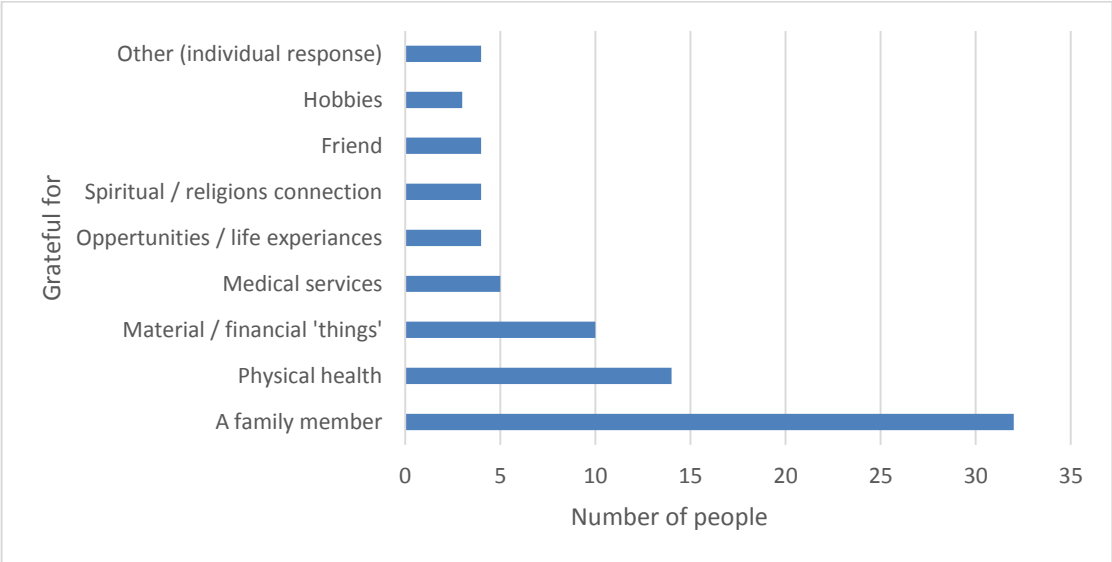


Figure 13. Things that OPWH are most grateful for, from the OPWHQ findings.

In this question the comments about partners were once again the most emotive. One man wrote, “deep caring from my wife and children of me” and another wrote “love of my family and wife.” However, relationships were also seen as a challenge or issue by six people, who discussed challenges associated with raising children, maintaining relationships and “facing life alone.” Five people also included comments about relationships in relation to their greatest fears (see Figure 9). Four people indicated friends as something they are most grateful for and five people mentioned them as important supports. However, the frequency of comments relating to friends was much lower than for family relationships.

Medical services were identified as the second best support system in participants’ lives, and in most cases (n = 13) this referred to support from the HTC. The medical services

were also represented in the responses about what the respondents were most grateful for in their life, with 4 specifically being about the HTC. One respondent stated he had "excellent service by HTC staff over the years."

Most of the comments about the respondents' experiences of HFNZ's services were very positive (18 of the 29 comments). The majority of these were not related to specific issues or reasons, with many just indicating the services were "very good," and "excellent." There were also general comments about the staff being "the kindest people in the world," "supportive," and "caring," and two people mentioned their specific outreach worker. There were two themes in the positive response category (other than the general positive comments about service and staff). The first theme reflected the advocacy and support the foundation provided, specifically regarding the HCV action and treatment products (n = 4). One respondent wrote "they gave excellent assistance during the Hep C compensation saga and also during my 12 months on interferon to combat the Hep C." The second positive theme was the communication from HFNZ, and the way they gave feedback, responded to different needs and gave updated information to members (n = 4), with one man stating "they are efficient and always respond." In contrast, there were five negative comments. Two participants did not feel supported by their specific outreach worker, and three participants believed there was poor access to and communication by HFNZ. One said HFNZ is "extremely difficult to access and phone calls are not returned when asking for help. Only young people are wanted on committees and the culture." The other said "HFNZ are there when you ask for support, but if you feel worthless inside you will not ask, and they will not connect." One individual also commented about his inability to attend events due to full time work and family responsibilities. There were also six neutral comments, with four of these from people who stated they did not need or did not use the HFNZ service, for instance, "I do not have a need for my Outreach Worker and prefer she give her time to others. I do not need the services."

The findings from the questionnaire illustrate that there are some large issues and challenges for older men with haemophilia, with mobility posing the biggest challenge or issue in their lives, as shown in the responses to both the standardised and open-ended questions. Decreasing physical ability, the effect haemophilia has on their working life, pain, and reliance on pain medications were also big issues that were identified. Medical health was also shown to be a serious challenge and something that caused participants fear (especially looking into the future). Although the respondents did not specifically say aging had a big effect on their independence, there were issues and fears closely related to this that did affect them. The quantitative and qualitative findings also demonstrated that the respondents had positive mental and emotional health.

Of the 21.20% who said HFNZ could change or improve its service, the main theme (mentioned by five participants) was that HFNZ needed to improve communication with members through more contact and interaction, be more inclusive for all to access, and review different ways to engage with members. One man said HFNZ needed to provide "more interaction with older members," another said "while HFNZ has a wonderful service, it has got to be more open, and its services not only available to those members who attend all its functions." The second theme (mentioned by three participants) was that HFNZ needed to review the services they provide, with one stating HFNZ need to "continue to evolve their service delivery model to ensure that it keeps up with the needs of an evolving community," and another saying it is "time for overall review of all services to find better ways to work." There were also a number of one-off suggestions by individuals.

Family was the biggest support for this group, (specifically partners, spouses or significant others) and something for which they were very grateful for. For those that were involved with medical professionals other than their HTC, respondents indicated the experience of collaboration between these two services was very positive. The support provided from participants' HTC, physiotherapist, and HFNZ was effective and the respective

staff supportive. However, there was very little support for issues to do with mobility and sexual activity.

This chapter has presented the methods behind the questionnaire construction and administration, and the findings resulting from the analysis of both the qualitative and quantitative questionnaire data. Chapter Six presents the triangulation and discussion of findings which have been presented thus far, and also the limitations to the research, recommendations, and considerations for the future.

CHAPTER 6 – DISCUSSION AND CONCLUSION

This thesis has aimed to provide an in-depth exploration of the unique issues and challenges faced by older men with haemophilia, and their perception of support services available in Aotearoa, New Zealand. This chapter discusses and triangulates the content which has been presented thus far. It considers the literature review from phase I (Chapter Two), the focus group discussion and outcomes from phase II (Chapter Four), and the phase III questionnaire findings from the previous chapter (Chapter Five). The findings are discussed in relation to the initial aims and objectives of the research, highlighting the important and interesting findings. After a discussion of the limitations of this study, this chapter concludes with recommendations for practice, the implications for this research for future use and concluding thoughts.

Unique Challenges and Issues Facing Older People with Haemophilia

An aging population with haemophilia has never been seen before (Franchini & Mannucci, 2010), therefore there is limited research, discussion, and literature available on this group of people and what they are facing, especially from a holistic perspective. Thus, the initial aim of this thesis was to explore the unique issues and challenges facing older men who are living with haemophilia in Aotearoa New Zealand and specifically, to explore both the prevalence and seriousness of these challenges and issues. Overall, the most serious and prevalent issues and challenges found for participants were decreasing mobility and physical ability, pain, medical health and comorbidities, work, and cost the of haemophilia treatment to the healthcare system. Other key findings of interest in the area of unique issues and challenges included being able to access their own veins to give themselves haemophilia treatment, losing independence, and positive mental and emotional wellbeing and outlook on life.

Physical Health

The most prevalent, serious, and frequently reported issue shown throughout the research was the loss of mobility and decreasing physical ability. OPWH in New Zealand who participated in the research spoke and wrote at length about the negative impact that decreased mobility and physical activity had on their lives. Many noted that mobility issues had caused their movement to slow and they were no longer able to participate in sports and activities which they had once enjoyed. Not only was it a significant issue and challenge for participants, but it was also a very real fear in the lives of many. They worried that in the future they would lose further mobility and physical ability or become completely immobile. These challenges are also echoed in the international literature, with many noticing decreasing mobility as being one of the most common complications for OPWH (Valintino, 2010) and, as explored by Chen et al. (2015), one that can have a serious effect on QOL. Interestingly, physical activity, exercise, and sport were the second most frequently used personal coping strategies in the lives of OPWH, as a way to help maintain their wellness. It seems problematic that physical activity was a major challenge for them, yet at the same time, it was often used as a way of coping with life's problems. Therefore, some OPWH may find that as their ability to engage in physical activity further reduces (due to age and mobility issues), they will also lose one of their valued coping mechanisms to maintain their wellness. This could have significant psychological and physical consequences for those individuals, and adds further complexity and challenge to the area of mobility and physical activity.

