The oral health of individuals with haemophilia: a mixed methods investigation

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Abstract

Those living with haemophilia can experience the same oral diseases as those without haemophilia; however, their dental treatment is complicated by prolonged bleeding and the reluctance of general dentists to treat such individuals. With these potential additional barriers to dental care, it is hypothesised that those with haemophilia have worse oral health than the general population.

There has been little research on the oral health of individuals with haemophilia, and none at all conducted in New Zealand. Limited conclusions can be drawn about the oral health of this population because much of the evidence that is available is of poor quality and is heterogeneous in setting, participants and measurements. However, the evidence that is available suggests that PWH are likely to have worse oral health than the general population.

In order to help improve the provision of oral healthcare to individuals with haemophilia, the aims of this research were to: describe the oral health and oral health behaviours of those with haemophilia; compare these findings with estimates from the New Zealand population; explore the perceptions of oral health and its importance expressed by those with haemophilia; and, identify the barriers to oral health perceived by those with haemophilia.

An embedded, correlational, mixed methods design was used to research the oral health of those with haemophilia in New Zealand. All individuals with moderate or severe haemophilia, aged over the age of two, and residing in the Capital & Coast, Hutt Valley or Wairarapa District Health Board regions, were invited to participate in a clinical examination, questionnaire and semi-structured interview.

Clinical examinations and questionnaires were completed by 33 individuals, comprising 25 adults and 8 children, giving a participation rate of 86.8%. Qualitative and quantitative data were considered in tandem to provide a snap-shot of the oral health of those with haemophilia and these data were compared with data from the general New Zealand population.
questionnaire, clinical examination and interview data paint a picture of a sample who have experienced oral disease and have varied perceptions of oral health. They acknowledge the importance of oral health but do not always carry out behaviours conducive to oral health, with many different barriers being described.

Having high dental anxiety and living further away from the hospital were significantly associated with being edentulous. Those who were edentulous were likely to have had periodontal disease and the decision-making process to render these individuals edentulous is different to that for individuals without haemophilia. When compared with the New Zealand population, those with haemophilia have more mild periodontal disease, which may be related to a reluctance to perform oral hygiene, especially flossing.

Generally, those with haemophilia appear to have better oral health (less caries experience and lower prevalence of severe periodontal disease) but where there is poor oral health it has more of an impact on those with haemophilia. A model has been suggested to show how better oral health is related to access to hospital dentistry.
Preface

The main body of this thesis is divided into five chapters.

The first chapter defines haemophilia and outlines the dental implications of this condition. A literature review describing the oral health of individuals with haemophilia is presented and this chapter finishes with a summary of the oral health of the New Zealand population.

The second chapter explains the mixed-methods approach used in this research. Methods of recruitment and the three methods of data collection (questionnaire, clinical examination, and semi-structured interview) are described.

The third chapter presents the results with the qualitative and quantitative results being presented together where they related to the same topic. Data concerning oral conditions, the impact of oral health, dental visiting behaviours, perceived barriers to dental care, self-perceived oral health, previous dental experiences, and oral hygiene behaviours are presented.

Chapter four compares these findings to current knowledge of the oral health of the New Zealand population. These findings are also discussed with reference to other haemophilia populations and other relevant international and national literature.

Chapter five draws conclusions and makes recommendations for further research.
Acknowledgements

Firstly, and most importantly, I would like to acknowledge the participants who gave up their time to take part in this study. I thoroughly enjoyed meeting and interviewing every one of them and without their willingness to take part this thesis would not exist.

My greatest thanks goes to my supervisors, Professor Murray Thomson and Dr Kate Morgaine, whose patience was unending with reading countless drafts and responding to, sometimes unrealistic, requests. I am still seeing commas in my sleep.

BJ Ramsay, Haemophilia Nurse Specialist at Capital and Coast District Health Board, was instrumental in recruiting participants and answered all sorts of questions about haemophilia whenever I thought of them. Without his enthusiasm this research would not have happened.

I am very grateful for financial support from the Ministry of Health Oral Health Research Fund administered by the New Zealand Dental Association.

Many thanks go to the staff within the Dental and Oral Health Service at Capital and Coast DHB who countenanced my arrival at all hours to examine participants.

Finally, I would like to thank the many family and friends who have taken an interest in what I have been doing for the last two and a half years and given encouragement at every turn. The best of these was Inga – I hope your thesis path is smooth.

My especial thanks and love go to Tony and Rosa who have spent many a weekend together at the zoo or Te Papa so ‘Mummy can do her work’. Thank you.
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<th>Full Form</th>
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<tbody>
<tr>
<td>CAL</td>
<td>Clinical attachment loss</td>
</tr>
<tr>
<td>CCDHB</td>
<td>Capital and Coast District Health Board</td>
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<td>DAS</td>
<td>Dental anxiety scale</td>
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<td>DDE</td>
<td>Developmental defects of enamel</td>
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<tr>
<td>DHB</td>
<td>District health board</td>
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<tr>
<td>DMFS</td>
<td>Decayed, missing and filled surfaces in the permanent dentition</td>
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<tr>
<td>dmfs</td>
<td>Decayed, missing and filled surfaces in the deciduous dentition</td>
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<tr>
<td>DMFT</td>
<td>Decayed, missing and filled teeth in the permanent dentition</td>
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<td>dmf</td>
<td>Decayed, missing and filled teeth in the deciduous dentition</td>
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<td>DS</td>
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<td>ds</td>
<td>Decayed surfaces in the deciduous dentition</td>
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<td>DT</td>
<td>Decayed teeth in the permanent dentition</td>
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<tr>
<td>dt</td>
<td>Decayed teeth in the deciduous dentition</td>
</tr>
<tr>
<td>FS</td>
<td>Filled surfaces in the permanent dentition</td>
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<tr>
<td>fs</td>
<td>Filled surfaces in the deciduous dentition</td>
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<tr>
<td>FT</td>
<td>Filled teeth in the permanent dentition</td>
</tr>
<tr>
<td>ft</td>
<td>Filled teeth in the deciduous dentition</td>
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<tr>
<td>GDP</td>
<td>General dental practitioner</td>
</tr>
<tr>
<td>HIV</td>
<td>Human immunodeficiency virus</td>
</tr>
<tr>
<td>IOTN</td>
<td>Index of orthodontic treatment needs</td>
</tr>
<tr>
<td>MS</td>
<td>Missing surfaces in the permanent dentition</td>
</tr>
<tr>
<td>ms</td>
<td>Missing surfaces in the deciduous dentition</td>
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<tr>
<td>MT</td>
<td>Missing teeth in the permanent dentition</td>
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<tr>
<td>mt</td>
<td>Missing teeth in the deciduous dentition</td>
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<tr>
<td>Acronym</td>
<td>Description</td>
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<tr>
<td>NHS</td>
<td>National Health Service</td>
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<td>NZ</td>
<td>New Zealand</td>
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<tr>
<td>NZOHS</td>
<td>New Zealand Oral Health Survey</td>
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<tr>
<td>OHIP</td>
<td>Oral Health Impact Profile</td>
</tr>
<tr>
<td>OHRQoL</td>
<td>Oral health-related quality of life</td>
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<tr>
<td>PD</td>
<td>Pocket depth</td>
</tr>
<tr>
<td>PWH</td>
<td>People with haemophilia</td>
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<tr>
<td>RAG-M</td>
<td>Research Advisory Group – Maori</td>
</tr>
<tr>
<td>RHNS</td>
<td>Regional haemophilia nurse specialist</td>
</tr>
<tr>
<td>TMJ</td>
<td>Temporomandibular joint</td>
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<tr>
<td>UK</td>
<td>United Kingdom</td>
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<tr>
<td>VAS</td>
<td>Visual analogue scale</td>
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<tr>
<td>WFH</td>
<td>World Federation of Hemophilia</td>
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<td>WHO</td>
<td>World Health Organization</td>
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</table>
1 Introduction

Those living with haemophilia can experience the same oral diseases as those without haemophilia; however, their dental treatment is complicated by prolonged bleeding and the reluctance of general dentists to treat such individuals. With these potential additional barriers to routine dental care, it is hypothesised that those with haemophilia have worse oral health than the general population.

In this section, haemophilia is described, along with its treatment and impact; the dental implications of haemophilia are outlined; a literature review of the oral health of individuals with haemophilia; and an overview of the oral health of the general New Zealand population is presented.

1.1 Haemophilia

Haemostasis in humans has been described as a complex cascade of clotting factors leading to fibrin formation. Each stage in the process shares a similar reaction whereby an inactive protein is converted to an active enzyme. Without this process the clot cannot be stabilised and bleeding is prolonged (Brummel-Ziedins et al., 2009).

Haemophilia is an X-linked recessive genetic disorder in which a clotting factor is greatly reduced or even absent. There are two types of haemophilia: Haemophilia A (missing Factor VIII) and Haemophilia B (missing Factor IX). They are indistinguishable clinically and were not identified as separate entities until 1952 (Biggs et al., 1952). Those with haemophilia are classified according to the amount of residual clotting factor as either mild (5-40% of normal level), moderate (1-5%), or severe (<1%) (Bolton-Maggs and Pasi, 2003).

Although the condition is usually familial, thirty percent of cases of haemophilia are caused by spontaneous mutations with no family history (Arruda and High, 2008;
Bolton-Maggs and Pasi, 2003; Hoyer, 1994). The incidence of haemophilia in New Zealand is 1/5000 male births, with Haemophilia A being four to six times more common than haemophilia B (Bolton-Maggs and Pasi, 2003; Park et al., 1995). Although predominantly affecting males, due to haemophilia being carried by a sex-linked recessive gene, female ‘carriers’ may display clinical signs of mild haemophilia (Friedman and Rodgers, 2009; Park et al., 1995).

Haemophilia is characterised by prolonged bleeding and even spontaneous bleeding (especially into the joints) in severe cases. The most common problem for those with haemophilia is bleeding into joints and muscles, which may lead to joint damage and loss of mobility if bleeding occurs in the same joint repeatedly (Arruda and High, 2008; Bolton-Maggs and Pasi, 2003). Those with mild haemophilia may have little spontaneous bleeding and may not be diagnosed unless they suffer major trauma or require surgical intervention (Roberts et al., 2010).

There is no cure for haemophilia. Until the 1960s, life expectancy averaged around 40 years, with the most common cause of early death being intracranial haemorrhage (Jones, 2000; Rizza and Spooner, 1983).

Treatment for haemophilia involves raising the concentration of the missing factor in the blood sufficiently to arrest bleeding or prevent spontaneous bleeding. Fifty years ago, this involved large-volume whole blood or fresh-frozen plasma transfusions often leading to circulatory overload (Jones, 2000). Haemophilia treatments have vastly improved since then, with the emergence of cryoprecipitate in the 1960s, blood derived factors in the 1970s, and recombinant factors in the 1990s, with the real possibility of gene therapy in the future (Jones, 2000). Nevertheless, modern day treatment is still not 100% effective, because inhibitors to factor proteins occur in 5-7% of individuals (12-13% of individuals with severe haemophilia), leading to complications of treatment (Wight and Paisley, 2003).
In New Zealand, there are six haemophilia centres (Auckland, Hamilton, Palmerston North, Wellington, Christchurch and Dunedin) providing multidisciplinary care for individuals with haemophilia. The New Zealand Haemophilia Foundation provides support for haemophilia patients and their families.

1.2 Dental implications of haemophilia

Routine dental appointments can result in hospital admissions because even those with mild haemophilia can experience excessive bleeding after dental procedures. In the past, this has been fatal (Archer and Zubrow, 1954).

A 1993 postal survey by Park et al of people with haemophilia (PWH) living in New Zealand (response rate 52%) investigated the type and frequency of bleeding experienced. The majority of bleeds were from joints (60%) while bleeding from the mouth or teeth accounted for only 3% of bleeds. However, the same survey identified that one-third of all surgical hospital admissions for PWH were for dental reasons. One explanation for this discrepancy in findings could be that routine dental appointments within the hospital setting were counted as hospital admissions in this data-set (Park et al., 1995).

Protocols are in place in New Zealand to prevent excessive bleeding associated with dental procedures; these include the addition of specific haemostatic treatment or overnight hospital admission (Medical Advisory Committee of the Haemophilia Foundation of New Zealand, 2005). However, this increases the cost of dental treatment and suggests that the prevention of dental disease is preferable. Prevention of dental disease is recognised as best practice by the World Federation of Hemophilia (Brewer and Correa, 2006; Scully et al., 2008), but prevention is not mentioned in New Zealand guidelines, which focus solely on the treatment of dental disease.
1.3 Oral health of individuals with haemophilia

There is very little literature available on the oral health of those with haemophilia. A literature search found only twelve studies fulfilling the search criteria¹ (Table 1). All twelve studies are of cross-sectional survey design and provide information only on the prevalence and/or severity of oral disease. Eight of the twelve studies compared a study group with a control group comprising individuals without haemophilia. No studies were found from New Zealand.

Between them, the studies investigated elements of caries, periodontal health, enamel defects (including fluorosis), malocclusion, temporomandibular joint disorders, and oral-health-related quality of life. The populations examined were demographically varied: ages ranged from 2 years to 70 years; developed and developing countries were included; and varying severity of bleeding disorder. However, there was little variation by sex/gender and, as expected, due to haemophilia’s sex-lined genetic basis, the majority of participants in these studies were male. Of interest is a Pakistani paper investigating severe haemophiliacs where 15.6% of participants were identified as female. Unfortunately, this was not explained further (Azhar et al., 2006).