Another prevalent and often reported issue for this population was challenges regarding medical health problems, both haemophilia and non-haemophilia related. This presented concerns in terms of practical management and treatment of conditions, and was also a major fear and worry for their future. As well as single medical problems, there were also challenges for those with comorbidities, as discussed by authors in the haemophilia field. Clear evidence exists that demonstrates co-morbidities are becoming more common in those

living longer with haemophilia (Franchini & Mannucci, 2010) and they are becoming more challenging for both the healthcare providers and the patients to manage (Miesbach et al., 2009). The new medical issues and complexities brought about by comorbidities was a mutually agreed area of concern for focus group participants. They talked at length about the challenges comorbidities presented for them in many areas of their lives (mental and emotional, physical, logistical, and financial) and the large negative effect comorbidities did or could have on their lives. It was found that the comorbidities this group is currently experiencing are in line with those experienced by OPWH around the world (see Hermans et al., 2014). Research participants primarily talked about comorbidities and medical issues as a fear and a worry for future deterioration, and not so much as a concern in their current lives. Interestingly, over half of the participants indicated that their other condition affected them more than haemophilia did. This is possibly because haemophilia is a lifelong condition that they grew up learning to manage and understand, whereas the comorbidity was a secondary condition which most often occurred later in life and involved new medical specialists, medications, and knowledge for the OPWH, hence the worries and fears associated with it. Despite the concerns around participants' medical health, both now and in the future, overall the participants reported their current general health to be 'good'. In fact, good general health was one of the things participants were most grateful for in their lives. Therefore, medical health provided serious challenges and fears for some OPWH (especially into the future), but for others they were extremely positive and grateful for their current good health, and it did not pose problems or concerns for them.

Pain also emerged as a salient topic in this research with regards to prevalence of people who experienced pain to some degree, and the prevalence of those somewhat reliant on pain medication. Although a large percentage of people in this research study did acknowledge pain in their lives, this is not unusual for OPWH, and the amount and severity of pain experienced in this study was similar to that identified in other studies (e.g. Genderen, et

al. 2006). Despite the pain experienced by participants, it was not a topic that was mentioned in any qualitative feedback (focus group or questionnaire), although there were multiple written and oral stories of accidents, bleeds, operations, hospital stays, and problems with mobility which one would assume would often lead to extensive pain. An explanation for this may be that pain is a norm for this population, and something that they have always had to deal with, hence, they minimize or choose not to focus on it or discuss it. Thus, it is very important for both medical and support people to note that just because pain may not be talked about by some OPWH does not necessarily mean that pain does not exist for them.

Age Related Changes

An area highlighted in the focus group that provided some challenges to participants was the ability of OPWH to be able to find their veins to intravenously inject their haemophilia factor replacement treatment themselves. The focus group discussions revealed that some participants were already struggling (or unable) to do this. Others were worried that, as their eyesight or veins deteriorate further with age, they will have to rely increasingly on medical professionals to do this for them (and the associated problems of regular hospital visits). This was especially a concern for those with severe haemophilia who needed to treat themselves more regularly. This was not a prevalent issue indicated in the questionnaire, with the vast majority reporting that their ability to self-infuse their haemophilia treatment had not changed as they had gotten older. However, the small percentage of respondents who now struggled to find and inject their own veins reported that the effect it had on their lives was, unsurprisingly, moderate to considerable. As the haemophilia population continues to age, there will be more and more OPWH who will also face old-age issues (i.e. vision and dexterity problems), which may mean they will also struggle to administer their own haemophilia treatment. This problem, accompanied by the new and very real possibility of support people (e.g. family) passing away before the OPWH that they support, adds further complexity to this issue of treatment at home, as it is often family who help with this when

the OPWH cannot do so themselves (this issue is explored further in the family support section). Lambing & Kachalsky (2009) mention the many effects that no longer being able to self-treat could have on a person's life, yet no research to date has been conducted on this area.

Independence was an issue mentioned, in sometimes subtle ways, throughout the research. It was a mutually agreed concern for the participants of the focus group- both the likelihood of having to spend more time in hospital as they get older, due to things such as longer recovery times and comorbidities, and the inability to treat or care for themselves. Multiple fears and worries relating to losing independence were also present throughout questionnaire respondents' written dialogues. On the other hand, the standardised questionnaire scale revealed that respondents had very low concern that aging was affecting their independence. A possible explanation for this (due to the contrary findings from the qualitative data) is that many OPWH may currently still feel independent on the whole, but there might be multiple areas in their lives where their independence is very slowly reducing, perhaps without them realising or naming it as such. Another consideration may be that they were expressing worries and fears about losing independence in the future, but it may not be an issue affecting them right now. There were brief mentions about independence in the international literature in terms of accessing veins (Lambing & Kachalsky, 2009) and going into residential care facilities (Allen & Kachalsky, 2010), but nothing about how to maintain independence, and the psychosocial and psychological consequences of losing independence, which could be substantial to the lives and wellbeing of OPWH.

Two issues that were not seen to be serious or prevalent challenges for most OPWH in this study, but still important to mention, were residential care facilities and falls. Transitioning to residential care facilities and rest homes was an area with very limited literature or research, however the small amount that is available suggests this may be a growing area of concern as OPWH will increasingly need care outside the home and hospital

setting as they live to an older age. This may bring worry and fears for this population (Allen & Kachalsky, 2010). Interestingly, this was not a fear that was shared by the questionnaire respondents, as on a whole they thought that residential care facilities would cope well or very well in caring for OPWH. Also, it was an issue that was not mentioned in any of the qualitative questionnaire responses. This does not mean that residential care facilities will cope well with OPWH, but rather that OPWH feel confident in the process and do not seem to be worried about it. This may be because very few PWH have gone into residential care to date (due to previous shorter life expectancy) and so there has been little feedback or discussion in the haemophilia community about the success or failure of OPWH transitioning into residential care. The other issue that was important to note was falls. In agreement with existing literature, this research demonstrated that OPWH are prone to falls which have led to a haemophilia bleed. Therefore, this is an important area to be taken into account because of the, often severe, negative consequences of falls for OPWH. Nevertheless, this research did not show anything new or surprising in this area.

Work, Income and Transport

Although employment was rarely discussed or written about by the participants in any qualitative aspect of this study, participants still rated that the impact that haemophilia had had on their working life was a substantial and serious challenge for them. In fact, work was found to be the area with the third highest impact (seriousness) on the lives and wellbeing of OPWH in this study. This finding was consistent with the international literature which explored at length the issues faced by PWH in the workforce. Bos (2007) reported that PWH participate in less full time work compared to an age-matched population for a wide variety of reasons, and Rolstad (2014) extended this to say that haemophilia has had a negative effect on the working life of OPWH, and that they have had fewer opportunities in the working world. It is not only important to acknowledge the difficulties many respondents have had in terms of work, but also important to understand the wider implications, and to realise that employment

issues could be a root cause for things such as troubles with finances, psychosocial wellbeing, and relationships (Iannone et al., 2012).

The focus group participants spent a considerable amount of time talking about the cost of haemophilia treatment and care to the health care system, specifically in terms of worry that the high costs may prevent them from getting the services, treatment, and care they would likely need in the future. Some participants also felt guilt that the high cost of their treatment prevented others from getting treatment or care they needed. These feelings were amplified by the perceived increase in clinicians talking about the cost of their treatment in front of them. Further, questionnaire respondents believed the cost of haemophilia treatment to the healthcare system impacted on the medical service and care they received, at least moderately, and five indicated that it severely affected their service and care. Therefore, many OPWH must be aware that haemophilia treatment is very expensive. According to the Pharmaceutical Management Agency in New Zealand (PHARMAC, 2015) approximately 250 patients use treatment each year, costing the New Zealand healthcare system approximately $25 million per annum.. It is surprising that this issue was not mentioned at all in the international literature, compared to this study, as it appears to have an evident effect on the lives of many OPWH in New Zealand who participated in this research. It may also be an important issue to research for those abroad without private insurance, or those living in countries where there is no or very limited treatment. It is clearly an area where more exploration is needed as some OPWH who participated in this research are thinking about it, internalising it, and discussing it with others. This could lead to OPWH trying to reduce their use of haemophilia treatment to save money (as is happening with one focus group participant), which could be very dangerous for their medical wellbeing.