¹ Search terms used: Haemophilia OR inherited bleeding disorder AND dent$ OR periodontal disease OR malocclusion OR mouth neoplasms OR temporomandibular joint OR mouth mucosa OR gingivitis OR dental caries OR oral health
<table>
<thead>
<tr>
<th>Paper</th>
<th>Type of paper</th>
<th>Type of study</th>
<th>Country</th>
<th>Study participants</th>
<th>Control participants</th>
<th>Outcomes measured</th>
</tr>
</thead>
<tbody>
<tr>
<td>Boyd and Kinirons, 1997</td>
<td>Refereed paper</td>
<td>Cross-sectional survey comparison</td>
<td>Northern Ireland</td>
<td>38 haemophiliacs 2-15 years</td>
<td>Weighted national survey</td>
<td>Caries</td>
</tr>
<tr>
<td>Meilnik-Blaszczak, 1999</td>
<td>Refereed paper</td>
<td>Cross-sectional survey comparison</td>
<td>Poland</td>
<td>80 individuals with inherited bleeding disorders 4-18 years</td>
<td>80 matched controls 4-18 years</td>
<td>Caries</td>
</tr>
<tr>
<td>Sonbol et al, 2001</td>
<td>Refereed paper</td>
<td>Cross-sectional survey comparison</td>
<td>UK</td>
<td>38 severe haemophiliacs 4-13.6 years mean 8.8 ± 3.4</td>
<td>30 matched controls 4-13.6 years mean 9.8 ± 3.1</td>
<td>Caries, Periodontal health, Developmental enamel defects</td>
</tr>
<tr>
<td>Fiske et al, 2002</td>
<td>Refereed paper</td>
<td>Cross-sectional survey</td>
<td>UK</td>
<td>50 individuals with inherited bleeding disorders 3-16-70 years</td>
<td>No controls</td>
<td>Caries</td>
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<tr>
<td>Ugolotti et al, 2004</td>
<td>Report of presentation</td>
<td>Cross-sectional survey comparison</td>
<td>Italy</td>
<td>65 individuals with inherited bleeding disorders 4-6-16 years and 30-55 years</td>
<td>24 healthy controls</td>
<td>Caries, Periodontal health</td>
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<td>Albrayrak et al, 2006</td>
<td>Report of presentation</td>
<td>Cross-sectional survey comparison</td>
<td>Turkey</td>
<td>36 type A haemophiliacs 6-12 years</td>
<td>36 healthy controls 6-12 years</td>
<td>Caries, Periodontal health</td>
</tr>
<tr>
<td>Azhar et al, 2006</td>
<td>Refereed paper</td>
<td>Cross-sectional survey comparison</td>
<td>Pakistan</td>
<td>52 severe haemophiliacs college students mean age 16.59 ± 3.24</td>
<td>192 matched college students mean age 16.70 ± 2.95</td>
<td>Caries, Periodontal health, Malocclusion, TMJD</td>
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</table>

2 77 with haemophilia, 3 with von Willebrands disease  
3 28 with haemophilia, 16 with von Willebrands disease and 6 others not differentiated  
4 Not specified
<table>
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<tr>
<th>Paper</th>
<th>Type of paper</th>
<th>Type of study</th>
<th>Country</th>
<th>Study participants</th>
<th>Control participants</th>
<th>Outcomes measured</th>
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<tr>
<td>Hanagavadi, 2006</td>
<td>Report of presentation</td>
<td>Cross sectional survey</td>
<td>India</td>
<td>30 haemophiliacs 15-25 years</td>
<td>No controls</td>
<td>Caries, Periodontal health</td>
</tr>
<tr>
<td>Kabil et al, 2007</td>
<td>Refereed paper</td>
<td>Cross sectional survey (part one of a randomised control trial)</td>
<td>Egypt</td>
<td>60 haemophiliacs 6-12 years means 8.52 and 7.03</td>
<td>No controls</td>
<td>Caries</td>
</tr>
<tr>
<td>Rodrigues, 2008</td>
<td>English abstract only</td>
<td>Cross-sectional survey</td>
<td>Brazil</td>
<td>40 haemophiliacs 3-12 years</td>
<td>No controls</td>
<td>Caries</td>
</tr>
<tr>
<td>Alpkilic Baskirt et al, 2009</td>
<td>Refereed paper</td>
<td>Cross-sectional survey comparison</td>
<td>Turkey</td>
<td>71 haemophiliacs 14-35 years mean 23 ±6.58</td>
<td>60 controls 14-35 years mean 21 ± 6.45</td>
<td>Oral-health related quality of life</td>
</tr>
<tr>
<td>Ziebolz et al, 2011</td>
<td>Refereed paper</td>
<td>Cross-sectional survey comparison</td>
<td>Germany</td>
<td>15 individuals with inherited bleeding disorders 18-60 years mean 39.2 ± 8.3</td>
<td>31 controls mean age 36.4 ± 9.6</td>
<td>Caries, Periodontal health</td>
</tr>
</tbody>
</table>

5 baseline results recorded from two groups with no significant difference between groups
6 8 with haemophilia, 7 with von Willebrands disease
1.3.1 Dental caries in individuals with haemophilia

Dental caries was the most common outcome investigated in studies of the oral health of individuals with haemophilia included in the literature review. This outcome was investigated by eleven authors (Table 2). The popular choice of ‘severity of caries’ as an outcome variable is unsurprising because indicators of the prevalence and severity of caries use good, well-recognised indices, and most countries collect these data at age twelve to send to the World Health Organization (WHO).

The severity of caries is represented by the mean number of decayed, missing or filled teeth (DMFT for permanent teeth and dmft for primary teeth). A wide range of severity of caries was found, from a mean DMFT of 0.5 (Boyd and Kinirons, 1997) to a mean DMFT of 18 (Ziebolz et al., 2011). Both of the studies reporting these DMFT extremes were carried out in Europe. The former was a child population assessed using WHO guidelines, with cavitation as the threshold of caries, while the Ziebolz et al. paper was based on an adult population and did not state the threshold at which caries was determined. Unfortunately, in three studies where DMFT had been recorded, these figures were not reported (Albayrak et al., 2006; Hanagavadi, 2006; Ugolotti et al., 2004).

The caries distribution is usually highly skewed, and therefore the percentage of the population who are caries-free (or the prevalence of caries, which is the inverse of the percentage caries-free) is often also quoted to give more information on the distribution. Caries prevalence in these studies ranged from 27% (Boyd and Kinirons, 1997) to 94% (Mielenik-Blaszczak, 1999).

In the seven studies where comparisons were made with healthy individuals, two found the study group to have less caries (Boyd and Kinirons, 1997; Sonbol et al., 2001), two found the study group to have more caries (Albayrak et al., 2006; Azhar et al., 2006) and three found no difference between the groups (Mielenik-Blaszczak, 1999; Ugolotti et al., 2004; Ziebolz et al., 2011) (Table 2).
The findings of these twelve studies vary considerably according to the population investigated. This heterogeneity in findings may be due to artefacts of the quality of the studies, confounding factors or actual differences in the populations involved. It is known that low socio-economic status is a risk factor for caries (National Advisory Committee on Health and Disability, 2003; Schou and Uitenbroek, 1995; Thomson et al., 2004), but only two studies considered this. Rodrigues found a higher mean DMFT in children whose mothers had not completed primary education (Rodrigues et al., 2008). Albayrak et al (2006) found that the socio-economic status of the study group was significantly lower than that of the control group but did not control for this in the analysis (Albayrak et al., 2006), meaning that no useful inferences can be taken from that study.

Five studies examining children all used the WHO standards; however, these are still not easily comparable because the DMFT/dmft values given were averages from the whole sample and not broken down to specific ages; only one study provided a breakdown of the study population by age (Boyd and Kinirons, 1997). The reason for this method of reporting is the often small number of individuals with haemophilia (for instance, Boyd reported just a single five-year-old) making it highly unlikely appropriate estimates could be identified for each age group.
<table>
<thead>
<tr>
<th>Paper</th>
<th>Age</th>
<th>Study group</th>
<th>Caries severity</th>
<th>Caries prevalence</th>
<th>Age</th>
<th>Comparison group</th>
<th>Caries severity</th>
<th>Caries prevalence</th>
<th>Other results found</th>
<th>Significant findings of comparison</th>
</tr>
</thead>
<tbody>
<tr>
<td>Boyd and Kinirons, 1997</td>
<td>2-10 years</td>
<td>dmft 0.86 (1.33)</td>
<td>33% (primary dentition)</td>
<td>2-10 years</td>
<td>dft 1.60</td>
<td>DMFT 2.3</td>
<td>55% (primary dentition)</td>
<td>59% (secondary dentition)</td>
<td>100% study population had no untreated caries in secondary dentition Study group had no teeth extracted due to caries</td>
<td>Study group had less children with any caries experience and less children with untreated caries (both primary and secondary dentition)</td>
</tr>
<tr>
<td></td>
<td>7-15 years</td>
<td>dft 0.86 (1.33)</td>
<td>27% (secondary dentition)</td>
<td>äche 2-10 years</td>
<td>DMFT 0.45</td>
<td></td>
<td></td>
<td></td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
</tr>
<tr>
<td>Meilnik-Blaszcak, 1999</td>
<td>4-18 years</td>
<td>dmfs 10.1 (13.5)</td>
<td>94%</td>
<td>4-18 years</td>
<td>dmfs 10.2 (8.62)</td>
<td>DMFS 8.5 (8.42)</td>
<td>DMFT 5.4</td>
<td>96%</td>
<td>No difference in caries prevalence or severity between groups</td>
<td>No difference in caries prevalence or severity between groups</td>
</tr>
<tr>
<td></td>
<td></td>
<td>dmf 4.1</td>
<td></td>
<td></td>
<td>dmf 4.6</td>
<td></td>
<td></td>
<td></td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
</tr>
<tr>
<td></td>
<td></td>
<td>DMFS 9.6 (10.5)</td>
<td></td>
<td></td>
<td>DMFS 8.5</td>
<td></td>
<td></td>
<td></td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
</tr>
<tr>
<td></td>
<td></td>
<td>DMFT 5.8</td>
<td></td>
<td></td>
<td>DMFT 5.4</td>
<td></td>
<td></td>
<td></td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
</tr>
<tr>
<td>Sonbol et al, 2001</td>
<td>4-13 years</td>
<td>dmfs 6.1 (11.1)</td>
<td>63.3%</td>
<td>4-15 years</td>
<td>dmfs 5.5 (6.2)</td>
<td>DMFS 3.6 (3.8)</td>
<td>DMFT 2.4 (2.8)</td>
<td>86.7%</td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
</tr>
<tr>
<td></td>
<td></td>
<td>dmf 2.3 (2.8)</td>
<td></td>
<td></td>
<td>dmf 2.9 (3.7)</td>
<td></td>
<td></td>
<td></td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
</tr>
<tr>
<td></td>
<td></td>
<td>DMFS 0.8 (1.5)</td>
<td></td>
<td></td>
<td>DMFS 3.6</td>
<td></td>
<td></td>
<td></td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
</tr>
<tr>
<td></td>
<td></td>
<td>DMFT 0.7 (1.3)</td>
<td></td>
<td></td>
<td>DMFT 2.4</td>
<td></td>
<td></td>
<td></td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
<td>Study group had lower DMFT, DMFS and lower caries prevalence</td>
</tr>
<tr>
<td>Fiske et al, 2002</td>
<td>16-70 years</td>
<td>DMFT 14.4 (range 0-36)</td>
<td>Not stated</td>
<td></td>
<td>No comparison</td>
<td></td>
<td></td>
<td></td>
<td>No difference in caries prevalence between groups</td>
<td>No difference in caries prevalence between groups</td>
</tr>
<tr>
<td></td>
<td></td>
<td>DT 3 (range 0-8)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Control group had lower DMFS/T (no difference in dmfs/t)</td>
<td>Control group had lower DMFS/T (no difference in dmfs/t)</td>
</tr>
<tr>
<td>U golotti et al, 2004</td>
<td>Not stated</td>
<td>Not stated</td>
<td>Not stated</td>
<td>Not stated</td>
<td>Not stated</td>
<td></td>
<td></td>
<td></td>
<td>No difference in caries prevalence between groups</td>
<td>No difference in caries prevalence between groups</td>
</tr>
<tr>
<td>Al bayrak et al, 2006</td>
<td>Not stated</td>
<td>Not stated</td>
<td>Not stated</td>
<td>Not stated</td>
<td>Not stated</td>
<td></td>
<td></td>
<td></td>
<td>Control group had lower DMFS/T (no difference in dmfs/t)</td>
<td>Control group had lower DMFS/T (no difference in dmfs/t)</td>
</tr>
<tr>
<td>Paper</td>
<td>Age</td>
<td>Study group Caries severity</td>
<td>Caries prevalence</td>
<td>Age</td>
<td>Comparison group Caries severity</td>
<td>Caries prevalence</td>
<td>Other results found</td>
<td>Significant findings of comparison</td>
<td></td>
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<td></td>
<td></td>
</tr>
<tr>
<td>Azhar et al, 2006</td>
<td>13-23 years</td>
<td>DMFT 2.07</td>
<td>57.7%</td>
<td>13-12 years</td>
<td>DMFT 0.95</td>
<td>40.63%</td>
<td>Control group had lower proportion of individuals with DMFT 6-15</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hanagavadi 2006</td>
<td>Not stated</td>
<td>No comparison</td>
<td>Untreated caries found in 40% participants</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Kabil et al, 2007</td>
<td>6-12 years</td>
<td>deft 5.91 (3.05) and 5.84 (2.62)</td>
<td>No comparison</td>
<td>No primary teeth restored or missing (all caries was untreated)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Rodrigues, 2008</td>
<td>3-12 years</td>
<td>dmft 2.00</td>
<td>No comparison</td>
<td>Caries more severe when parents had not completed primary school education</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Ziebolz et al, 2011</td>
<td>18-60 years</td>
<td>DMFT 18</td>
<td>18-60 years</td>
<td>DMFT 15</td>
<td>No difference between groups</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

**Table:** Caries severity and prevalence findings from different studies, along with other relevant results. The table includes age ranges, DMFT and dmft scores, and findings regarding caries severity and treatment outcomes.
Caries initiation and development is a complex interaction of protective factors (fluoride, calcium phosphate) and destructive factors (cariogenic bacteria and sugar consumption). This review did not consider any determinants of oral health in the inclusion criteria, so, although some of these aspects are covered in some of these studies, it is possible that other authors have looked at these issues and the review in this area is incomplete. Bacterial counts were measured by Sonbol et al (2001) as a proxy for caries risk. The control group was found to have higher levels of streptococci bacteria, as well as more caries. In a Turkish study, sugar consumption was found to be higher in the study population, who also had more caries (Albayrak et al., 2006).

One population, part of a comprehensive, dedicated preventive dental programme in Northern Ireland (Boyd and Kinirons, 1997), had no active caries at all. In contrast, PWH in Egypt were found to have active caries in the primary dentition but no treatment (fillings or extractions) had been carried out (Kabil et al., 2007). It may be that no treatment had been completed because the children had not visited a dentist; the lack of dental services for PWH in developing countries is lamented in this paper.

In summary, it appears that in those countries where an intensive dental programme is provided for those with haemophilia, the prevalence and severity of caries is lower for (than in a comparison group).

1.3.2 Periodontal condition in individuals with haemophilia

Six of the twelve reviewed papers investigated periodontal conditions (Table 3). However, the majority used a visual index of inflammation only, with no probing depth or loss of attachment measurements. One study used radiographic measures of bone loss (Ziebolz et al., 2011). The study that carried out probing (Ugolotti et al., 2004) did not properly describe what was measured and did not report any values. In fact, three of the six studies investigating periodontal condition did not report any values and the three that did used different methods of reporting their results. The failure to probe may be due to fear of causing bleeding by probing and the ethical considerations associated with this, but none of the studies explained why pocket depth probing was not carried
out. The use of radiography can give a measure of bone loss without the need for probing but in itself raises ethical issues.