Issues of less significance, but still of note were related to income and finances. Only two studies have been conducted on PWH and money (other than those focused on insurance), and both articles took a different stance on whether money was a problematic area for PWH

(Dolatkhah et al., 2014; Siboni, 2009). In the current research it was found that finances had a moderate effect on the lives of participants. However, participants had very mixed responses in terms of income and financial security. Those without financial security found it a large challenge and likely need support and help in this area (i.e. budgeting advice) and those who had financial security were very grateful. Transport was found to be an issue with little to no impact on the lives of OPWH.

Mental, Emotional and Spiritual Wellbeing

This research showed extremely low depressive tendencies in older people with haemophilia in the study sample. Using the Major Depression Index (12 item, 6 point scale) it was found that respondents had an exceptionally low average score (*M = 0.36, SD 0.60),* and using the ICD-10 scoring mechanism it was found that all of the respondents were under the 'mild depression' mark. Mild depression in the ICD-10 is indicated by a score of 50, and in this research no respondent scored higher than 29, and nearly half (*n* = 19) scored zero, therefore answering 'at no time' to all 12 questions. This is a very surprising finding, particularly when compared to the international literature, where the vast majority of prior research has shown elevated levels of depression in OPWH (Canclini et al., 2003). The two questions that scored the highest and pushed the total depression score upwards were "have you felt lacking in energy and strength?" and "have you had trouble sleeping at night?". However, it is not unusual for older people, specifically those with a chronic condition to have trouble with energy, strength, and sleep (Hardy, 2010). These are also issues known to significantly affect people with HCV (Carlson, Hilsabeck, Barakat & Perry, 2010), which at least five respondents indicated they had.

An area that may have contributed to the positive mental and emotional wellbeing of participants in this study was their positive attitude and outlook on life. OPWH in this study clearly indicated their most common coping mechanism in life was their positive outlook and attitude, and other common strategies they used were building their resilience, and their

internal and mental strength. These, along with the wide range of support people and systems identified, and the handful of people who said they had no fears or worries for the future, and no issues or challenges in their life, show there may be the other factors that are contributing to this group's wellbeing, and perhaps in some cases superseding depression. Overall, depression in this group was extremely low and quite inconsistent with any prior international research. One possible explanation for this is response bias, which is further explored in the limitations section.

Perception of Services and Supports for Older People with Haemophilia

OPWH are a new and growing group that face many unique issues and challenges, as previously discussed. Due to this it is important that OPWH are supported through these life challenges by a variety of support services and people. Therefore, the second aim of the thesis was to explore the supports and services available to OPWH by identifying them, describing OPWHs perceptions of these supports and services, and gauging how effective they are for OPWH. Overall responses regarding services and supports utilised by the study cohort in Aotearoa were very positive. The most effective supports, as discussed below, were family (specifically significant others), the HTC, the coordination of care between healthcare providers and specialists, physiotherapists, and wider sources of social support such as social groups and others with bleeding disorders. Services to aid in problems with engagement in sexual activity and mobility were rarely accessed, and HFNZ was seen as an effective service, but participants also suggested some areas HFNZ could address to better meet their needs.

Social Supports

Without doubt, participants felt that family was the best support for them, and the thing they were most grateful for in their lives. Although this research uses the word family, it is clear that for those with a significant other, that person provided the most effective support as they were most frequently mentioned and most highly rated. Further, significant others were described with the most emotive language. This research aligns with international

literature which states most PWH are married or in long-term relationships and are satisfied with the support from their partners and family (Cassis, et al. 2014). This however raises very real concerns for those OPWH without a significant other, and poses questions about where and how those people access support at the deepest level. It also raises concerns for the respondents that stated that relationships were a challenge for them, or brought them fear. Another worry is that as OPWH are living longer than ever before, there is a likelihood that their significant other or closest support people may die before them and leave massive gaps, both in their hearts and in terms of their care, which is a concern also shared by Rolstad (2014).

Social participation in groups, clubs, and teams was found to be an important part of life for OPWH. The participants attended a considerable number and variety of groups including sport, hobby, spiritual, community, and educational (in order of prevalence). In approximately half the cases these men participated in two or more different groups, and perceived themselves to be just as involved in groups, clubs and teams as others their age (without haemophilia). The groups that the most people participated in were sport groups, which is surprising considering the challenges of waning mobility and physical ability that were also identified in the research. However, it seems these men have chosen wisely about appropriate sports which they can partake in, despite their limitations (i.e. golf), and for other sports perhaps they have chosen alternative ways to be involved in groups which do not jeopardise their health (i.e. some respondents said they were involved with rugby and tennis clubs, which does not necessarily indicate they play the game, but perhaps they are involved in other ways such as management or coaching). Both Bos (2007) and Iannone et al. (2012) argue that there needs to be much more research into this area as, to date, little is known about OPWH, social groups, and connection. Based on the findings of this current research, and international literature identifying that mobility, physical ability and pain were issues for OPWH, one may assume that OPWH would be less involved in social groups, however this

was clearly not the case in this study. Some OPWH must truly value their social participation to be attending so many groups (despite the other challenges they face). This may be because of their resilience, positive attitude, and determination to let nothing stand in their way as shown in their positive coping strategies. It may also because people with haemophilia have had a lifetime of conversations with both medical and social supports about what their strengths and limitations are and have taken a considered approach when deciding what groups, clubs, or teams are appropriate and sustainable for them to participate in.

Throughout the focus group, the men mentioned the importance of bonding and connecting with other people who have haemophilia. In fact, this was the only support (other than medical) that the focus group talked about. Therefore this topic was identified as another type of social support that was valued by respondents. They discussed the importance of supporting and encouraging one another with their bleeding disorder journey and challenges, and also mentioned the importance of comradery and their ability to understand one another. HFNZ was one identified way of connecting people with bleeding disorders with one another, however some participants felt HFNZ were not supplying the types of events that benefited and suited older men, and that HFNZ may not understand how older men communicate and engage with one another. These comments were also in line with participants' recommendations of ways HFNZ could change to better meet the needs of OPWH, with the most commonly reported ways including: being more open for all to access; looking at different and appropriate ways to engage; and more interaction with older members. However, this may not be unusual, as there is some similarity with these comments and a recent study by Rolstad (2014) in relation to *how* to involve OPWH in NMOs through age and gender appropriate activities at appropriate times and places. Despite this, the social connection HFNZ provided was obviously important to participants, as approximately one third used HFNZ as a way to connect socially and had attended either a camp or workshop organised by HFNZ. It is therefore interesting that HFNZ did not come up in the open-ended

question asking about effective supports in OPWH lives. Perhaps participants only view HFNZ as an educational resource or facilitator of social connection, and not as a direct support per se.

Otherwise, in general, participants found HFNZ an effective service and felt supported by their outreach workers. Despite this positivity, a lot of the services were not well attended by the respondents, (ten out of sixteen services were used by less than half the respondents) so although they may not engage fully with the services provided, perhaps they can see the benefit for others and are appreciative of them being available. These findings were consistent with the minimal international literature on the subject and Street et al.'s (2006) comments that it is hard for NMOs to meet the needs of OPWH when their needs are unknown, and therefore NMOs need to gain information on their needs and develop 'wellness' programs, practices, policies, and supports which are directed at preventative strategies to reduce the physical and psychological impacts of aging.

Comprehensive Care

The HTC (primarily nurses and doctors) was the most discussed group within the comprehensive care teams. There was a lot of written and oral comment on HTCs and, for the most part, this was all positive. Staff and their effectiveness were rated very highly, and overall the HTC was seen as the second best support (after family). The majority of the international literature indicates that HTCs have made critical contributions toward improving QOL for individuals with haemophilia (Evatt et al., 2004). However, while Konkle et al. (2011) praise the HTC they also state that HTCs are in a predicament as they have limited experience with the wide scope of issues OPWH are facing. Despite these concerns, throughout the research participants conveyed very high praise for their specialist nurse and spoke fondly of others in the comprehensive care team. They were very grateful and appreciative for what they have here in New Zealand in comparison to other countries, and it was the access they had to their medical team that most impressed them. The HTC is thus a

very highly valued and praised service for most of the research participants, as shown in both the focus group and the questionnaire. However, an important issue was raised by focus group participants in terms of the HTC and how they can retain staff. Participants are very fond of their medical staff and acknowledge their specialist skills and extensive haemophilia expertise. They are very fearful of 'losing' their HTC care specialists who they 'rely on' and are worried about being left with gaps in services (and in their lives) when medical staff leave, or if positions are cut back or no longer funded through the healthcare system; so it is also important that staff retention strategies are put in place.

Another valued support was the specialist physiotherapist. The focus group spoke very fondly of their haemophilia physiotherapist, group physiotherapy sessions, and other physiotherapy initiatives, which have made a huge positive impact on their lives. In all, OPWH in this study thought that physiotherapy was a very effective service. Similarly, the literature also suggests specialist physiotherapists are crucial in the management and recovery of PWH (Wittmeier & Mulder, 2007). Dolan (2010) indicates that there is some concern about whether physiotherapists have new and updated information and skills to address the needs of an older population with haemophilia. However, this sentiment is obviously not felt by people in this study.

The results regarding two medical services in this research differed dramatically to the literature. The first of these and perhaps the most important was the coordination of care between the HTC and other healthcare professionals and specialists, including General Practitioners (GPs). Rolstad's (2014) study of social work perceptions of men with haemophilia in Utah, USA, raised many concerns about coordination of care, claiming there were problems with the lack of knowledge about haemophilia and its management by other medical professionals; worries about communication between specialists and the HTC; frustrations for PWH having to provide education and explain their condition to healthcare professionals; and that many OPWH would rather rely on their HTC to be able to provide all

of their care. The participants in the focus group agreed with Rolstad's views as they also talked at length about the coordination of care with other medical specialists, and all agreed the presence of comorbidities would mean they would need many more specialists involved in their care and this could be problematic on many levels. Despite this, respondents rated the coordination of care between HTC and specialists as being very effective and no one indicated that the coordination of their care was either ineffective or very ineffective. Surprisingly, this was the service that was rated the highest in the section of the questionnaire asking about service effectiveness. The international literature also indicates that GPs are crucial in the lives of OPWH in terms of helping with the management of comorbidities and general health as a person ages, yet typically OPWH do not have GPs and instead prefer to rely on their HTC for all of their healthcare needs (Mauser-Bunschoten et al., 2009). Due to this there is a lot of discussion by healthcare professionals on how to connect OPWH to a GP and how to make that engagement successful. Therefore it was also extremely surprising to find that every participant in this research indicated that they already had their own GP and that they found this GP effective in meeting their needs. So perhaps the care professionals in New Zealand have educated their service users well and supported them in obtaining GPs, which was dissimilar to the international literature.

The second medical support service where the literature and research findings differed considerably was around support for pain. In this study it was found that HTCs were the leading provider in terms of support for pain issues. This did not align with much of the international literature in terms of reports that HTCs do not effectively deal with pain (Witkop et al., 2012; Elander, 2014). Not only does the international literature state that HTCs are not good at dealing with pain, but that pain in OPWH is not managed well by any service, and in fact, specialists and supports are rarely consulted at all (Elander, 2014; Riley et al., 2011). This is different to the current research which showed approximately half had received support for pain issues and they perceived the service they accessed (most often the HTC) as

effective in meeting their needs. Therefore, perhaps HTC clinicians in New Zealand have more access to training on pain management, or work in structures which allow them to more effectively deal with pain than overseas.

Two services that were rarely accessed or consulted with by OPWH in this research were mobility services, and those to assist with issues around engagement in sexual activity. Sexual activity in older men with haemophilia was a topic that was regularly discussed in the international literature, and it was often acknowledged that there are many issues and barriers which prohibit OPWH from engaging in sexual activity (Gianotten & Heijnen, 2009). Prior research has clearly identified the importance of health care professionals and services talking about this issue with OPWH, and making referrals to specialists where needed (Mauser-Bunschoten et al., 2009). The HERO study showed 28% of adults had talked to doctors about sexual intimacy (Cassis, et al. 2012), which is much higher than the rate reported by respondents in this research, where only one respondent (2.50%) indicated he had received any professional support to help with challenges about this issue, and it was therefore the lowest service used by far. It is possible that, because it is a sensitive issue, respondents had not sought out any support due to shame and embarrassment, although there may be support services available to them. This is in line with the findings from Arnold et al. (2013) who found that knowledge seeking by patients in this area of sexual engagement was low.

The second rarely accessed service was mobility support. Less than half of the respondents reported that they had received professional help to support them with mobility issues. This is concerning considering the current prevalence and severity of these challenges, and their associated fears for the future as previously discussed. For those who had received some support, a wide variety of different services had been used for mobility support (most of which seemed to be self-referred). This may present some problems if OPWH are not seeing the best or most appropriate service and raises questions if routine discussions, assessments,

and referrals are or are not taking place through HTCs or medical providers for this very important and serious issue.

People who provided support for emotional issues and those who provided therapeutic interventions were rarely discussed in any phase of this research. However, as previously deliberated on, most people did not seem to have emotional concerns, and of those that did, most respondents found the support sought from these professionals to be effective.

This research has clearly shown that there are some substantial challenges for OPWH in this study, specifically in the area of physical health. The most prevalent and serious issues arose from decreasing mobility and decreasing physical ability but pain, medical health, and comorbidities were also large challenges. Employment and the cost of haemophilia treatment to the healthcare system were two other key challenges for OPWH. There were no widely experienced challenges which correlated specifically to age related issues. However, some, such as being able to access their own veins to give themselves haemophilia treatment and losing independence, did provide a select few participants with substantial challenges and did provide worries and fears for the future. In contrast to other studies and reports from haemophilia authors abroad, it was found that the respondents to this research had positive mental and emotional health and wellbeing. Overall participants in this study perceived that they had access to very effective and caring supports and services. The best and most effective supports were family (specifically significant others), the HTC, the coordination of care between healthcare providers and specialists, specialist physiotherapists, and wider social support such as social groups and others with bleeding disorders. However, of some concern was the low use of services to aid in problems with engagement in sexual activity and particularly mobility.

Limitations

Some important findings have been highlighted in the discussion, but it is also important to acknowledge the limitations to this research, the biggest of which was response bias. Forty individuals responded to the questionnaire out of a possible ninety people, and making generalisations and assumptions on a whole population based on input from only half of the population raises some concern over the representativeness of the research. As individuals with low mood may have been reluctant or unable to answer the questionnaire, this presents particular problems in terms of generalising the findings for mental and emotional wellbeing. Further, for those with severe haemophilia or comorbidities, pain, mobility issues, or who were in hospital, this may have prevented them from answering the questionnaire, thus the results are likely to be biased in a positive direction. Fortunately, the age range of respondents was representative of the population who the questionnaire was sent to, and also all severities of haemophilia were represented (most experienced a mild level of severity, followed closely by severe haemophilia). The vast majority of respondents lived within two hours of the nearest HTC, however it is difficult to identify if this is due to missing experiences from those who live rurally, or if it is representative of the population as there are no population-based statistics available. The majority of respondents identified as NZ European, with only four people identifying with another ethnicity, and only two of those respondents were Māori. Therefore, there are queries about the representativeness of ethnicities, those who live away from versus close to a HTC, and those who were unable or unwilling to answer the questionnaire.

This research only involved participants who were recorded on the HFNZ database (HFNZ members). Therefore, people unknown to HFNZ, or those who had asked to be removed from the HFNZ contact list at some point were unable to provide feedback. This has specific ramifications for the HFNZ section of the questionnaire as people who had asked to be removed from the HFNZ database were likely to be people who had experienced

dissatisfaction or discontentment with HFNZ; therefore, their views and opinions were not represented in the research. To navigate this issue would have required access to each DHB's hospital database (as there is no central database for people with haemophilia). Access to this hospital information presented many ethical challenges and thus was not feasible for the current research. Although people who were unidentified by HFNZ or who had asked to be removed from the database are thought to be few, it is acknowledged that there may be some important views missing from the research.