The study comparing radiographic measures of bone loss found that PWH had more alveolar bone loss (although this was not clinically significant) and a higher prevalence of sites with bone loss (Ziebolz et al., 2011).

Of the five studies that compared periodontal condition between populations, two found the control group had better gingival health (Albayrak et al., 2006; Ugolotti et al., 2004), two found no difference (Azhar et al., 2006; Sonbol et al., 2001), and one found that the control group had more attachment loss (Ziebolz et al., 2011) (Table 3).

Although the evidence is sparse, it appears that ‘healthy’ controls have less gingival inflammation than PWH. There are several possible reasons for this, including confounding factors, such as lifestyle differences like smoking and oral hygiene practices; and biological determinants, that is, a difference in underlying susceptibility.
## Table 3 - Summary of studies that investigated periodontal health

<table>
<thead>
<tr>
<th>Paper</th>
<th>Outcomes measured</th>
<th>Study group</th>
<th>Comparison group</th>
<th>Significant findings of comparison</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sonbol et al, 2001</td>
<td>Gingival index score Mean score (SD)</td>
<td>0.6 (2.4) primary dentition</td>
<td>1.0 (2.8) primary dentition</td>
<td>No difference in gingival index score between groups</td>
</tr>
<tr>
<td></td>
<td></td>
<td>1.3 (4.2) secondary dentition</td>
<td>3.8 (8.6) secondary dentition</td>
<td>Control group had lower gingival index scores</td>
</tr>
<tr>
<td>Ugolotti et al, 2004</td>
<td>Loe Gingival Index Partial probing</td>
<td>Not stated</td>
<td></td>
<td>No difference in probing between groups</td>
</tr>
<tr>
<td>Albayrak et al, 2006</td>
<td>Gingival index</td>
<td>Not stated</td>
<td></td>
<td>Control group had lower gingival index scores</td>
</tr>
<tr>
<td>Azhar et al, 2006</td>
<td>Modified gingival index</td>
<td>Not stated</td>
<td></td>
<td>No difference in gingival index score between groups</td>
</tr>
<tr>
<td>Hanagavadi, 2006</td>
<td>Gingival Index</td>
<td>57% participants had mild gingivitis</td>
<td>No comparison</td>
<td>Study group had higher prevalence of severe or moderate periodontal disease</td>
</tr>
<tr>
<td></td>
<td></td>
<td>27% participants had moderate gingivitis</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Ziebolz et al, 2011</td>
<td>Alveolar bone loss (mm) Prevalence of periodontal disease</td>
<td>2.73mm (0.73) 80%</td>
<td>2.00mm (0.77) 48%</td>
<td></td>
</tr>
</tbody>
</table>
1.3.2.1 Lifestyle factors and individuals with haemophilia

Four studies asked participants about oral hygiene practices (Albayrak et al., 2006; Alpkilic Baskirt et al., 2009; Azhar et al., 2006; Kabil et al., 2007). Of these, the most striking finding was that 48.3% of Egyptian children with haemophilia did not carry out any oral hygiene practices (Kabil et al., 2007). Alpkilic Baskirt et al (2009) found that controls were more likely to be regular brushers.

Seven studies included in this review considered oral hygiene. Two papers found that the study group had more plaque than the controls (Mielnik-Blaszczak, 1999; Ugolotti et al., 2004), one found the study group had less plaque than the controls (Ziebolz et al., 2011) one found similar amounts of plaque as the controls (Albayrak et al., 2006), two did not compare groups (Hanagavadi, 2006; Kabil et al., 2007), and one did not report those findings (Azhar et al., 2006).

None of the studies considered the smoking status of participants.

1.3.2.2 Biological factors and individuals with haemophilia

Those with haemophilia who were part of a targeted prevention programme had the same gingival index scores as healthy individuals, suggesting that, although targeted programmes may result in less caries, they may not result in healthier gingivae (Sonbol et al., 2001). One theoretical reason for this lack of improvement in gingivae may be an (as yet unidentified) underlying predisposition to periodontal disease linked to haemophilia.

One study reported on oral hygiene practices, plaque levels and gingival inflammation (Albayrak et al., 2006). It found the controls brushed more frequently than children with haemophilia but had similar levels of plaque and less inflammation. In general, patients with more inflammation (such as the study group in that paper) will find their gingivae will bleed more, which may cause some individuals to stop performing oral
hygiene procedures. Gingival inflammation is caused by the presence of plaque but is modified by the underlying susceptibility of the individual. Similar levels of plaque leading to more inflammation in the study group may indicate that haemophilia changes the underlying susceptibility of the individual.

1.3.3 Other measures of oral health in individuals with haemophilia

Four studies investigated other aspects of oral health (Table 4). These other aspects include temporomandibular joint (TMJ) dysfunction, malocclusion, enamel defects and oral-health-related quality of life (OHRQoL).

Individuals with haemophilia were found to have a higher prevalence of TMJ dysfunction (Azhar et al., 2006). This may indicate haemarthrosis occurring, because the participants in this study were severe haemophiliacs (residual clotting factor <1%), and spontaneous bleeding into joints is a feature of severe haemophilia. However, the total numbers were small and bleeding into joints is more common in the knees, ankles and elbows (Park et al., 1995), so this finding could merely be due to chance (type I error\(^7\)).

Azhar et al (2006) also found that haemophiliacs had a higher orthodontic treatment need. This may be due to reluctance among orthodontists to treat these patients, or patients’ reluctance to seek more medical interventions (or, again, an artefact of the study).

\(^7\) A type 1 error is one where the null hypothesis is rejected incorrectly
Table 4 - Summary of studies which investigated other measures of oral health

<table>
<thead>
<tr>
<th>Paper</th>
<th>Area of interest</th>
<th>Outcomes measured</th>
<th>Significant findings of comparison</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sonbol et al, 2004</td>
<td>Developmental defects of enamel</td>
<td>DDE</td>
<td>No significant difference between groups found</td>
</tr>
<tr>
<td>Azhar et al, 2006</td>
<td>Symptoms of TMJ dysfunction</td>
<td>Questionnaire, form unspecified</td>
<td>Study group had higher prevalence of existing TMJ symptoms</td>
</tr>
<tr>
<td></td>
<td>Malocclusion</td>
<td>IOTN</td>
<td>Study group had higher orthodontic treatment needs</td>
</tr>
<tr>
<td>Hanagavadi, 2006</td>
<td>Fluorosis</td>
<td>Deans Index</td>
<td>No comparison made</td>
</tr>
<tr>
<td>Alpkilic-Baskirt et al, 2009</td>
<td>Oral health-related quality of life</td>
<td>OHIP, OHQoL-UK, VAS of oral health status</td>
<td>Study group perceived lower oral health-related quality of life</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Study group perceived lower oral health status</td>
</tr>
</tbody>
</table>

Developmental defects of enamel (DDE), or disruptions in the normal formation of enamel, can be caused by a variety of factors such as excess of systemic fluoride, local or systemic trauma, or local or systemic infection (Soames and Southam, 2005). Children with haemophilia who are in a high-intensity prevention programme may be exposed to higher fluoride levels, thus leading to fluorosis. Conversely, it may be the case that those in prevention programmes have less caries experience and therefore less periapical infection in primary teeth and subsequently fewer DDE. An Indian study found fluorosis in 20% of PWH (Hanagavadi, 2006), which is lower than New Zealand estimates of 40% to 50% (Mackay and Thomson, 2005; Schluter et al., 2008) in the general population. Unfortunately, this result was not compared with a control population which, as fluorosis is considered endemic in some areas of India (Meenakshi and Maheshwari, 2006), does not allow these estimates to be considered in context. Sonbol et al (2001) found that the prevalence of developmental defects in enamel did not differ between groups.

It is worth noting that health cannot be defined purely as the absence of disease; it also encompasses subjective elements such as pain, discomfort and the ability to function in society. These patient-reported outcome variables are important indicators alongside
the more objective/clinical outcome variables described above. One study investigated oral-health-related quality of life using the Oral Health Impact Profile (OHIP-14) and the oral-health-related quality of life (OHQoL-UK) indices (Alpkilic Baskirt et al., 2009). It found that PWH perceived their OHRQoL (and general quality of life) to be poorer than those without haemophilia. Interestingly, the OHIP score recorded for the control population in this group (8.4) is similar to that reported from a New Zealand general population (8.0) (Lawrence et al., 2008).

No studies were found that investigated oral cancer or mucosal disease in PWH. The epidemiology of these conditions is fraught with difficulties even in ‘healthy’ populations due to their low incidence and difficulties with diagnosis, so this is perhaps unsurprising.

1.4 Oral health of the general New Zealand population

This section summarises what is known about the oral health of the New Zealand population. Where significant differences between males and females have been reported, these are also presented, in order to aid comparison with the haemophilia population who are all male.

The oral health of New Zealand children at ages five and twelve has been recorded by the Ministry of Health for many years (Ministry of Health, 2010a) but data concerning the oral health of other age groups, including adults, have been sparse until recently. Previous national oral-health surveys were held in 1976 (SAOH) (Cutress et al., 1979) and 1988 (SOHO) (Hunter et al., 1992). SAOH used a sample of New Zealand adults over the age of 15 years while SOHO sampled adults in the age groups 20-24, 35-44 and 65-74. Smaller surveys were held in 1962 (Beck, 1968) and 1982 (MAOH) (Cutress et al., 1983), but the samples were not representative. In 2009 the third national survey of the oral health of the New Zealand population (NZOHS) was held (Ministry of Health, 2010b). This involved nearly 5000 interviews and over 3000 clinical examinations.
1.4.1 Oral health status of New Zealand children

Historical data from the Ministry of Health for children aged five and twelve show that the oral health of those aged twelve has gradually improved since 2008, while that of five-year-olds has remained static over the same period (Ministry of Health, 2010a).

The 2009 New Zealand Oral Health Survey (Ministry of Health, 2010b) found that half of all New Zealand children (49.3%) were caries-free while one in six children (15.9%) had untreated dental caries. Less caries was seen in permanent teeth than in deciduous teeth, with both the mean number of teeth with caries experience being fewer (1.2 and 1.6 respectively) and the proportion of children with untreated caries being lower (7.6% and 16.6% respectively). Data from this national survey show that, although inequalities are not as wide in school-age children as in adults, significant inequalities in oral health still exist, with Maori children being more likely to have untreated caries and to have more caries experience.

1.4.2 Oral health status of New Zealand adults

Some 9.4% of New Zealand adults were edentulous. Of those with some teeth remaining, 61.8% had one or more missing teeth, while 88.6% had a ‘functional dentition’ (defined as 21+ natural teeth). Those living in deprived neighbourhoods and Maori were more likely to be edentulous.

1.4.2.1 Caries experience of New Zealand adults

Caries is widely prevalent in New Zealand adults. The proportion of adults who were caries-free was presented only for those aged 18-24 years, with it being 22.7%.

Adults had an average of 13.9 teeth with some caries experience. Men were found to have significantly fewer filled teeth than women (8.0 and 8.9 respectively) and more
decayed teeth than women (1.0 and 0.7 respectively) when adjusted for age, and to have experienced less caries overall (DMFT 13.6 and 14.1 respectively).

One-third of adults (35.3%) and significantly more males than women (41.5% and 29.7% respectively) had some untreated caries, with male adults, Maori and Pacific adults and those living in deprived neighbourhoods more likely to have untreated caries. Fewer adults had untreated root caries (9.5%); again, significantly more males than females had untreated root caries (12.2% and 7.0 respectively).

1.4.2.2 Periodontal condition of New Zealand adults

Periodontal disease is characterised by attachment loss and the development of pockets. This is usually reported at different thresholds of pocket depth and attachment loss. As the threshold rises the prevalence decreases.

One-third of adults (33.5%) had one or more pockets of at least 4mm; this proportion was 10.5% for pockets 5mm or more and 5.1% for pockets 6mm or more. When considering attachment loss half of all adults (49.9%) had one or more sites with 4mm or more attachment loss, 27.5% at 5mm or more attachment loss, and 13.5% with 6mm or more attachment loss. The prevalence of periodontal disease was found to be significantly greater in men than women over all measures (except for the experience of pockets which were 6mm or more).

The NZOHS did not present data for gingival bleeding or for oral hygiene in adults.

1.4.3 Self-reported oral health of the New Zealand population

The NZOHS found that the parents of 7% of children reported that their child had had oro-facial pain in the previous year, while 12.6% of parents reported that their child’s oral health was fair or poor. In the previous four weeks, 25.4% of adults had experienced oro-facial pain, while 26.6% of adults (and significantly more males, at 30.6%) described their oral health as fair or poor. Nearly one-half of adults (45.9%) felt that they currently needed dental treatment (this estimate was not reported for children).
The impact of oral health was recorded by the Oral Health Impact Profile (OHIP), a validated measure of oral-health-related quality of life which consistently shows an association between OHIP scores and oral health, with lower OHIP scores strongly associated with better oral health (Slade and Spencer, 1994). Respondents answering ‘often’ or ‘very often’ to one or more of the fourteen statements are counted as having one or more impacts. In the NZOHS, 15.6% of adults (with significantly fewer males at 12.4%) were reported as experiencing one or more impacts over the previous twelve months.

1.4.4 *Oral hygiene practices in the New Zealand population*

Two out of three New Zealand adults (65.3%) and children (63.5%) brushed their teeth twice a day or more. Similar numbers of six-year-olds in Auckland (66.8%) also brushed their teeth twice a day or more (Paterson et al., 2011). Men and boys were significantly less likely to brush their teeth than women or girls. Current Ministry of Health recommendations are that teeth are brushed twice a day with a fluoride toothpaste (New Zealand Guidelines Group, 2009). One-third of the New Zealand population are falling short of this.

1.4.5 *Dental visiting by the New Zealand population*

Four out of five children (81.2%) had visited a dental professional in the previous year, with the highest attendance rate in children aged 5-11 years (90.3%) and the lowest in pre-schoolers (59.7%). Fewer than half of all adults (47.1%) had visited a dental professional in the last year, with significantly fewer men (44.1%) (Ministry of Health, 2010b).

Of the adults who had not visited a dental professional in the previous year, 47.5% reported that they had not visited because they had had no dental problems and 46.8% had not visited due to cost. In addition, 44.1% of adults (39.6% of males) reported they had avoided dental care in the previous year due to cost; 25.3% (22.5% of males) had
gone without recommended treatment due to the cost. When asked why they usually visited a dental professional, 38.9% of adults (36.4% of males) reported that they usually visited for a check-up rather than a dental problem, and 55.3% felt that they did not see a dental professional often enough.

1.4.6 Summary

There has been little research on the oral health of individuals with haemophilia, and none at all conducted in New Zealand. Limited conclusions can be drawn about the oral health of this population because much of the evidence that is available is of poor quality and is heterogeneous in setting, participants and measurements. However, the evidence that is available suggests that PWH are likely to have worse oral health than the general population.

Poor oral health can cause pain, infection, long-term morbidity and, in rare cases, death (Currie and Ho, 1993; Green et al., 2001). This has an impact on the individual, both in terms of quality of life and the resources required to deal with these problems. Moreover, the treatment of oral disease in those with haemophilia puts additional burden onto the healthcare provider in terms of hospital use and factor costs. Because PWH are living longer (Franchini and Mannucci, 2010), these costs have become more important.

While there is no known biological reason (such as altered tooth tissue structure or immune response) for the oral health of PWH to be different from the general population, the features of this chronic condition and its treatment may lead to different oral health care behaviours, attitudes, barriers and clinical outcomes.
1.5 Aims

In order to help improve the provision of oral healthcare to individuals with haemophilia, the aims of this research are to:

- describe the oral health and oral health behaviours of those with haemophilia;
- compare these findings with estimates from the New Zealand population;
- explore the perceptions of oral health and its importance expressed by those with haemophilia; and
- identify the barriers to oral health perceived by those with haemophilia.
2 Methods

This section outlines the methods used in this study, including study design, ethical considerations, participant selection and recruitment, data collection, and analysis.

2.1 Design

An embedded correlational mixed methods design was used to research the oral health of those with haemophilia in New Zealand.

The choice of methodology and methods used to answer research questions should be guided by the epistemology (theory of knowledge) and theoretical perspective of the topic. Clinical science, such as dentistry, sits in a post-positivist theoretical framework where, while it is believed that reality is based in data, there is also acceptance that there are things that cannot be known, observed, or quantified (Crotty, 1998). This pragmatic approach to the research problem is reflected in the choice of a mixed methods methodology for producing evidence to address the specific research questions. This ‘embedded’ design is led by a quantitative approach and supported by qualitative methods, bringing breadth and depth to the study. In this design, the primary, quantitative data-set is supported by the secondary, qualitative data-set, and questions requiring the interrogation of the different data-sets can be investigated.

The researcher in this study is a hospital dentist with an interest in public health. The researcher had a prior dentist-patient relationship with some participants.

2.2 Ethical considerations

Prior to the collection of data, the ethical issues of this research were considered. The Health and Disability Ethics Committees of the NZ Ministry of Health are responsible for granting ethical approval of research projects. Ethical approval was granted by the
Central Regional Ethics Committee in August 2010 (Ref CEN/10/05/015) (Appendix 1).

Cultural beliefs must also be considered, with special recognition of Māori culture and the principles of the Treaty of Waitangi. Researchers within Capital and Coast District Health Board (CCDHB) can work with the “Research Advisory Group – Maori” (RAG-M), a subcommittee of the Maori Partnership Board, in order to engage Maori input and support for their research. This group is mandated by all local iwi in the CCDHB region. Approval from the RAG-M was obtained in June 2010 (Appendix 2).

In addition, support was also sought from the Ngāi Tahu Research Consultation Committee in Otago because the research was carried out under the umbrella of the School of Dentistry in Dunedin. Support from the Ngāi Tahu Research Consultation Committee was given in February 2010 (Appendix 3).

Support was also obtained from the Haemophilia Foundation of New Zealand (Appendix 4). The Haemophilia is a community agency providing education, care and support for people with haemophilia in New Zealand.

2.3 Participants

2.3.1 Inclusion criteria

All individuals with moderate or severe haemophilia, over the age of two, and residing in the Capital & Coast, Hutt Valley or Wairarapa District Health Board regions, were eligible to participate. These individuals were identified from their local registers by the Regional Haemophilia Nurse Specialists (RHNS) covering this region. There were no exclusion criteria.
2.3.2 Recruitment

The thirty-eight potential participants meeting the study eligibility criteria were sent an introductory letter and information sheet by the RHNS, inviting them to participate in the study. Letters were sent out in batches of two or three, rather than all at once, to allow timely examinations and interviews with those who agreed to participate. Those interested in participating were invited to contact either the study principal investigator or the RHNS. Those who did not reply to the initial letter were followed up by a phone call from the RHNS after two weeks.

The principal investigator telephoned all potential participants who expressed an interest in participating. Consent and confidentiality issues were considered and highlighted during recruitment procedures. All potential participants were given a study information sheet (Appendix 5) and were allowed time to consider their participation in the study. Written consent was obtained by participants and/or their parent/guardian freely signing the study consent form (Appendix 6). There were four different information sheets and consent forms available depending on the individual’s age.

Demographic details were collected (age, distance residing from hospital, rural status of residence, ethnicity, severity of haemophilia) for those who declined to participate (or who did not respond) so that comparisons could be made between participants and non-participants.

It was originally intended that 25 of the individuals who agreed to participate in the study would be purposively sampled using maximum variation sampling (stratified by specific demographic variables including age, severity of haemophilia, geographic location, ethnicity) in order to ensure a wide cross-section of individuals. However, due to the small number of participants and the staggered nature of recruitment, all individuals were invited to participate in a semi-structured interview.

All participants completing any element of the research were given a $20 voucher towards the costs of attending the research appointment.
2.4 Data collection

Data collection was through three component parts: a questionnaire, a clinical examination and a semi-structured interview, administered in that order. Data from each study participant were linked using a confidential, unique ID number.

Confidentiality was maintained through all data being stored securely and electronic data being password-protected. Data were identified only by an identification code, the key to which was stored separately and only accessible by the researchers. It was destroyed after the research was complete. Linked, de-identified data will continue be kept securely for an indefinite period in order to allow future analysis and historical comparison.

2.4.1 Questionnaire

All participants were asked tick-box questions which were similar to those asked in the National Oral Health Survey (questions utilised with permission from the Health and Disability Intelligence Unit, Health and Disability Systems Strategy Directorate of the Ministry of Health). Two different questionnaires were used, one for participants 15 years and older and one for the parents of participants fourteen years and under, with an optional part for participants aged 9-14 years. The questionnaires included questions about self-perceived oral health (including the Oral Health Impact Profile) and oral health behaviours, in addition to demographic questions (Appendix 7 and Appendix 8). Answers were entered directly into a spreadsheet (Microsoft Office Excel 2007).

2.4.1.1 The Oral Health Impact Profile (OHIP)

The OHIP (Slade and Spencer, 1994) is a validated oral-health-related quality of life measure comprising of fourteen questions in the seven domains of functional limitation, pain, psychological discomfort, physical disability, psychological disability, social disability, and handicap. Each statement is answered on a five point Likert response scale with each response given a score; ‘never’ (scores 1), ‘hardly ever’ (2), ‘occasionally’ (3), ‘fairly often’ (4) and ‘very often’ (5).
OHIP data can be presented as the proportion of individuals who have experienced one or more impact – those who responded with ‘fairly often’ or ‘very often’ to one or more statements – or can be presented as a mean OHIP score either overall, or for each domain.

2.4.1.2 The Dental Anxiety Score

The Corah Dental Anxiety Score (DAS) is a well-validated method of assessing dental anxiety (Corah, 1969; Corah et al., 1978). It consists of four questions each answered on a five-point scale. The scores for the responses to the questions are summed to give a DAS score which can range from 4 (least anxious) to 20 (most anxious).

The severity of dental anxiety is represented by the DAS score. The prevalence of dental anxiety is given as the proportion of the population with a DAS score higher than a threshold, usually 13 (Corah et al., 1978).

2.4.2 Clinical examination

All participants were invited to undergo a clinical examination that included surface-level data on dental caries, and, for those aged over 15 years, gingival recession and probing depth at three sites per tooth.

Following standard World Health Organization guidelines for epidemiological surveys, caries were to be identified at the cavitated stage only (World Health Organization, 1997). Each tooth and tooth surface was coded as described in Table 5.
Table 5 - Criteria used to code teeth and tooth surfaces

<table>
<thead>
<tr>
<th>Code</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Tooth Code</strong></td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>Present permanent tooth</td>
</tr>
<tr>
<td>2</td>
<td>Missing tooth with no replacement</td>
</tr>
<tr>
<td>3</td>
<td>Present deciduous tooth</td>
</tr>
<tr>
<td>5</td>
<td>Carious retained root</td>
</tr>
<tr>
<td>6</td>
<td>Missing tooth replaced by bridge pontic</td>
</tr>
<tr>
<td>7</td>
<td>Missing tooth replaced by acrylic partial denture</td>
</tr>
<tr>
<td>8</td>
<td>Missing tooth replaced by metal partial denture</td>
</tr>
<tr>
<td>9</td>
<td>Missing tooth replaced by full denture</td>
</tr>
<tr>
<td><strong>Surface code</strong></td>
<td></td>
</tr>
<tr>
<td>0</td>
<td>Sound surface</td>
</tr>
<tr>
<td>1</td>
<td>Decayed surface – any cavitation of the enamel and/or its extension into dentine</td>
</tr>
<tr>
<td>2</td>
<td>Filled surface – one that has a restoration (including preventive resin restorations)</td>
</tr>
<tr>
<td>3</td>
<td>Filled and decayed surface – a restoration with secondary caries or a fissure sealant with visible caries around the margins</td>
</tr>
<tr>
<td>4</td>
<td>Crown or bridge abutment. For the purposes of analysis crowned deciduous teeth were judged equivalent to three filled surfaces; crowned permanent teeth were excluded from analysis</td>
</tr>
<tr>
<td>5</td>
<td>Missing surface – assumed to be due to caries unless tooth was a third molar or one of matching premolars judged to be extracted for orthodontic reasons</td>
</tr>
<tr>
<td>6</td>
<td>Missing surface due to other reasons (see above)</td>
</tr>
<tr>
<td>7</td>
<td>Unerupted surface – as determined by individual’s age</td>
</tr>
<tr>
<td>8</td>
<td>Excluded (unexposed) root surface</td>
</tr>
</tbody>
</table>

Participants who would require antibiotics for periodontal probing (those considered high-risk for endocarditis or those who had had a joint replacement in the previous two years) were excluded from this part of the clinical examination.

The site for the examinations was a dental surgery within the CCDHB. The principal investigator carried out all clinical examinations. Because some individuals with haemophilia may be at risk of bleeding upon periodontal probing, the bleeding risk of each participant was assessed prior to the clinical examination. An assessment was conducted by the Regional Haemophilia Nurse Specialist as to their bleeding risk and haemostatic cover was administered to participants for whom the nurse deemed it to be necessary.
All clinical data were recorded and entered post-examination into a statistical software package (SPSS version 19).

All participants received an oral health report that summarised the findings (and limitations) of the clinical examination and provided details of how to access appropriate dental care. Provision had been made for referral of patients discovered to have potentially serious oral conditions.

2.4.3 Semi-structured interviews

All participants were invited to participate in a semi-structured interview conducted by the principal researcher. The interviews were guided by a template formulated to include questions about perception of oral health and its importance, and perceived barriers to oral health (Appendix 9). However, this part of the study continued to be emergent, and provision had been made for new paths of inquiry to be identified and followed.

The semi-structured interviews were recorded, transcribed and checked for accuracy by comparing the transcript with the recording.

2.5 Analyses

2.5.1 Quantitative data

The data from the questionnaire and the clinical examinations were analysed using SPSS version 19. Descriptive statistics (percentages, means with standard deviations) were reported. Chi-square tests and Mann-Whitney tests were used, as appropriate, to test the statistical significance of observed associations.
2.5.2 *Qualitative data*

Inductive thematic analysis of the transcriptions of the semi-structured interviews was carried out. As each transcript became available, the researcher read it several times to become familiar with its content. Themes and categories were then identified from relevant text, and a preliminary coding frame was developed. Transcripts were coded and, as new transcripts became available (and as new codes were identified), changes were made to the coding frame to combine, separate or redefine themes and categories. All transcripts were reread and coded according to the new coding frame.

Responses were linked back to the quantitative data in order to identify discordance or agreement in the data, and to help in interpreting and explaining the data.
3 Results

This chapter presents the results of this study. Qualitative and quantitative results are presented together where they relate to the same subject matter. Where direct quotes are presented, the number in square brackets at the end of the quote represents the participant’s unique ID number. Italicised text in square brackets represents the researcher’s comments or interpretation.

3.1 Participants

The Regional Haemophilia Nurse Specialists (who support individuals with moderate or severe haemophilia residing in the Capital & Coast, Hutt Valley or Waiararapa DHB catchment areas) identified 38 eligible individuals. Clinical examinations and questionnaires were completed by 33 individuals, comprising 25 adults and 8 children, giving a participation rate of 86.8%. Semi-structured interviews were held with 32 of these participants (or their parents). The characteristics of participants and non-participants can be seen in Table 6. All participants were male. Information on the socio-economic status of non-participants was not available.

Twenty-five adults participated in the study. This gave an adult response rate of 83.3%. and the demographic characteristics of adult participants and non-participants can be compared with data presented in Table 6. There were more Maori and city-dwellers among the non-participants than the participants.