Three other minor limitations were identified. First, there was a slim possibility that participants could have completed the questionnaire twice (via paper copy and the online survey monkey link). This risk was mitigated by clearly stating in the participant information sheet (PIS) that the questionnaire was only to be completed and submitted once (through either method) and questionnaire responses were also carefully reviewed for potential duplicate entries of which there was no evidence. Secondly, some participants deviated from answering questions correctly in the paper (hand written) copies of the questionnaire. For instance, some respondents ticked two answers (instead of one), ticked in-between answers, included written notes by answers (which did not require notes), and wrote answers which were illegible. Each of these cases (n = 8) was reviewed and discussed with the researcher's academic supervisors and decisions were made on how to best record the information. Finally, one questionnaire was received which was omitted from analysis as the person stated that they were not a member of HFNZ. Therefore, they could not have been on the HFNZ database, and did not receive the questionnaire through legitimate means, so did not fit the research criteria. In all, these anomalies were mitigated where possible and carefully thought through with the researcher's academic supervisors.

Recommendations

Despite these limitations, the research has provided a holistic overview of the key areas of concern, and a thorough exploration of the current services for a sample of older men

with haemophilia in New Zealand. The findings are very important for OPWH, and the services and individuals who support OPWH in New Zealand. Based on the findings of this research, several recommendations are offered.

Family members are highly valued by the respondents in this research. It is therefore important that all service providers (both medical teams, and psychosocial support practitioner) endeavour to keep family and other valued support people involved and engaged in their processes with OPWH (i.e. meetings, discussions, planning, reviews and educational and social events). Not only should family be included, but their opinions should be sought, and their input valued. However, services also need to be wary and mindful that family relationships can cause challenges and fears for some (specifically for those without a significant other) so OPWH must be asked to define their own support systems, and how they want these support people to be involved in their care.

OPWH in this study also highly value social participation, connection, and support from social, community, and bleeding disorder groups. Therefore, services and support people should encourage and help connect OPWH with wider social supports and social groups that appeal to that individual and also to others with bleeding disorders. This connection could help reduce some of the psychological impacts of aging by sharing, connecting, and increasing their understanding that they are not alone. HFNZ and all service providers need to think creatively and specifically about the demographic they are working with, and ensure that all of their social initiatives are relevant, appropriate and enticing to OPWH.

Many OPWH are facing fear and uncertainty, as there are still many unknowns for what their future may hold. This was found particularly with regards to the fear of additional comorbidities, aging and decreasing medical health, independence, and mobility. To overcome these fears and worries some OPWH may require support to address and work through these fears. This could be achieved through therapeutic support (e.g. counselling), or

through practical planning and preparing for the future (e.g. with social workers). It is likely that psychosocial professionals will play an increasingly important role in supporting PWH as they age and face new and unique challenges in their lives, and therefore referrals and connection with these professionals should be made when necessary. This recommendation is one which has rarely been discussed by authors in the haemophilia field .

Though most of the supports and services were perceived by participants to be effective and highly valued, there were still some concerns with access to some services and inconsistencies with the services that were used to deal with some issues. This was especially so for mobility, sexual engagement, pain, and emotional problems, all of which are sensitive topics to discuss. To ensure these crucial areas are addressed appropriately it is important for regular and routine discussions to occur about these sensitive topics with medical and psychosocial professionals. These professionals need to be aware that OPWH may be reluctant to bring these issues up themselves so professionalism, sensitivity, and compassion are required. Following discussions, verified and appropriate tools for professionals to use to easily and quickly assess OPWH (i.e. depression screening, mobility assessments) are needed along with having systems and processes in place to make referrals to specialists to address and assist with these issues where appropriate. This is an issue that other authors in the haemophilia field have also suggested as important, but these discussions do not seem to be effectively taking place at present.

It is important that services and supports address not only haemophilia and medical related issues, but also general aging issues faced by OPWH. Although these issues may be slightly different for the haemophilia population, they still share some similarities with the general aging population (i.e. mobility, comorbidities, falls and independence). Input needs to be solicited from specialists with extensive experience in these areas, such as gerontologists, local aging organisations (e.g. age concern), aging specialists, and people outside the

haemophilia realm who are involved with positive aging. To date it seems that aging specialists and groups have rarely been consulted for advice regarding OPWH.

As highlighted in the discussion there were some inconsistencies between research overseas and what was found in the current study. There were also many areas where OPWH had significantly different views, opinions and experiences to one another. Therefore it is important that services and supports see each OPWH as an individual and avoid making assumptions about them, instead engaging in discussions with that individual and making tailored plans and decisions appropriate to their specific needs.

Future Directions

This research has wider implications for spheres such as social work practice and policy, and provides a foundation, and a tool, that could potentially lead into larger or different pieces of research. The OPWHQ is a tool that could be used again (and again) with the same population over time to assess any changes (positive or negative) in the wellbeing of OPWH in New Zealand and the support they receive. The OPWHQ also has the scope to be used by OPWH in other countries. Then results between countries could be compared, and similarities, differences and trends analysed.

The current research only used univariate analysis to explore the quantitative OPWHQ findings. This was due to the MSW scope, word and time constraints, and the aim to look at general holistic wellbeing of OPWH in New Zealand broadly. However, there is much that can still be done using the findings generated from this research. There is opportunity for a researcher to extend this research to investigate OPWHQ results in greater depth by using bivariate analysis i.e. relationships between: mobility issues and both age and severity of bleeding disorder; social support and age; ability to infuse haemophilia treatment and both age and severity of bleeding disorder; life satisfaction and age; experiences of medical services between those who live close to the medical facilities and those which live further away.

Opportunities also exist to use this piece of research as a starting point to conduct more in-depth qualitative work based on the key findings identified, specifically in terms of the aspects which were absent from discussions in the international literature such as the fear that the cost of haemophilia treatment would prevent OPWH receiving the treatment and care they will need in the future, loss of independence, and changes in the ability to infuse their own haemophilia factor treatment, residential care homes and spiritual wellbeing. Therefore, not only has this thesis presented some interesting and useful findings and recommendations, but there is still much that could be drawn from the data or further investigated in the future.

Conclusion

This thesis offers an in-depth exploration of the unique issues and challenges facing older men with haemophilia, and their perceptions of services and supports available to them in Aotearoa, New Zealand. The rationale for this research was straightforward: the literature established that life expectancy for people with haemophilia has been slowly increasing over time, and is now approaching that of the general male population (Mannucci et al., 2009). As a result, OPWH are now facing the same aging issues as the general population for the first time. This brings new and unique challenges for this group and adds further complexity to their treatment, care, and support. However, despite this growing issue there were significant gaps in the broad knowledge base pertaining to the needs and experiences of OPWH (Young, 2012). Prior to this research, the experience and needs of older men with haemophilia were explored in a limited manner with limited acknowledgment or assessment of holistic wellbeing, and throughout the literature academics and service providers alike recognised that assessment of the needs of OPWH were urgently required (Dolatkhah et al., 2014; Franchini & Mannucci, 2010).

Consequently, this study was conducted to provide a thorough examination of two research questions 1) what are the unique issues and challenges facing older men who are living with haemophilia in Aotearoa New Zealand? and 2) how do older men with

haemophilia perceive the support services available to them in Aotearoa New Zealand? These questions were addressed by meeting the research objectives:

- Identifying the unique issues and challenges faced by these men
- Exploring the prevalence and seriousness of these issues
- Describing the impact of these issues and challenges on their holistic well-being
- Identifying which services and supports are being used
- Describing their perceptions of the effectiveness of these services and supports

It was argued that meeting these objectives would provide a broad picture of OPWH in New Zealand, and based on that, discussion and recommendations could be drawn to contribute to the assessment and improvement of the wellbeing of OPWH in New Zealand.

This research has clearly shown that there are some substantial challenges for OPWH specifically in the area of their physical health such as decreasing mobility and decreasing physical ability, pain, medical health, and comorbidities. Also causing concern was the effect haemophilia had on their working life, and the fear that the cost of their haemophilia treatment would prevent them getting the care, medications, and operations they may need in the future. However, despite these challenges some findings which were very optimistic, such as the positive mental and emotional wellbeing of OPWH in New Zealand. Overall, participants in this study perceived that they had very effective and caring supports and services around them. First and foremost was their family (specifically their significant other), followed by the haemophilia treatment centre, coordination of care they received between healthcare providers and specialists, specialist physiotherapist, and wider social supports. However, of concern was the low use of services to aid in problems with engagement in sexual activity and mobility.

This research provides professionals working with people with haemophilia with a starting point for conversations about OPWH, presents some of the indicated major

challenges, and describes the perceptions regarding the support received by the men in this study. In addition, this thesis considers ways in which the lives and experiences of OPWH in this study could be improved and provides clear consideration for the future. The findings are useful as they stand, but there is also more that can be done with the results and with the tool created. The thesis has achieved what it set out to do - produce new knowledge, raise awareness of needs, and gain a deeper understanding of the holistic wellbeing of OPWH, but most importantly provides sound research that will hopefully improve and enhance the lives and wellbeing of older men with haemophilia in New Zealand.

APPENDICES

Appendix A: Participation Information Sheet (Focus Group Participant)

Project Title
Explorations into the unique issues and challenges faced by older men with Haemophilia

The Researchers
My name is Sarah Elliott and I am completing a Master of Social Work qualification from the University of Auckland under the supervision of Dr Kelsey Deane and Dr Barbara Staniforth. I also work for HFNZ who have contacted you on my behalf.

Project Description
I am very interested in investigating the impact of the unique issues and challenges facing men aged 45 years and over who are living with haemophilia, including their perceptions of supports and services available to them in Aotearoa New Zealand.

The Study
After consultations with Haemophilia Foundation New Zealand (HFNZ) to identify males with haemophilia who are 45 years or older and are registered on the HFNZ database and who live in the Wellington region, you have been identified as someone who fits the criteria for this study and may be able to make a contribution due to your knowledge and insight about the research topic. I am inviting you to participate in a small focus group to discuss your opinions on this topic, and to review a draft questionnaire that will be used in a larger survey study sent to all males 45 years and older with haemophilia in New Zealand.

The focus group will be held in the Wellington area and should take approximately 90 minutes of your time. A further 10 to 20 minutes of your time will be requested approximately one month later via a follow up email to look over the revised questionnaire and give any final feedback or comments.

Benefits to being involved?
This study is an opportunity to contribute to an important piece of research which could potential help to raise awareness of the needs of older people with haemophilia in NZ. Therefore, by being a part of this research you will be adding great value and depth to the findings. You could also be helping to provide older people with haemophilia, their support people, and individuals working with the bleeding disorder community an insight into what things this cohort may face as they continue to age. This will be useful in finding how best to support them, therefore offering opportunities for increasing awareness and preventative action.

Food and non-alcoholic drinks will be provided at the focus group and each participant will be provided with a $20 grocery voucher as a token of appreciation (koha) for your contribution.

Conflicts of interest
I am a practicing social worker with HFNZ however; this academic research project is completely separate to my role within the organisation.

HFNZ have an interest in in the outcome/findings of this research and are therefore partial financial sponsors of the research project. HFNZ have provided their assurance that this will in no way compromise the integrity of the results and HFNZ will only have access to the final report, so I ask participants to please be open and honest in all their disclosures.
Confidentiality, consent and safeguards

This focus group will be audio recorded for verification purposes only; however, this recording will not be made available to you at any point nor will the transcription of the recording because the information will reflect group and not individual discussion.

Participants will all be asked to sign a consent form and to maintain the privacy and confidentiality of other members; however, in the group situation we cannot guarantee that other participants will honour this. Only the research team will have access to individual information and no individual participants will be identified when we write up the results.

Participation is completely voluntary. The acting CEO of HFNZ has given their assurance that your participation or non-participation in this research will in no way impact upon your relationship with the organisation. Participants are free to withdraw from this study or leave the focus group discussion at any time without having to give reason and without penalty. However, participants will not be able to withdraw anything which has been recorded because it will form part of the group data.

The hardcopy data and consent forms will be securely stored in separate locked filing cabinets in Dr Kelsey Deane's office at the University of Auckland's Epsom campus for 6 years, after which point they will be confidentially destroyed in compliance with the University of Auckland's secure destruction of research data procedures. The audio recording will be transferred to the researchers' password protected computer and kept for 6 years. The original will be deleted immediately after transferring the file to the researchers' computer. The anonymized notes will be entered into an electronic file and stored on the researchers' password protected computer indefinitely.

We do not believe that there will be any risks to individuals taking part in the focus group however some of the focus group questions are sensitive in nature and this could potentially cause you some discomfort. You should not feel obliged to answer any questions you feel uncomfortable about. If you would like to discuss any concerns arising from the questionnaire you are welcome to contact me, my supervisors, or the Chair of the University of Auckland Ethics Committee at any time using the contact details at the bottom of the form. If you would prefer to discuss your concerns with your outreach worker, she will be able to support you. Call Freephone: 0508factor.

Dissemination
The findings will form part of my Master of Social Work thesis and may be published in academic journals or discussed in presentations or at conferences.

The final report and outcome of the research will be made available to you once it is completed in 2015. HFNZ will make you aware of this via an article in the Bloodline magazine and they will have a copy of the report which you could contact them to obtain.

Next Steps
Thank you for your consideration to participate in this focus group. If you have any concerns or questions, please contact us at any time. If you would like to participate in this focus group, please contact Sarah Elliott on the details bellow to indicate your interest and I will respond with further details of the location and time of the focus group.

Contact details for the researcher, supervisor and Head of Department
Sarah Elliott 0275121114 Email: spre607@aucklanduni.ac.nz

Dr Kelsey Deane 09 623 8899 ext. 48685 Email: k.deane@auckland.ac.nz

Dr Barbara Staniforth 096238899 ext 48349 Email: b.staniforth@auckland.ac.nz

Chair contact details
For any concerns regarding ethical issues you may contact the Chair, the University of Auckland Human Participants Ethics Committee, The University of Auckland, Research Office, Private Bag 92019, Auckland 1142, Telephone 09 383 7599 ext. 87830/83761. Email: humanethics@auckland.ac.nz

APPROVED BY THE UNIVERSITY OF AUCKLAND HUMAN PARTICIPANTS ETHICS COMMITTEE ON 19 JUNE 2014 FOR 3 YEARS, REFERENCE NUMBER 012105

Appendix B: Consent Form for Focus Group Participants

THE UNIVERSITY OF AUCKLAND
FACULTY OF EDUCATION
School of Counselling, Human Services and Social Work

University of Auckland
Faculty of Education
Epsom Campus
Private Bag 92601 Symonds Street
Auckland

Consent Form
For focus group participants

THIS FORM WILL BE HELD FOR A PERIOD OF 6 YEARS

Researchers
Sarah Elliott, Dr Kelsey Deane, Dr Barbara Staniforth (University of Auckland)

Research Title
Explorations into the unique issues and challenges faced by older men with haemophilia

Agreement

- I have read the information provided and understand the explanation of the research and why I have been invited to participate
- I have had the opportunity to ask questions and my questions have been answered to my satisfaction
- I understand that participating in this research is voluntary (my choice), and that my participation OR non-participation will not affect my relationship with HFNZ
- I understand that my participation in this research is confidential and that no material which could identify me will be used in any reports on this study
- I understand that I may withdraw from the study at any time without having to give reasons and without penalty
- I understand that I cannot withdraw data already provided
- I understand that I have been asked to attend one focus group lasting approximately 90 minutes and one follow up email to review changes which will take approximately 10-20 minutes
- I understand that information shared by everyone in the focus group is to be kept confidential
- I understand some of the questions in the focus group could potentially cause me some discomfort. If this is the case, I am aware of who I can contact to discuss any issues
- I understand that only the research team will have access to the research notes
- I understand that the focus group will be audio recorded
- I understand that I will not have access to the audio recording
- I understand that this consent form shall be kept separate to the hard copy of the focus group notes and the audio recording, all of which will be kept for 6 years in a secure filing cabinet in Dr Kelsey Deane's office at the University of Auckland's Epsom campus, after which time they will be destroyed and digital audio recordings will be deleted

- I understand that the electronic versions of written files will contain no information that can identify me, and that these files will be kept indefinitely on the researchers' password-protected computer
- I know who to contact if I have any concerns about the study

I_____ (full name) hereby give my consent to take part in this focus group

Signature_____ Date_____

Thank you for your consideration to participate in this study. If you have any concerns or questions, please contact us at any time.