Eight children participated in the study. This was 100% of all eligible children; their demographic data can be seen in Table 6.
### Table 6 - Demographic characteristics of participants and non-participants (brackets contain column percentages unless otherwise indicated)

<table>
<thead>
<tr>
<th></th>
<th>Adults</th>
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<th></th>
<th>Children</th>
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<th>All</th>
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<tbody>
<tr>
<td></td>
<td>Participants</td>
<td>Non-participants</td>
<td>Total</td>
<td>Participants</td>
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<td>Participants</td>
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<td>Total</td>
<td>Non-participants</td>
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<td>Participants</td>
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<tr>
<td><strong>Severity of haemophilia</strong></td>
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<tr>
<td>Severe</td>
<td>20 (80.0%)</td>
<td>3 (60.0%)</td>
<td>23 (76.7%)</td>
<td>7 (88%)</td>
<td>27 (81.8%)</td>
<td>3 (60.0%)</td>
<td>30 (78.9%)</td>
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<tr>
<td>Moderate</td>
<td>5 (20.0%)</td>
<td>2 (40.0%)</td>
<td>7 (23.3%)</td>
<td>1 (13%)</td>
<td>6 (18.2%)</td>
<td>2 (40.0%)</td>
<td>8 (21.1%)</td>
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<td><strong>Ethnicity</strong></td>
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<tr>
<td>NZ European</td>
<td>16 (64.0%)</td>
<td>3 (60.0%)</td>
<td>19 (63.3%)</td>
<td>4 (50%)</td>
<td>20 (66.6%)</td>
<td>3 (60.0%)</td>
<td>23 (60.5%)</td>
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<tr>
<td>Maori</td>
<td>6 (14.0%)</td>
<td>2 (40.0%)</td>
<td>8 (26.7%)</td>
<td>1 (13%)</td>
<td>7 (21.2%)</td>
<td>2 (40.0%)</td>
<td>9 (23.7%)</td>
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<tr>
<td>PI</td>
<td>2 (8.0%)</td>
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<td>2 (6.7%)</td>
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<td>2 (6.1%)</td>
<td>0 (0.0%)</td>
<td>2 (5.3%)</td>
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<tr>
<td>Other</td>
<td>1 (4.0%)</td>
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<td>1 (3.3%)</td>
<td>3 (38%)</td>
<td>4 (12.1%)</td>
<td>0 (0.0%)</td>
<td>4 (10.5%)</td>
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<td><strong>Domicile</strong></td>
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<tr>
<td>City</td>
<td>17 (68.0%)</td>
<td>5 (100.0%)</td>
<td>22 (73.3%)</td>
<td>6 (75%)</td>
<td>23 (69.7%)</td>
<td>5 (100.0%)</td>
<td>28 (73.7%)</td>
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<td>Large town</td>
<td>3 (12.0%)</td>
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<td>3 (10.0%)</td>
<td>2 (25%)</td>
<td>5 (15.2%)</td>
<td>0 (0.0%)</td>
<td>5 (13.2%)</td>
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<tr>
<td>Small town</td>
<td>5 (20.0%)</td>
<td>0 (0.0%)</td>
<td>5 (16.7%)</td>
<td>0 (0.0%)</td>
<td>5 (15.2%)</td>
<td>0 (0.0%)</td>
<td>5 (13.2%)</td>
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<tr>
<td><strong>Age</strong></td>
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<tr>
<td>Mean (SD)</td>
<td>42.5 (16.2)</td>
<td>39.8 (9.2)</td>
<td>42.0 (15.2)</td>
<td>9 (4)</td>
<td>34.5 (20.2)</td>
<td>39.8 (9.2)</td>
<td>35.1 (19.1)</td>
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<td><strong>Distance living from hospital (kms)</strong></td>
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<tr>
<td>Mean (SD)</td>
<td>30.4 (18.7)</td>
<td>22.0 (13.0)</td>
<td>29.0 (18.0)</td>
<td>23 (19)</td>
<td>28.6 (18.7)</td>
<td>22.0 (13.0)</td>
<td>27.8 (18.0)</td>
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<td><strong>Highest education level reached</strong></td>
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</tr>
<tr>
<td>None</td>
<td>4 (16.0%)</td>
<td>0 (0.0%)</td>
<td>4 (16.0%)</td>
<td>4 (12.1%)</td>
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<tr>
<td>High school</td>
<td>7 (28.0%)</td>
<td>4 (50.0%)</td>
<td>11 (33.3%)</td>
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<tr>
<td>Diploma</td>
<td>10 (40.0%)</td>
<td>2 (25.0%)</td>
<td>12 (36.4%)</td>
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<tr>
<td>University</td>
<td>4 (16.0%)</td>
<td>2 (25.0%)</td>
<td>6 (18.2%)</td>
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<td></td>
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<td></td>
<td></td>
</tr>
</tbody>
</table>

1 Of participant or parent of participant  
2 Not available for non-participants
3.2  Clinical data for adults

3.2.1  Presence of teeth

Four individuals (16%) had no remaining teeth. A significantly higher proportion of those with no remaining teeth had not completed any formal education and lived further away from the haemophilia centre. Table 7 compares the characteristics of dentate and edentulous participants.

<table>
<thead>
<tr>
<th>Table 7 - Demographic characteristics of edentulous and dentate adult participants</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ethnicity</td>
</tr>
<tr>
<td>N (%)</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td></td>
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<tr>
<td></td>
</tr>
<tr>
<td>Age</td>
</tr>
<tr>
<td>Mean (SD)</td>
</tr>
<tr>
<td>Range</td>
</tr>
<tr>
<td>Highest education level reached</td>
</tr>
<tr>
<td>N (%)</td>
</tr>
<tr>
<td></td>
</tr>
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<td></td>
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<tr>
<td></td>
</tr>
<tr>
<td>Severity of haemophilia</td>
</tr>
<tr>
<td>N (%)</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>Distance living from hospital (kms)</td>
</tr>
<tr>
<td>Mean (SD)</td>
</tr>
<tr>
<td>Range</td>
</tr>
<tr>
<td>Domicile</td>
</tr>
<tr>
<td>N (%)</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td></td>
</tr>
</tbody>
</table>

aP<0.05
Twenty-one individuals (84.0%) had some teeth remaining, and the following data in this section refers only to these dentate individuals. Dentate individuals had a mean average of 26.9 (SD 3.1; median 27.0) teeth remaining. The distribution of the number of remaining teeth can be seen in Figure 1. Nine individuals (42.9%) had no missing teeth (due to caries) at all. All but one (95.0%) had a ‘functional dentition’ (considered to be 21 teeth or more (Steele et al., 1998)). Ten percent of dentate participants thought that they would require full dentures in the future.

![Figure 1 - Distribution of number of remaining teeth in dentate individuals](image)

3.2.2 Decayed, missing or filled surfaces

An average of 120 coronal surfaces per individual were recorded. The mean number of decayed, missing and filled coronal surfaces found in dentate individuals is presented by demographic characteristics in Table 8.

Seven individuals (33.3%) had untreated caries, with three having more than one decayed surface.
Table 8 - Summary data on caries experience at surface and tooth level

<table>
<thead>
<tr>
<th></th>
<th>N</th>
<th>No of decayed coronal surfacesa (DS)</th>
<th>No of filled coronal surfacesb (FS)</th>
<th>No of missing coronal surfacesc (MS)</th>
<th>Total no of decayed, filled or missing coronal surfaces (DMFS)</th>
<th>Number of decayed teeth (DT)</th>
<th>Number of filled teeth (FT)</th>
<th>Number of missing teeth (MT)</th>
<th>Total number of decayed, filled or missing teeth (DMFT)</th>
<th>Number of sound teeth</th>
</tr>
</thead>
<tbody>
<tr>
<td>All adults</td>
<td>21</td>
<td>0.9 (2.1)</td>
<td>14.0 (15.1)</td>
<td>10.2 (12.1)</td>
<td>25.1 (22.3)</td>
<td>0.7 (1.3)</td>
<td>5.8 (5.5)</td>
<td>2.1 (2.6)</td>
<td>8.6 (7.0)</td>
<td>18.2 (8.8)</td>
</tr>
<tr>
<td>Mean (SD)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Median</td>
<td></td>
<td>0.0</td>
<td>6.0</td>
<td>5.0</td>
<td>24</td>
<td>0.0</td>
<td>4.0</td>
<td>1.4</td>
<td>9.0</td>
<td>18.0</td>
</tr>
<tr>
<td>Range</td>
<td></td>
<td>0 - 9</td>
<td>0 - 42</td>
<td>0 - 42</td>
<td>0 - 70</td>
<td>0 - 5</td>
<td>0 - 17</td>
<td>0 - 9</td>
<td>0 - 23</td>
<td>2 - 31</td>
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<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>European</td>
<td>14</td>
<td>1.2 (2.5)</td>
<td>17.4 (16.9)</td>
<td>11.6 (12.9)</td>
<td>30.4 (24.3)</td>
<td>0.9 (1.5)</td>
<td>6.6 (6.0)</td>
<td>2.5 (2.7)</td>
<td>9.9 (7.6)</td>
<td>16.7 (9.6)</td>
</tr>
<tr>
<td>Non-European</td>
<td>7</td>
<td>0.3 (0.5)</td>
<td>7.3 (8.1)</td>
<td>7.0 (10.5)</td>
<td>14.6 (13.4)</td>
<td>0.3 (0.5)</td>
<td>4.3 (4.4)</td>
<td>1.4 (2.1)</td>
<td>6.0 (5.0)</td>
<td>21.3 (6.6)</td>
</tr>
<tr>
<td>Age</td>
<td></td>
<td></td>
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<td></td>
<td></td>
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<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>18 – 24</td>
<td>5</td>
<td>0.4 (0.5)</td>
<td>5.2 (5.3)</td>
<td>5.0 (8.7)</td>
<td>10.6 (12.1)</td>
<td>0.4 (0.5)</td>
<td>2.8 (3.0)</td>
<td>1.0 (1.7)</td>
<td>4.2 (4.4)</td>
<td>23.6 (7.1)</td>
</tr>
<tr>
<td>25 – 34</td>
<td>3</td>
<td>0.0</td>
<td>1.3 (1.5)</td>
<td>4.0 (6.9)</td>
<td>5.3 (6.8)</td>
<td>0.0</td>
<td>0.7 (0.6)</td>
<td>1.0 (1.7)</td>
<td>1.7 (2.1)</td>
<td>26.3 (5.0)</td>
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<tr>
<td>35 – 44</td>
<td>7</td>
<td>1.4 (3.6)</td>
<td>7.7 (7.4)</td>
<td>13.0 (16.2)</td>
<td>22.1 (17.3)</td>
<td>0.9 (1.9)</td>
<td>4.4 (3.0)</td>
<td>2.7 (3.5)</td>
<td>8.0 (5.0)</td>
<td>18.1 (7.6)</td>
</tr>
<tr>
<td>45 – 54</td>
<td>2</td>
<td>0.5 (0.7)</td>
<td>26.5 (7.8)</td>
<td>7.5 (10.6)</td>
<td>34.5 (2.1)</td>
<td>0.5 (0.7)</td>
<td>10.0 (4.2)</td>
<td>1.5 (2.1)</td>
<td>12.0 (2.8)</td>
<td>16.0 (2.8)</td>
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<tr>
<td>55+</td>
<td>4</td>
<td>1.5 (1.7)</td>
<td>39.3 (4.2)</td>
<td>18.0 (9.6)</td>
<td>58.8 (12.7)</td>
<td>1.3 (1.5)</td>
<td>13.8 (3.0)</td>
<td>3.8 (2.1)</td>
<td>18.8 (4.3)</td>
<td>6.8 (4.9)</td>
</tr>
<tr>
<td>Education</td>
<td></td>
<td></td>
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<td></td>
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<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Low educationd</td>
<td>7</td>
<td>0.7 (1.1)</td>
<td>20.4 (17.2)</td>
<td>14.1 (17.1)</td>
<td>35.3 (27.6)</td>
<td>0.6 (0.8)</td>
<td>8.6 (6.2)</td>
<td>3.0 (3.7)</td>
<td>12.1 (8.2)</td>
<td>14.4 (11.1)</td>
</tr>
<tr>
<td>Diploma</td>
<td>10</td>
<td>1.4 (2.8)</td>
<td>12.4 (14.4)</td>
<td>11.1 (8.9)</td>
<td>24.9 (18.7)</td>
<td>1.0 (1.7)</td>
<td>4.8 (4.3)</td>
<td>2.3 (1.8)</td>
<td>8.1 (5.5)</td>
<td>18.0 (6.8)</td>
</tr>
<tr>
<td>University</td>
<td>4</td>
<td>0.0</td>
<td>6.8 (11.6)</td>
<td>1.3 (2.5)</td>
<td>8.0 (10.9)</td>
<td>0.0</td>
<td>3.5 (6.4)</td>
<td>0.3 (0.5)</td>
<td>3.8 (6.2)</td>
<td>25.5 (5.4)</td>
</tr>
<tr>
<td>Severity of haemophilia</td>
<td>N</td>
<td>No of decayed coronal surfaces (^a) (DS)</td>
<td>No of filled coronal surfaces (^b) (FS)</td>
<td>No of missing coronal surfaces (^c) (MS)</td>
<td>Total no of decayed, filled or missing coronal surfaces (DMFS)</td>
<td>Number of decayed teeth (DT)</td>
<td>Number of filled teeth (FT)</td>
<td>Number of missing teeth (MT)</td>
<td>Total number of decayed, filled or missing teeth (DMFT)</td>
<td>Number of sound teeth</td>
</tr>
<tr>
<td>------------------------</td>
<td>----</td>
<td>-----------------------------------------</td>
<td>----------------------------------------</td>
<td>------------------------------------------</td>
<td>-------------------------------------------------</td>
<td>----------------------</td>
<td>----------------------</td>
<td>----------------------</td>
<td>-----------------------------------------------</td>
<td>---------------------</td>
</tr>
<tr>
<td>Severe</td>
<td>17</td>
<td>0.9 (2.2)</td>
<td>12.9 (13.9)</td>
<td>10.5 (13.5)</td>
<td>24.3 (22.7)</td>
<td>0.6 (1.3)</td>
<td>5.8 (5.5)</td>
<td>2.2 (2.8)</td>
<td>8.6 (7.2)</td>
<td>18.5 (9.3)</td>
</tr>
<tr>
<td>Moderate</td>
<td>4</td>
<td>0.8 (1.5)</td>
<td>18.8 (21.3)</td>
<td>9.3 (3.0)</td>
<td>28.8 (23.5)</td>
<td>0.8 (1.5)</td>
<td>6.0 (6.5)</td>
<td>2.0 (0.8)</td>
<td>8.8 (7.3)</td>
<td>17.3 (7.5)</td>
</tr>
<tr>
<td>Distance from hospital</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>≤20kms</td>
<td>12</td>
<td>0.5 (0.9)</td>
<td>10.0 (13.0)</td>
<td>6.7 (8.6)</td>
<td>17.2 (19.9)</td>
<td>0.4 (0.7)</td>
<td>4.4 (5.2)</td>
<td>1.4 (1.8)</td>
<td>6.3 (6.6)</td>
<td>21.4 (7.8)</td>
</tr>
<tr>
<td>&gt;20kms</td>
<td>9</td>
<td>1.4 (3.0)</td>
<td>19.3 (16.8)</td>
<td>15.0 (14.8)</td>
<td>35.8 (21.8)</td>
<td>1.0 (1.8)</td>
<td>7.7 (5.7)</td>
<td>3.1 (3.1)</td>
<td>11.8 (6.5)</td>
<td>14.0 (8.8)</td>
</tr>
</tbody>
</table>

\(^a\)Excluding carious retained roots; \(^b\)Excluding crowns; \(^c\)Due to caries; \(^d\)No qualifications or high-school certificate only
3.2.3 Decayed, missing or filled teeth

Summary data on decayed, missing and filled teeth are presented in Table 8. A breakdown of decayed, missing and filled teeth by demographic characteristics can be seen in Table 8, with summaries in Figure 2 and Figure 3. Participants who were: younger, had higher levels of education, non-European, and lived closer to the hospital tended to have less caries experience.