Contact details for the researcher, supervisor and Head of Department
Sarah Elliott 0275121114 Email: spre607@aucklanduni.ac.nz

Dr Kelsey Deane 09 623 8899 ext. 48685 Email: k.deane@auckland.ac.nz

Dr Barbara Staniforth 096238899 ext 48349 Email: b.staniforth@auckland.ac.nz

Chair contact details
For any concerns regarding ethical issues you may contact the Chair, the University of Auckland Human Participants Ethics Committee, The University of Auckland, Research Office, Private Bag 92019, Auckland 1142, Telephone 09 383 7599 ext. 87830/83761. Email: humanethics@auckland.ac.nz

APPROVED BY THE UNIVERSITY OF AUCKLAND HUMAN PARTICIPANTS ETHICS COMMITTEE ON 19 JUNE 2014 FOR 3 YEARS, REFERENCE NUMBER 012105

Appendix C: Participant Information Sheet (Questionnaire Participant)

THE UNIVERSITY OF AUCKLAND
FACULTY OF EDUCATION
School of Counselling, Human Services and Social Work

University of Auckland
Faculty of Education
Epsom Campus
Private Bag 92601 Symonds Street
Auckland

Participation Information Sheet (Questionnaire participant)

Project Title
Explorations into the unique issues and challenges faced by older men with Haemophilia

The Researchers
My name is Sarah Elliott and I am completing a Master of Social Work qualification from the University of Auckland under the supervision of Dr Kelsey Deane and Dr Barbara Staniforth. I also work for HFNZ who have forwarded this questionnaire to you on my behalf.

Project Description
I am very interested in investigating the impact of the unique issues and challenges facing men aged 45 years and over who are living with haemophilia, including their perceptions of supports and services available to them in Aotearoa New Zealand. This questionnaire has been developed through a review on the literature of people living with Haemophilia, as well as through consultation with a focus group of HFNZ members.

The Study
After consultations with Haemophilia Foundation New Zealand (HFNZ) to identify males with haemophilia who are 45 years or older and are registered on the HFNZ database you have been identified as someone who fits the study criteria; therefore, I am inviting you to contribute to this research project by answering the attached questionnaire. This questionnaire should take approximately 20 minutes for you to answer.

Your involvement in this study is completely voluntary and you do not have to answer any questions that you are not happy to answer. If you are happy to participate, then please complete and return the questionnaire in the postage-paid envelope provided. Alternatively, you can complete the questionnaire online if this is easier for you. Please do so by going to: www.surveymonkey.com/s/D5VTHVL Please only complete the questionnaire once, either using the online or this hard copy version.

Benefits to being involved?
This study is an opportunity to contribute to an important piece of research which could potential help to raise awareness of the needs of older people with haemophilia in NZ. Therefore, by being a part of this research you will be adding great value and depth to the findings. You could also be helping to provide older people with haemophilia, their support people, and individuals working with the bleeding disorder community an insight into what things this cohort may face as they continue to age. This will be useful in finding how best to support them, therefore offering opportunities for increasing awareness and preventative action.

Confidentiality, consent and safeguards
Please note that by completing and returning the questionnaire in the envelope provided you are consenting to the use of your information for this research project. To ensure your anonymity is protected please do not put any personal names or contact details in the questionnaire or on the return envelope. While we will do our best to protect your anonymity, there is a very slight chance that people who know you may recognise some of the comments you have made if they represent a unique experience or view that other individuals are aware of.

Responses from the hardcopy questionnaire will be entered into an electronic file and these along with the questionnaires entered online will be kept on the researchers' password protected computers indefinitely. The original hard copies of the completed questionnaires will be securely stored in a locked filing cabinet in Dr Kelsey Deane's office at the University of Auckland's Epsom campus for 6 years, after which point they will be confidentially destroyed in compliance with the University of Auckland's secure destruction of research.

We do not believe that there will be any risks to individuals completing the questionnaire however some of the questions are sensitive in nature thus this could potentially cause you some discomfort. If you would like to discuss any concerns arising from participation in this research project you may contact me, either of my supervisors or the Chair of the University of Auckland Human Participants Ethics committee using the contact details provided below. You could also contact your Outreach Worker who will be able to support you, Call Freephone: 0508factor. For those of you in Northern region where I am your social worker you may choose to discuss any concerns with my manager: Colleen McKay (phone: 033717477) instead.

Conflicts of interest
I am a practicing social worker with HFNZ; however, my role within this academic research project is completely separate to my role within the organisation.

Please note that HFNZ have expressed an interest in in the findings of this research and have agreed to partially fund the research. HFNZ have provided their assurance that this will in no way compromise the integrity of the results. Furthermore, HFNZ will only have access to the final report.

Dissemination
The findings will form part of my Master of Social Work thesis and may be published in academic journals or discussed in presentations or at conferences.

The final report and outcome of the research will be made available to you once it is completed in 2015. HFNZ have also agreed to disseminate the findings via an article in the Bloodline magazine and they will have a copy of the report which you could contact them to obtain.

Thank you for your consideration to participate in this study. If you have any concerns or questions, please contact us at any time.

Contact details for the researcher, supervisor and Head of Department
Sarah Elliott 0275121114 Email: spre607@aucklanduni.ac.nz

Dr Kelsey Deane 09 623 8899 ext. 48685 Email: k.deane@auckland.ac.nz

Dr Barbara Staniforth 096238899 ext 48349 Email: b.staniforth@auckland.ac.nz

Chair contact details
For any concerns regarding ethical issues you may contact the Chair, the University of Auckland Human Participants Ethics Committee, The University of Auckland, Research Office, Private Bag 92019, Auckland 1142, Telephone 09 383 7599 ext. 87830/83761. Email: humanethics@auckland.ac.nz

APPROVED BY THE UNIVERSITY OF AUCKLAND HUMAN PARTICIPANTS ETHICS COMMITTEE ON 19 JUNE 2014 FOR 3 YEARS, REFERENCE NUMBER 012105

Appendix D: Questionnaire

A Health and Wellbeing Questionnaire for Older Men with Haemophilia

The purpose of this questionnaire is to investigate the impact of the unique issues and challenges facing men aged 45 years and over who are living with haemophilia, including their perceptions of supports and services available to them in Aotearoa New Zealand.

Some questions are sensitive; however, obtaining accurate information about the nature of your experiences is important for understanding and improving service delivery, so please be open and honest in all your responses.

*Your answers are **completely confidential** and no one other than the researcher will have access to the questionnaires. Please do not put any identifying information on the questionnaire.*

If you would prefer to fill this in online please go to:
www.surveymonkey.com/s/D5VTHVL

The survey should take approximately 20 minutes to complete. Please circle the correct answer for each question unless you are instructed otherwise. Your participation is highly appreciated.

Demographics

1. Which age group do you fall within?

45-49	50-59	60-69	70-79	80-89	90-99	100+

2. Where do you normally live?

Within two hour drive to the closest haemophilia treatment centre	More than two hours' drive to the closest haemophilia treatment centre

3. What is your haemophilia factor level?

Mild	Moderate	Severe

4. What is your ethnicity?

New Zealand European	Maori	Samoan	Cook Island Maori	Tongan	Niuean	Chinese	Indian	Other:

Comprehensive Care

5. How supported do you feel by your Haemophilia Foundation of New Zealand (HFNZ) Outreach Worker?

Very unsupported	Unsupported	Fairly supported	Supported	Very supported

6. What services do you use within HFNZ? (Please tick all appropriate options)

	Outreach workers support
	Conferences
	Workshops and camps
	Opportunity to connect socially with others
	Bloodline
	Panui
	HFNZ educational resources
	Advocacy/help by the HFNZ to others on your behalf
	Exercise programme
	Supportive footwear programme
	Defensive driving programme
	Needs grants
	Member Representative Group (MRG) activities
	Website
	Piritoto
	Masters Group
	Other (please list)