Two people (9.5%) had no caries experience (a DMFT of 0). These individuals were aged 22 and 30 years, respectively.

![Figure 2 - Mean DMFT by ethnicity, highest education achieved and distance residing from hospital](image-url)
3.2.4 Root surface caries

Summary data on the numbers of exposed roots, sound, filled and carious root surfaces are presented in Table 9.

Table 9 - Numbers of carious, filled and sound root surfaces

<table>
<thead>
<tr>
<th></th>
<th>Number of carious root surfaces</th>
<th>Number of filled root surfaces</th>
<th>Number of roots with caries experience</th>
<th>Number of sound root surfaces</th>
<th>Total number of exposed root surfaces</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean (SD)</td>
<td>0.05 (0.2)</td>
<td>1.0 (2.4)</td>
<td>1.1 (2.4)</td>
<td>11.0 (10.8)</td>
<td>12.1 (12.4)</td>
</tr>
<tr>
<td>Median</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>7.0</td>
<td>7.0</td>
</tr>
<tr>
<td>Range</td>
<td>0 - 1</td>
<td>0 - 10</td>
<td>0 - 10</td>
<td>0 - 36</td>
<td>0 - 42</td>
</tr>
</tbody>
</table>

The mean Root Caries Index was 4.0% (SD 7.9). Of the five individuals (23.8%) who had had some experience of root caries, one individual had untreated root caries.

3.2.5 Periodontal disease

All twenty-one adult dentate individuals had a periodontal examination. An average of 76 (SD 8.1; range 54 to 84) sites per dentate individual were recorded.
Table 10 summarises the prevalence and extent of periodontal disease in the sample. The prevalence of periodontal disease is defined by the presence of one or more sites with clinical attachment loss (CAL) or pocket depth (PD) measuring more than a certain threshold. The extent of periodontal disease is defined by the proportion of all sites that measured more than that threshold. For both prevalence and extent, the threshold has been set at different values, in order to test robustness and enable comparison with other studies.

Table 10 - Prevalence and extent of periodontal disease

<table>
<thead>
<tr>
<th></th>
<th>CAL 4+mm</th>
<th>PD 4+mm</th>
<th>CAL 5+mm</th>
<th>PD 5+mm</th>
<th>CAL 6+mm</th>
<th>PD 6+mm</th>
</tr>
</thead>
<tbody>
<tr>
<td>Prevalence N (%)</td>
<td>15 (71.4)</td>
<td>12 (57.1)</td>
<td>6 (28.6)</td>
<td>4 (19.0)</td>
<td>1 (4.8)</td>
<td>0</td>
</tr>
<tr>
<td>Extent % (SD)</td>
<td>4.7 (8.4)</td>
<td>1.3 (1.4)</td>
<td>2.0 (5.2)</td>
<td>0.3 (0.5)</td>
<td>0.4 (1.7)</td>
<td>0</td>
</tr>
</tbody>
</table>

3.2.6 Gingival inflammation

The presence or absence of bleeding on probing was recorded for each periodontally examined site. The mean bleeding index (the percentage of sites with bleeding on probing) was 25.9% (SD 19.7). One individual had no bleeding sites at all and another had more than three-quarters of his sites bleeding.

3.3 Clinical data for children

The data in this section pertain to deciduous teeth, permanent teeth and all teeth. The assumption was made that, in children, teeth were crowned due to caries, and an allowance of three filled surfaces was given for each crown.

3.3.1 Deciduous teeth

This section includes only data from deciduous teeth in the five children aged 2–11 years. They had a mean number of 15.6 (SD 3.2) deciduous teeth, with 14.4 being
sound. Summary data on the numbers of decayed, missing and filled surfaces can be seen in Table 11.

No children had any unrestored caries in their deciduous teeth, and 80% of children were caries-free in their deciduous teeth. Table 11 presents summary data on the numbers of decayed, missing or filled deciduous teeth.

3.3.2 Permanent teeth

Seven children aged 5–17 were included in this analysis. They had an average of 16.4 permanent teeth, with 14.3 of those being sound. Table 11 presents summary data on the numbers of decayed, missing and filled surfaces in permanent teeth.

Only two individuals had any caries experience, meaning that 71.4 % were caries-free. One individual (14.3%) had untreated caries in a permanent tooth and a DMFT score of 14. Table 11 presents summary data on the numbers of decayed, missing and filled permanent teeth.
### Table 11 - Summary data on numbers of decayed, missing and filled surfaces and teeth

<table>
<thead>
<tr>
<th></th>
<th>Number of decayed surfaces</th>
<th>Number of missing surfaces</th>
<th>Number of filled surfaces</th>
<th>Total number of decayed, missing or filled surfaces</th>
<th>Number of decayed teeth</th>
<th>Number of missing teeth</th>
<th>Number of filled teeth</th>
<th>Total number of decayed, missing or filled teeth</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Deciduous teeth</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mean (SD)</td>
<td>0.0 (0.0)</td>
<td>1.0 (2.2)</td>
<td>2.8 (6.3)</td>
<td>3.8 (8.5)</td>
<td>0.0 (0.0)</td>
<td>0.2 (0.5)</td>
<td>1.0 (2.2)</td>
<td>1.2 (2.7)</td>
</tr>
<tr>
<td>Median</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
</tr>
<tr>
<td>Range</td>
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<td>0 - 5</td>
<td>0 - 14</td>
<td>0 - 7</td>
<td>0</td>
<td>0 - 1</td>
<td>0 - 5</td>
<td>0 - 6</td>
</tr>
<tr>
<td><strong>Permanent teeth</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mean (SD)</td>
<td>0.3 (0.6)</td>
<td>4.7 (8.1)</td>
<td>5.0 (7.8)</td>
<td>10.0 (16.5)</td>
<td>0.1 (0.4)</td>
<td>0.4 (1.1)</td>
<td>1.6 (3.7)</td>
<td>2.1 (5.2)</td>
</tr>
<tr>
<td>Median</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
</tr>
<tr>
<td>Range</td>
<td>0 - 1</td>
<td>0 - 14</td>
<td>0 - 14</td>
<td>0 - 29</td>
<td>0 - 1</td>
<td>0 - 3</td>
<td>0 - 10</td>
<td>0 - 14</td>
</tr>
<tr>
<td><strong>All teeth</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mean (SD)</td>
<td>0.1 (0.4)</td>
<td>2.4 (5.0)</td>
<td>3.6 (6.4)</td>
<td>6.1 (11.4)</td>
<td>0.1 (0.4)</td>
<td>0.5 (1.0)</td>
<td>2.0 (3.7)</td>
<td>2.6 (5.0)</td>
</tr>
<tr>
<td>Median</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
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</tr>
<tr>
<td>Range</td>
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<td>0 - 14</td>
<td>0 - 14</td>
<td>0 - 29</td>
<td>0 - 1</td>
<td>0 - 3</td>
<td>0 - 10</td>
<td>0 - 14</td>
</tr>
</tbody>
</table>
3.3.3 Both dentitions combined

This data involves all teeth in all children aged 2–17. These children had an average of 24.1 teeth each, with 21.5 being sound. Table 11 presents summary data on the numbers of decayed, missing and filled surfaces in both dentitions combined.

One individual had untreated caries and five (62.5%) of the child participants were caries-free. Table 11 presents summary data on the numbers of all decayed, missing and filled teeth. The distribution of caries experience can be seen in Figure 4.

![Figure 4 - Distribution of caries experience in either dentition](image_url)

3.4 Impact of oral health

3.4.1 Oral Health Impact Profile (OHIP)

All adult participants completed the OHIP questionnaire. Table 12 shows the OHIP scores over all the subscales. The mean overall score was 5.2 (SD 6.6). Pain had the highest score of any subscale with physical disability and handicap having the lowest.
Table 12 - OHIP scores for all subscales

<table>
<thead>
<tr>
<th>Subscales</th>
<th>OHIP (Mean (SD))</th>
<th>Functional limitation</th>
<th>Pain</th>
<th>Psychological discomfort</th>
<th>Physical disability</th>
<th>Psychologic al disability</th>
<th>Social disability</th>
<th>Handicap</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean (SD)</td>
<td>5.2 (6.6)</td>
<td>1.0 (2.3)</td>
<td>1.5 (1.8)</td>
<td>1.1 (2.0)</td>
<td>0.2 (0.5)</td>
<td>0.7 (1.2)</td>
<td>0.4 (0.8)</td>
<td>0.2 (0.7)</td>
</tr>
<tr>
<td>Median</td>
<td>3.0</td>
<td>0.0</td>
<td>1.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
<td>0.0</td>
</tr>
<tr>
<td>Range</td>
<td>0 - 26</td>
<td>0 - 8</td>
<td>0 - 7</td>
<td>0 - 7</td>
<td>0 - 2</td>
<td>0 - 4</td>
<td>0 - 2</td>
<td>0 - 3</td>
</tr>
</tbody>
</table>

Table 13 shows the prevalence of impacts of oral health in each of the OHIP subscales. The majority of participants (72%) reported no impact of their oral health. No participants reported any impact in the physical disability or social disability subscales.

Table 13 - Prevalence of one or more impact over all OHIP domains

<table>
<thead>
<tr>
<th>OHIP subscale</th>
<th>No impacts N (%)</th>
<th>1+ impacts N (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Functional limitation</td>
<td>18 (72.0)</td>
<td>7 (28.0)</td>
</tr>
<tr>
<td>Pain</td>
<td>20 (80.0)</td>
<td>5 (20.0)</td>
</tr>
<tr>
<td>Psychological discomfort</td>
<td>22 (88.0)</td>
<td>3 (12.0)</td>
</tr>
<tr>
<td>Physical disability</td>
<td>25 (100.0)</td>
<td>0</td>
</tr>
<tr>
<td>Psychological disability</td>
<td>24 (96.0)</td>
<td>1 (4.0)</td>
</tr>
<tr>
<td>Social disability</td>
<td>25 (100.0)</td>
<td>0</td>
</tr>
<tr>
<td>Handicap</td>
<td>24 (96.0)</td>
<td>1 (4.0)</td>
</tr>
</tbody>
</table>

3.4.2 Time-off taken for dental appointments

Four participants (19% of adult dentate participants) needed to take time off work or school to attend dental appointments in the last year. This time ranged from 7.5 hours to four days. Three participants (21.4% of those who have ever had to ask) have had difficulty getting time off work in the past to attend dental appointments.

Two children (25%) needed to take time from school to attend dental appointments during the last year. This time ranged from 30 to 60 minutes.
3.4.3  Impact of oral health

Participants expressed the impact of oral health in terms of either functional problems (usually as a theoretical issue), or social problems (usually as a direct experience).

You know, it’s how you eat. Be a sad place if you could only have milkshakes all the time. [26]

So mouth and teeth, obviously, they are very useful for you, food, like eating your food and all that. So if that part is not working properly, or if you are in pain, then, you know, you can’t really enjoy your food and can’t really eat that well, and all that, so, I think yes, it is quite important to look after it. [22]

It’s like having a racehorse with no feet. Haven’t got good feet, it’s not going to race. You’re not going to feel very healthy, are you, if your teeth are aching and you bite on something and it hurts. [37]

The social importance of a healthy mouth was also highlighted in the direct experiences of some participants.

It, it, it, [bad breath from unhealthy gums] broke down my social life because I didn’t want to go out. I’d stay home a lot, too, you know, and my wife, you know, you know, kissing and all that. It was just, I tell you, it wasn’t bloody nice and it can, it can bloody get you. [24]

When I was having problems with my teeth before I had them all out, I had, bad breath and it was to the point of, you know, to talk to
someone, you know, I couldn’t. It was awful to talk to someone, you know, I’ve got bad breath and I was conscious of all that. [27]

Many participants described memories of waking up with ‘blood on the pillow’ that had come from their mouths during the night.

I was waking up every morning with blood all over the place, you know. I used to sleep with a towel around me. And on the pillow you know, you’d clot, it was just – or I’d sleep with my mouth and in the morning, so clots, oh, it was just – I’m not for saying nasty words, you know, it was a total crap, it was. [24]

Like, I used to bleed every morning. I used to wake up with blood on the pillow, even if it was a small amount. But, like, when I was a teenager, I, I ruined every single pillow in the house because I’d wake up and it’s like it was covered in blood. [19]

I remember like, pillows just soaked with, you know, big clots, you know. [27]

3.5 Dental visiting characteristics

3.5.1 Adult attendance at dental appointments

Adult participants were asked whether they had attended a dental appointment in the previous year or not, and whether they usually attended the dentist for check-ups or only when they had a dental problem. These data are summarised in Table 14. Those who had attended in the previous year and who reported that they usually attended for check-ups were classified as regular attenders. While over half the participants (52.0%) had attended in the previous year, nearly half of these had attended only when they had dental problems.
### Table 14 - Patterns of dental attendance (adults)

<table>
<thead>
<tr>
<th></th>
<th>Usually attends for check-ups</th>
<th>Usually attends with problems</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Attended in previous year N (%)</td>
<td>7 (28%)</td>
<td>6 (24%)</td>
<td>13 (52%)</td>
</tr>
<tr>
<td>Did not attend in previous year N (%)</td>
<td>4 (16%)</td>
<td>8 (32%)</td>
<td>12 (48%)</td>
</tr>
<tr>
<td>Total N (%)</td>
<td>11 (44%)</td>
<td>14 (56%)</td>
<td>25 (100%)</td>
</tr>
</tbody>
</table>

#### 3.5.2 Child attendance at dental appointments

One child participant (aged three) had never visited the dentist. Of the others, 85.7% had been to see a dentist within the previous year. All participants (who could remember) had attended a dental professional by age five (median age of three).

#### 3.5.3 Place of dental visit

Each participant was asked the location of their most recent dental visit and where (other things being equal) they would prefer to make their dental visits (Figure 5). The majority of participants (80% of adults, 71% of children) had made their most recent dental visit at a hospital department though only 68% of adults and 43% of children would prefer their appointments to be at the hospital. Five participants made their most recent visit to a private practitioner. However, these were not the same five who would prefer to make their appointment at a private practitioner.
3.5.4 Choosing a dental provider

Figure 6 presents the reasons given, in the questionnaire, by adults for their choice of practitioner at their most recent visit to a dentist.

Figure 5 - Location of most recent dental visit and desired location for dental visits

When participants talked about why they attended the hospital for their dental care, they gave three main reasons: streamlining, safety and cost.
Attendance at the hospital was seen as being easy or streamlined.

They’ve got people like *the haemophilia nurse* here, you know, makes it so much easier. He arranges things for you here and gets things sorted out, you know. [14]

Benefits *of going to the hospital*. It’s more streamlined if you do need surgery. It takes one step out. [31]

The good thing with haemophilia now is like what *the haemophilia nurse* does when you come and see *the hospital dentist* when she’s going to do something like burn back some gum or cut off some gum to get to a thing to put a cap on something is that they, they take away your haemophilia, so they give you enough clotting factor so you no longer have haemophilia. So you rock up to the chair now just like everyone else and that’s great, that makes a huge difference, the fact that you just turn up. *[The hospital dentist]* knows you’re coming, she can do what she likes, not going to cause a drama. [21]

Some participants expressed a desire to be seen at the hospital because it was perceived to be safer than a private practice.

It would freak me out *to go to a private dentist*. To think I wasn’t near the people I needed to get access to were if something went wrong. [20]

If I bring *my son* to hospital I know 100% they look after him 'cos they know how they deal with him as a haemophiliac, you know, so they can deal with two problems same time. [28]
To be honest I wouldn’t feel as confident that I was going to be getting good care. The, the times that I have gone out and seen someone with Dr in their title who was outside the hospital system, it’s never ended particularly well. [13]

Treatment at the hospital was acknowledged to be cheaper than private practice.

It’s cheaper here [8]

Benefits [of going to the hospital]. Historically it was cheap. Yeah, but still is. [31]

Not all participants liked going to the hospital for their dental treatment. The reasons given for not attending the hospital were either practical (waiting times, not necessary to be seen in hospital) or more philosophical.

The hospital was perceived as having long waiting lists.

Sometimes, having to go to the dentist at the hospital, meant bigger delays than if you went private. [15]

Well, if you’re haemophiliac and you need an operation, forget it. Because it could take years. [29]

Some participants felt that a private dentist was sufficient and that there was no need for the additional care at the hospital.
If, I know if I have like a treatment beforehand I’m going to be in pretty good shape [if I see a private dentist]. [26]

No, I don’t think so. As soon as I spoke to my dentist about it he was like “it’s fine.” He said “as long as”, you know, he said “that’s not a problem”, he said, because the understanding of the school dental nurse was that if he needed anything done with his teeth, like a tooth pulled or fillings, it’s going to the hospital. And they just reckon that they would let me know and I could get in touch with.... the haemophilia nurse. It was all organised. So as soon as I told my dentist that he was like “no, that’s not a problem.” [4]

A more philosophical view taken by some participants was that, as individuals with haemophilia, they already received a lot from the health system and did not need to take any more.

Well, the hospital is kinda looking after me in other ways. I don’t want to put too much pressure on them. I don’t mind paying my way. Don’t have any trouble with that although as you get older you seem to rely on the government or the system to take care of you. [37]

But personally there’s probably no reason why I should have it any cheaper than anyone else. For check-ups and things. I think surgery it makes sense, for haemophilia, but for check-ups, I don’t see a point. [31]

I’m sure there’s lots of people out there worse than we’ve got, we just bleed. [35]
3.6 Barriers to dental care

The reasons for not attending in the previous year are summarised in Figure 7 for the 12 adults who had not done so. All adult participants were asked whether they felt that they attended a dental professional often enough and, if not, why not. Ten participants (40.0%) felt that they did not attend the dentist often enough and the reasons are summarised in Figure 7.

![Figure 7 - Reasons given by adult respondents for not visiting dental professional in previous year or more generally](image)

*Figure 7 - Reasons given by adult respondents for not visiting dental professional in previous year or more generally*  
*(total may be more than 100% because more than one response could be given)*

One in five adult participants (20.0%) reported that they had experienced difficulty accessing dental care due to their haemophilia in the past. All of these felt that the dentist would not see them in case they bled.

When asked in more detail why they did not visit a dental professional, participants gave many reasons. These could be broken into three main groups: extrinsic factors (time and cost); intrinsic values or perceptions (fear, additional medical interventions, need not perceived, and laziness); and reasons related to their haemophilia (joint bleeds and unwilling dentist).
Participants talked about the time that dental appointments took, particularly when time off work was needed.

That’s what put me off before having it done, was the time off work, you know, and I hate having time off work. [14]

I mean, I don’t come here [hospital] for regular check-ups because I know simply if I say “hey, I’d like to come in at ten” it won’t be until eleven o’clock that they see me. And I’ve organised to take time off work, you know, parking and all the rest of that sort of thing, you know, then. [26]

And life’s quite busy, so it is that that makes it difficult to get to appointments and fit things in really well. [11]

Some participants mentioned their fear of dental treatment, as opposed to dental visits being unpleasant or disliked.

I’m very relaxed but when I’m a bit nervous in the waiting room and I sit in the chair and I just always break out in a sweat, just straight away. [20]

It was then I had to have a filling, you know, oh man, I was sweating just thinking about it. Just thinking about it, you know. Lot of people [say] “oh, toughen up”, you know. Not when it comes to the dentist, mate – not when it comes to the dentist. I hated, just the thought, you know, got into bed at night thinking I’ve got to go to the dentist at ten o’clock tomorrow morning. Can’t sleep, you know. [29]
'Cos I was shit-scared of dentists. [39]

The cost of dental treatment was also mentioned as a reason for not attending the dentist.

You know it’s not just going to be one visit, you know, there’s going to be $400 that time and $600 that time and then a $200 check-up afterwards. [26]

I didn’t want to spend lots of money on them - my teeth. [18]

Visiting the dentist was seen as yet another medical intervention by some participants and, as such, to be avoided.

Because I come to the hospital so often for other things that I try and avoid it. [20]

Obviously I have to come and pick up my Factor 8 about once a month, and that’s about as frequent as I would like to come to the hospital. So it’s not dentist in particular. [39]

I’m also not a great one for going and taking care of medical things at hospitals. Ah, erm, I prefer to stay out of them. [13]

Some participants did not attend the dentist because they did not perceive the need to go.
[My teeth] weren’t giving me any trouble. [so I didn’t go to dentist] [27]

It’s largely I think that my mouth is, is healthy, that I don’t experience pain, I don’t experience major amounts of bleeding. Um, I, when I brush my teeth I give them a big poke and none of them seem to wiggle or move around in my mouth. [13]

I never really had problems other than wisdom teeth which is a one-off thing that everyone goes through. But otherwise, I think I’m in reasonably good shape so I come as little as possible. [39]

Some participants recognised that lack of attendance came from their own inactivity.

Because I’m slack probably. [20]

Couldn’t be bothered, I suppose. I just never went. [27]

Some participants experienced joint bleeds that made it difficult to attend appointments. These were either single bleeds causing short-term problems, or the long-term disability from repeated bleeds.

Haemophilia can affect it because sometimes I haven’t been able to get down here because I’ll bleed. Um, so that can be a bit annoying. Especially like if I woke up in the morning and I think oh, shit, I’ve got a bleed, I can’t come down here. I’ve got to ring up and cancel and then like explain why, explain why and then rah, rah, rah, and then there’s the hassle of like getting that appointment cancelled and catching up on the other because with haemophilia you just don’t
know if you’re going to wake up with a bleed or whatever, ‘cos like last night I had some Factor 8 just in case so I didn’t wake up with a bleed. [17]

Nothing is easy for me because, as you see, my mobility’s fairly limited and it takes a bit of planning and thing to do, so it’s, it’s not, not easy but it’s not difficult either. It’s just not as easy. When I drove it wasn’t so bad ‘cos I just drove in and, er, but now I don’t drive so, er, so I’ve got to find some other way of getting there. [12]

Some participants had experience of dentists being unwilling to treat someone with haemophilia.

Wasn’t so much I that I didn’t go to them, it was they didn’t want to see, didn’t want to know me. [12]

Oh, they [private dentists] are just scared of haemophiliacs, you know. Most of the dentists I’ve tried to get to give me, you know, a clean or a service or whatever, they just – they think you’re going to die on their chair, you know, the way they act, a lot of them. [36]

Basically the [dentist] wouldn’t do anything at all. [10]

3.6.1 Overcoming barriers

Many participants talked about why they made the effort to attend dental appointments. Generally, they wanted to avoid the consequences (dental, haematological or other) of not going, or they had a philosophical approach to the benefits to society.
Some participants recognised that their teeth or mouth would suffer without the regular input of a dental professional.

I think now I would come for more check-ups, now that I realise the importance. So now that I’ve actually had problems. [39]

Well, I'm paranoid about losing my teeth. I've got a family of grandmothers and mothers who have got false teeth and it's not something I would ever want, so I've been, you know, usually have a hygienist appointment every six months and a dental check-up every 12 months because – my gums I think, because I carry the haemophilia gene, tend to be quite sensitive and they do bleed. [1]

For some participants, the additional complications of providing dental treatment to someone with haemophilia was the motivation for attending.

I do spend the money on taking the kids to a private dentist as well as the school check-ups, because I do worry, especially about [my son]. Because I've seen the consequences of haemophiliacs not looking after their teeth, and I don't want him to be in that situation. [5]

Just – especially when your gums bleed and if it does get to a point where you do have to have teeth removed, it can be quite an ordeal... so you’d be better of to maintain a decent set of teeth from the word go and you’ll be okay. [18]

I don’t leave [dental care] too long. If I didn’t have haemophilia I might leave it a bit longer. But because I’ve got it I think well, it’s just going to make things worse if it goes too far, so I get it done earlier rather than later. [37]
Some had other reasons for attending.

I do try and diligently come or my wife gives me a quick, swift kick up the butt. [20]

While not necessarily a factor in their own attendance, some participants had a wider awareness of the benefits to society if those with haemophilia looked after their teeth.

We don’t have to go through some big drama having the teeth, having a haemophiliac’s teeth out, it’s going to be enormously expensive through blood products for a kick-off. [21]

Catch it early. That’s my opinion. If we look after ourselves then we, our requirement of the health services overall would lessen, I would think. [35]

And from a, from a kind of wider population point of view it does have an impact on everybody from sort of the pragmatic thing and for haemophilia is so expensive to treat that by making the case that, you know, by treating this population really well you’re actually making huge savings over here for other, you know, a whole bunch of people could have elective surgery if you cut this, you know. [11]

Some participants observed that having haemophilia actually meant their oral health was better.

And in fact [my son] does end up having the double dentist treatment sometimes because we’ll take him to the dentist as well. We’ll go as a family every six months. So you get to go twice! [to son] [40]
In fact I’ve probably, probably over the years gone to the dentist more than a lot of my friends of my age would. [11]

Well, I’ve been to the dentist a lot. I had two dentists up until I was 18, ‘cos I would go to the hospital and the school and then after 14 the local dentist, privately. [31]

3.7 Time commitment for dental treatment

Figure 8 shows the time taken by adult respondents to get to their last dental appointment. One respondent (who travelled 30-60min) felt that this was too long to travel.

![Time taken by adult respondents to get to dental appointment](image)

**Figure 8 - Time taken by adult respondents to get to dental appointment**

3.8 Dental anxiety

The dental anxiety scale (DAS) was administered to all adult participants. Six individuals (24.0%) were considered dentally anxious (described by Corah (1979) as
those with a DAS score higher than 13). Table 15 summarises the DAS scores for each group.

Table 15 - DAS scores for anxious and non-anxious participants

<table>
<thead>
<tr>
<th></th>
<th>Dentally anxious</th>
<th>Non-dentally anxious</th>
<th>All adult participants</th>
</tr>
</thead>
<tbody>
<tr>
<td>N (%)</td>
<td>6 (24.0)</td>
<td>19 (76.0)</td>
<td>25 (100.0)</td>
</tr>
<tr>
<td>Mean (SD)</td>
<td>16.3 (2.1)</td>
<td>6.8 (2.2)</td>
<td>9.1 (4.7)</td>
</tr>
<tr>
<td>Median</td>
<td>16.0</td>
<td>6.0</td>
<td>7.0</td>
</tr>
<tr>
<td>Range</td>
<td>14 - 20</td>
<td>4 - 12</td>
<td>4 - 20</td>
</tr>
</tbody>
</table>

Table 16 shows the DAS scores for dentate and edentulous participants. Those who were edentulous were significantly more likely to be classified as dentally anxious.

Table 16 - DAS scores for dentate and edentulous participants

<table>
<thead>
<tr>
<th></th>
<th>Dentate (N=21)</th>
<th>Edentulous (N =4)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number dentally anxious (%)</td>
<td>3 (14.2)</td>
<td>3 (75.0)*</td>
</tr>
<tr>
<td>Mean DAS (SD)</td>
<td>7.9 (3.7)</td>
<td>15.3 (4.6)</td>
</tr>
<tr>
<td>Median</td>
<td>6.0</td>
<td>16.0</td>
</tr>
<tr>
<td>Range</td>
<td>4-17</td>
<td>9-20</td>
</tr>
</tbody>
</table>

*P<0.05

3.9 The cost of dentistry

Table 17 shows how adult participants perceive cost affecting their dental visits. One in four reported that the cost of dentistry was a burden to them. One in six had avoided a dental visit due to the cost and one participant had refused treatment due to the cost. Fewer than half would visit more often if the cost was lower.
### Table 17 - Cost as a barrier to dentistry

<table>
<thead>
<tr>
<th></th>
<th>Yes</th>
<th>No</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>N (%)</td>
<td>N (%)</td>
</tr>
<tr>
<td>Agree cost of dentistry is a burden</td>
<td>6 (24)</td>
<td>19 (76)</td>
</tr>
<tr>
<td>Has avoided a dental visit in last year due to cost</td>
<td>4 (16)</td>
<td>21 (84)</td>
</tr>
<tr>
<td>Has refused treatment due to the cost</td>
<td>1 (4)</td>
<td>24 (96)</td>
</tr>
<tr>
<td>Would visit dentist more if cost was lower$^a$</td>
<td>11 (44)</td>
<td>13 (56)</td>
</tr>
</tbody>
</table>

$^a$One missing response

Thirteen participants spent money on dentistry in the last year. These individuals spent an average of $302.31 (range $100-850).

Adult participants were asked whether there were any groups that should receive subsidised dental health-care. Five respondents thought that no-one should receive subsidised dental care. Other responses are shown in Figure 9.

![Figure 9 - Groups which respondents felt should receive subsidised dental care](image)

**Figure 9 - Groups which respondents felt should receive subsidised dental care**

(Can total more than 100% because respondents could give more than one answer)
When participants were discussing the cost of dentistry, it was either described as a negative influence or as something that was not an issue. The negative effects of the cost were expressed either as a general perception, or a description of how the cost of dentistry had impacted on someone they knew, or how the cost of dentistry had impacted on themselves.

I think for people in the lower [socio-economic group] I would imagine, um, it [cost of dentistry] would be a burden. [35]

Oh, in general it’s expensive, I reckon. It’s always been expensive, hasn’t it? [36]

Lack of money would be one thing [stopping people going to the dentist]. [37]

Some participants could describe how the cost of dentistry had impacted on someone they knew.