7. How effective is HFNZ in addressing your needs?

Very ineffective	Ineffective	Fairly effective	Effective	Very effective

8. Please comment on your experiences of the HFNZ service:

_____
_____
_____
_____

9. Do you think HFNZ could change or improve their services?

No	Maybe	Yes

Please explain how: _____
_____
_____
_____

10. Have you ever seen a specialist haemophilia physiotherapist?

Yes	No *Go to Q 12*

11. How effective is the specialist physiotherapy service in addressing your needs?

Very ineffective	Ineffective	Fairly effective	Effective	Very effective

12. How effective is your Haemophilia Treatment Centre (HTC) in meeting your needs?

Very ineffective	Ineffective	Fairly effective	Effective	Very effective

13. How supported do you feel by the HTC staff?

Very unsupported	Unsupported	Fairly Supported	Supported	Very supported

14. Do you have a specific GP?

Yes	No *Go to Q 16*

15. How effective is your GP in meeting your needs?

Very ineffective	Ineffective	Fairly effective	Effective	Very effective

16. Are you involved with medical specialists other than your HTC and comprehensive care team?

Yes	No *Go to Q 18*

17. What is your experience of co-ordination of your care between the HTC and other specialist services?

Very Negative	Negative	Fair	Positive	Very positive

18. Have you been to the A&E for haemophilia-related problems since you were 45 years of age?

Yes	No *Go to Q 20*

19. How effective was the A&E service in addressing your needs?

Very ineffective	Ineffective	Fairly effective	Effective	Very effective

Physical Health

Yes	No *Go to Q 22*

20. Do you have any medical conditions other than haemophilia?

If yes, please specify:

_____

21. What concerns you more - your haemophilia or another (other) medical condition(s)?

Haemophilia	My other condition(s)

22. To what degree does having haemophilia affect your ability to engage in physical activity?

Not at All	Slightly	Moderately	Considerably	Severely

23. To what degree is your joint mobility/movement limited?

Not at All *Go to Q26*	Slightly	Moderately	Considerably	Severely

24. Have you ever received any professional support to help you overcome some of these mobility barriers or to discuss mobility equipment and aids?

Yes	No *Go to Q 26*

25. Which service provided this support?

_____

26. How would you rate your current diet?

Very unhealthy	Unhealthy	Fairly healthy	Healthy	Very healthy

27. I currently consider myself to be:

Very underweight	Underweight	Of normal weight	Overweight	Very overweight

28. To what degree do you experience pain?

Not at all *Go to Q33*	Slightly	Moderately	Considerably	Severely

29. Have you ever received any professional help or advice to make decisions about dealing with pain?

Yes	No *Go to Q 32*

30. Which service provided this support?

_____

31. How effective was this service in meeting your needs?

Very ineffective	Ineffective	Fairly effective	Effective	Very effective

32. To what degree do you consider yourself dependent on pain medications?

Not at all	Slightly	Moderately	Considerably	Severely

33. To what degree does having haemophilia affect your sexual activity?

Not at all *Go to Q 37*	Slightly	Moderately	Considerably	Severely

34. Have you ever received any professional help or advice to support you to overcome barriers to sexual activity?

Yes	No *Go to Q 37*

35. Which service provided this support?

_____

36. How effective was this service in meeting your needs?

Very ineffective	Ineffective	Fairly effective	Effective	Very effective

37. In general would you say your health is:

Poor	Fair	Good	Very good	Excellent

Work, Income and Transport

38. To what degree has haemophilia affected your working life? *Note: If you have retired, please respond with regard to your working life prior to retirement.*

Not at all	Slightly	Moderately	Considerably	Severely

39. To what degree do you experience transport difficulties to get to medical appointments?

Not at all	Slightly	Moderately	Considerably	Severely

40. To what degree does haemophilia impact on you financially?

Not at all	Slightly	Moderately	Considerably	Severely

41. Do you receive any benefits other than superannuation?

Yes	No *Go to Q 43*

42. To what degree do you feel supported by the government to obtain and access this/these benefit(s)?

Very unsupported	Unsupported	Fairly supported	Supported	Very supported

43. To what degree do you think the cost of haemophilia treatment and care <u>to the healthcare system</u> impacts on the medical service or care you receive?

Not at all	Slightly	Moderately	Considerably	Severely

Social Support

44. How involved are you with social groups, clubs or teams (e.g. religious, cultural, volunteer, sport, hobbies or educational groups)?

Not at all involved	Slightly involved	Moderately involved	Considerably involved	Very involved

Please list all groups you are involved with:

_____

45. Compared to others your age, how often would you say you participate in social activities

A lot less than most	Less than most	The same as most	More than most	A lot more than most

46. Please tick the appropriate response for each question on the rating grid below

	Very Strongly Disagree	Strongly Disagree	Mildly Disagree	Neutral	Mildly Agree	Strongly Agree	Very Strongly Agree

There is a special person who is around when I am in need							
There is a special person with whom I can share my joys and sorrows.							
My family really tries to help me.							
I get the emotional help and support I need from my family.							
I have a special person who is a real source of comfort to me.							
My friends really try to help me.							
I can count on my friends when things go wrong.							
I can talk about my problems with my family							
I have friends with whom I can share my joys and sorrows.							
There is a special person in my life who cares about my feelings.							
My family is willing to help me make decisions.							

I can talk about my problems with my friends							

The above questions are sourced from:
Zimet, G., Dahlem, N., Zimet, S., & Farley. (1988). The multidimensional Scale of Perceived social Support. *Journal of Personality Assessment*, 52(1).

Mental, Emotional and Spiritual Wellbeing

Please tick the appropriate response for each question on the rating grid below

47. Over the **last two weeks** how much of the time…..

	All of the time	Most of the time	Slightly more than half the time	Slightly less than half the time	Some of the time	At no time
have you felt low in spirits or sad?						
have you lost interest in your daily activities?						
have you felt lacking in energy and strength?						
have you felt less self-confident?						
have you had a bad conscience or feelings of guilt?						
have you felt that life wasn't worth living?						
have you had difficulty in concentrating?						
have you felt very restless?						
have you felt subdued or slowed down?						
have you had trouble sleeping at night?						
have you suffered from reduced appetite?						
have you suffered from increased appetite?						

The above questions are sourced from: Psychiatric Research Unit, WHO Collaborating Center for Mental Health. (1998). *Major Depression Inventory.* Retrieved from: http://www.psykiatri-regionh.dk/who5/menu/WHO-5+Questionnaire/

48. Please tick the appropriate response for each question on the rating grid below

	Strongly agree	Agree	Slightly agree	Neither Agree or disagree	Slightly disagree	Disagree	Strongly disagree
In most ways my life is close to my ideal							
The conditions of my life are excellent.							
I am satisfied with life.							
So far I have gotten the important things I want in life.							
If I could live my life over, I would change almost nothing.							
The above questions are sourced from: Diener, E., Emmons, A., Larsen, R., & Griffin, S. (1985). The Satisfaction With Life Scale. *Journal of Personality assessment*, 49(1).							

49. How often have you seen a professional (such as a doctor, counsellor or psychologist) for emotional concerns?

Never *Go to Q 52*	A few times	More than a few times	Regularly

50. Which service provided this support?

_____

51. How effective was this service in meeting your needs?

Very ineffective	Ineffective	Fairly effective	Effective	Very Effective

52. To what degree do you feel connected to your ethnic culture or heritage?

Not at all connected	Slightly connected	Moderately connected	Considerably connected	Extremely connected

53. Are you religious?

No	Sort of	Yes

54. How important is spirituality to you?

Note. *In this questionnaire, spirituality is defined as the attachment to or regard for things of the spiritual world as opposed to material or worldly interests e.g. Connection to a god, the mystical or nature; or contemplation/meditation on the meaning of life.*

Not at all important	Slightly important	Moderately important	Considerably important	Extremely important

Age related changes

55. Have you experienced a fall which has led to a haemophilia bleed since the age of 45?

Yes	No

56. Has your ability to self-infuse your own haemophilia factor treatment changed as you have gotten older?

Yes	No *Go to Q58*

57. To what degree has this affected your life?

Not at all	Slightly	Moderately	Considerably	Severely

58. To what degree is aging having an effect on your independence?

No effect at all	A slight effect	A moderate effect	A considerable effect	A severe effect

59. To what degree do you think residential care would cope (or is coping) with looking after you?

Very poorly	Poorly	Fairly well	Well	Very well

Closing questions and thoughts

60. What are the biggest challenges, issues or concerns in your life at present?

_____
_____
_____
_____
_____
_____

61. What are your biggest fears for your future?

_____
_____
_____
_____
_____
_____

62. What are the most effective support systems in your life?

_____
_____
_____
_____
_____
_____

63. What are you most grateful for in your life?

_____
_____
_____
_____
_____
_____

64. What personal coping strategies have you developed which help keep you going?

_____
_____
_____
_____
_____
_____

Thank you for taking your time to participate in this questionnaire. We truly value the information you have provided.

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