But um, my flatmate, he’s got to have um, a tooth done and it’s going to cost him about five, six hundred dollars. And that’s, that’s just for a filling. So, and, um. And that’s, that’s quite, quite expensive, I think. [17]

Also it’s the things like the cost of getting a crown. It tends to have a financial – a friend of mine had some crown work done, quite significant crown work and it was $12,000.00 Australian dollars. So it was, I think everyone who was near went “Woah - that’s a lot of money”. [40]
Two participants had direct experience of how the cost had affected them.

Yeah, cost is, I mean, even through a subsidised rate it still took a fair chunk out of my pay cheque each week, so, you know, it’s definitely a big factor in going. [25]

I mean crikey, the last time I went it was about $800 and that was like, you know, a couple of fillings and a bit of a clean-up, and you sort of think well, you know, I can’t really do that two or three times a year. You know, its..... I’d rather, I don’t know, take the kids away on a holiday or whatever, you know. That’s more important to me than sort of, I don’t know, fixing a non-drastic problem with my mouth. [26]

However, for the majority of participants, being able to access dental care through the public system meant that cost, although accepted as an issue for other people, was not an issue for them.

It’s a lot cheaper going through here [hospital] than through a private practice [25]

Well, [dentistry at the hospital] cost a lot less. [10]

Well, [cost is not an issue] for me. Normal dentistry, because I’ve always had to go through the hospital because of the bleeding. [27]

I think [my son] would be able to see the dentist, here [at the hospital] maybe annually just for check-ups which is great, so in a way haemophilia has an advantage in getting that care [at a reduced cost]. [2]
3.10 Self-perceived oral health

3.10.1 Self-perceived oral health of adults

Nine individuals (36.0%) thought that they currently needed dental treatment.

All adults were asked to describe their oral health on a Likert scale. These responses are summarised in Figure 10.

Figure 11 summarises how satisfied adult respondents were with the health and appearance of their mouth. In general, participants were more satisfied with the health of their mouth than with the appearance.

When asked in general terms about pain or discomfort in the last four weeks, nearly a quarter (24.0%) reported that they had experienced pain or discomfort (Figure 12). When asked about specific oral symptoms (Table 18) three out of five respondents had experienced at least one symptom during the last four weeks.

Table 18 - Oro-facial symptoms experienced in last four weeks

<table>
<thead>
<tr>
<th>Symptom experienced</th>
<th>N (%)</th>
<th>Symptom not experienced</th>
<th>N (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Toothache</td>
<td>2 (8)</td>
<td>23 (92)</td>
<td></td>
</tr>
<tr>
<td>Sensitivity</td>
<td>5 (20)</td>
<td>20 (80)</td>
<td></td>
</tr>
<tr>
<td>TMJ discomfort</td>
<td>0</td>
<td>25 (100)</td>
<td></td>
</tr>
<tr>
<td>Gingival bleeding</td>
<td>6 (24)</td>
<td>19 (76)</td>
<td></td>
</tr>
<tr>
<td>Bad breatha</td>
<td>6 (29)</td>
<td>15 (71)</td>
<td></td>
</tr>
<tr>
<td>Any symptom</td>
<td>15 (60)</td>
<td>10 (40)</td>
<td></td>
</tr>
</tbody>
</table>

*aFour missing responses

The majority of adult participants (90.5%) think that they will retain some of their natural teeth throughout their lifetime.
Figure 10 - Self-reported oral health (adults) (one missing answer)

Figure 11 - Satisfaction with oral health or appearance (adults)

Figure 12 - Self-reported pain/discomfort in last four weeks (adults)

Figure 13 - Self reported dryness (one missing response)
One-third of participants (33.3%) reported feeling dry mouth frequently or always (Figure 13).

3.10.2 Self-perceived oral health of children

The parent of one individual (14.3%) thought that their child currently needed dental treatment. Figure 14 summarises the self-reported oral health; Figure 15 summarises self-assessment of appearance of the mouth.

No children had avoided eating in the last twelve weeks but two children (25.0%) had experienced pain in that period (see Figure 16).

![Chart showing self-reported oral health of children]

**Figure 14 - Self-reported oral health (children)**
3.11 Understanding of oral health

All participants were asked during the semi-structured interview how they rated their own (or their child’s) oral health and why they felt that way. Participants justified their self-assessment of their oral health in many different ways: the appearance of their teeth; the presence or absence of symptoms; the ability to eat; information given by an oral health professional; in terms of treatment need, either current or historic; and, the quality or quantity of home-care.
Those who based the judgement of oral health on the appearance of teeth were interested in either the colour or straightness of the teeth.

Usually how much whiter they are *[makes me think my teeth are average compared to other people my age]*. [18]

*[My oral health is poor because]* I don’t like the way my bottom jaw sits between my top jaw, so it affects like I’ve got an under bite. [8]

I think they’re fairly straight considering I never had braces or anything like that *[so I would give my mouth 6 out of 10]*. [25]

The presence or absence of symptoms (such as bleeding, halitosis or discomfort) was one way of assessing their oral health for some participants.

I think *[the health of my own mouth is]* pretty good, but because I notice sometimes bleeding gums when I floss my teeth, that’s why I sort of think you know they need to be examined and cleaned. [11]

My breath doesn’t stink *[so my oral health is ok at the moment]*. [17]

*[My oral health is good]* and I don’t have any discomfort. [2]

The ability to eat (or not) was felt to be an important indicator for some.

I don’t have any trouble eating on them *[so my oral health is ok at the moment]*. [17]
Also, I do need to be a bit careful what I chew on, you know, really, because the teeth that are broken are a multiple number of times have just snapped off, you know, eating. Banana chip in the museli or something dumb like that just sheared off, so I’m a bit cautious about how I chew and what I do so. [21]

Information from an oral health professional was considered indicative. No respondents offered evidence from a third party who was not an oral health professional.

I know from my last check-up that my gums were in good health. [39]

And he’s had check-ups and got the all clear and told that the teeth are good. [2]

Treatment need (whether it be historical treatment, knowledge that there is a current need for treatment, or a view that no treatment has been needed in the past), was proffered as evidence by some respondents.

[Base my assessment of my oral health on] the number of fillings and the broken teeth, root canals er, and, you know, the amount of work that’s gone into keeping all my teeth. [21]

With that tooth that’s almost all gone, you know [so my oral health is not too flash] [26]

I’ve got no fillings [so my oral health is pretty good]. [31]
A large number of respondents explained that their oral health was good (or poor) because of the care they put into looking after their teeth (or not). There seemed to be a logic that, if they put the effort in, they would be rewarded with good oral health.

[My oral health is not too bad] because I brush my teeth every day and I try and look after my teeth. [7]

[My oral health could be better] I suppose I don’t take very good care of them. [8]

Well, ‘cos I look after it regularly [which is why my oral health is just above average]. [37]

In general, it seemed to be agreed that good oral health is characterised by white, straight teeth, which have needed little treatment in the past, currently give no symptoms, allow trouble-free eating, are well cared for, and have been labelled as healthy by an oral health professional. With only one exception, those who expressed any of the above considered their oral health in a positive light, while those expressing the converse saw their oral health in a negative light. However, one individual who knew he needed fillings still felt that his oral health was not bad.

I know I’ve got a couple of holes in there, but yeah. I think it’s [my oral health is] not bad, you know, for my age. [14]

All participants were invited to discuss, during the semi-structured interview, what they could do (or they would recommend others could do) to improve oral health. All participants could list the elements of oral hygiene, diet and dental visiting as being important for oral health. Very few participants felt the need to explain these further and many statements were accompanied by qualifiers such as ‘they do say’ or ‘obviously.....’ indicating that such activities are well known as being good for oral
health. All respondents were correct in their suggestions of good oral health behaviour, except one parent of a child with haemophilia who stated that his son would keep healthy teeth by drinking Bournvita (hot chocolate) every night.

3.12 Previous dental experience

The majority of participants described previous dental experiences, usually as an illustration of why they did or did not attend appointments. Many participants described unpleasant past experiences, with the three main themes being: lack of anaesthesia; bleeding; and the additional procedures required to facilitate dental treatment. A few participants described previous experiences as good, usually expressing that this was a surprise.

Many participants described how dentists were reluctant to use local anaesthesia when treating them, which they ascribed to the fear of causing bleeding.

And I’m also very – have long memories of sitting in the chair and it just being utterly, excruciatingly painful because we didn’t have any sort of pain killer on board, local anaesthetics or anything [20]

He just. He said to me “do you want an injection?” He said “I’m not over keen on giving you an injection in case it bleeds.” I said “oh, don’t worry about it. Just go ahead and do it.” He said “it’s gonna hurt.” I said “well, it’ll be over and done with in a minute. What the hell.” Put up with it. Yeah. Didn’t like it, but. [14]

Like, we had a dentist friend in Tauranga and he knew my condition but his associates didn’t, who I went to see, and they were quite nervous about putting a needle in my gums for fear of bleeding. [25]
Bleeding after dental treatment (particularly extractions) was widely described by participants.

I had an experience about 25 years ago now where I had a tooth out and the dentist said, you know, everything was fine and I had the Factor 8 treatment but then it kept bleeding, just oozing a wee bit, and I went back to the dentist and he said it was natural, it was just a little bit of blood and mixed in with your saliva. So we ignored it basically and then about three days later I had a faint spell up town and when I got in they said that I’d lost a huge amount of blood. [15]

I mean having a tooth removed, which I’ve had several removed, is just a major drama. The last – well, not the last one, the second to last tooth I had removed, even though it was stitched up and I was on treatment, bled profusely for six weeks. [20]

Oh, the first few times [I had teeth out] I used to need blood transfusions. [10]

The additional input needed when someone with a bleeding disorder requires dental treatment was described by participants as adding to the overall unpleasant experience.

They do, um, because, you know, the process of even having a tooth out is made harder if, for those with haemophilia, because you need to then correspond with two sets of people, the dentist, the er, you know, the haemophilia people, clinic, so it sort of just makes it a little bit harder if you have to get all that work done. [15]

You know, cos it wasn’t just as simple as me treating myself and just rock up to the dentist and, you know, fine. It would be like go to the
dentist, go to the hospital or someone had to give me treatments and then I had to go up to the dentist and – or else they’d keep me overnight just for a tooth extraction type of thing or they’d keep me for days. [19]

So that was, that was sort of a choice not to go through the whole hassle of trying to get cover and it wasn’t so easy then we didn’t have a haemophilia nurse and just lots of drama. [21]

Not all dental experiences were unpleasant, and some participants expressed surprise that the visit was not as bad as they were expecting.

But I had my teeth out here, this year, didn’t feel a damn thing, you know, I didn’t even know I was having them out. Magic. It’s beautiful. [14]

The hospital have done it twice [extractions] and been absolutely perfect. Didn’t look like it, but. Bunch of kids in there doing it, they made a great job. You don’t always get what you think you’re going to get. Yeah. Didn’t spill a drop. It was very good. [37]

I had to have my tumour out, 24 [teeth out] and stitches there. There was no pain and there was hardly any bleeding. You know, I was, I was, I was amazed. [24]
3.13 Perceived importance of oral health

The majority of participants expressed the view that oral health was important. When adult participants were asked various questions using a Likert scale, oral health was deemed by most participants to be somewhat or very important (Table 19).

When asked to prioritise spending on dental care against spending on other healthcare, dental spending was considered less important than most other areas of healthcare except spending on non-urgent surgery (such as cataracts or hip-replacements; Figure 17).

Table 19 - Perceived importance of oral health

<table>
<thead>
<tr>
<th>Survey questions</th>
<th>Very important</th>
<th>Somewhat important</th>
<th>Neutral</th>
<th>Not very important</th>
<th>Not important at all</th>
</tr>
</thead>
<tbody>
<tr>
<td>Do you think that dental problems in adult teeth are:</td>
<td>18 (72)</td>
<td>7 (28)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>How important is it to you that the government spends money on improving the oral health of adults in New Zealand?</td>
<td>9 (36)</td>
<td>11 (44)</td>
<td>5 (20)</td>
<td>0 (0)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>Do you think that dental problems in children's teeth are:</td>
<td>21 (84)</td>
<td>4 (16)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>How important is it to you that the government spends money on improving the oral health of children in New Zealand?</td>
<td>20 (80)</td>
<td>4 (16)</td>
<td>1 (4)</td>
<td>0 (0)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>How important do you think the health of your mouth and teeth is to your general health and wellbeing?</td>
<td>15 (60)</td>
<td>10 (40)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td>0 (0)</td>
</tr>
</tbody>
</table>
Figure 17 - Number of respondents who thought government spending on other areas of healthcare was more important or less important than government spending on dental care
(Responses may total less than 21 because participants could also respond that the issue was of equal importance)

When explaining why oral health was important, participants expressed this either by describing the impact on day-to-day functioning or quality of life, (which is reported on page 44), or the wider effect on health. Some participants felt they understood how oral health impacted on the body, either specifically or on a more generic level.

Because if you get an infection there, it travels to the rest of your body. And if you’ve got heart problems, well, you’d probably know that anyway, that can cause big problems, bigger problems, which you don’t need. [14]

I think it’s very important when - from what I understand it [oral health] has a lot of adverse reactions later on in life as far as heart disease goes. [25]

If you’ve got a bad mouth it, that can muck up your immune system, so if you’ve got an infected mouth it can make things a lot worse. For your general health. [17]
Other participants had examples of how their bad oral health had affected the rest of their body, whether real or imagined.

You know, I always, I wondered why I was always feeling crook. But it was, yeah, you know. Yeah, so, but now [I've had my teeth out] I'm good as gold. [29]

I’ve had infections in the mouth before that’s caused headaches, neck pain, back pain, all the rest of it, you know, there are other sort of things that lead on, you know. [26]

I think it was causing me to bleed more as well in my joints. Might have just been psychological, I don’t know, but I did, anyway. [interviewer asked for clarification] So my target joint is this knee, and I had quite a bit of problems with that until I got my tooth fixed. [39]

However, despite one participant responding that he felt that the health of his mouth and teeth was very important to his general wellbeing, he was a little more realistic during the interview.

It’s not like the sort of life and death health problems that you tend to consider and worry about more than anything else. [12]
3.14 Oral hygiene behaviours

3.14.1 Frequency of oral hygiene behaviours

Figure 18 shows the frequency of tooth-brushing by adults and children. All participants used a fluoride toothpaste every time they brushed their teeth. Fewer than half of the participants (13 or 44.8%) brushed twice a day or more.

Figure 19 shows the use of interdental aids and mouthwash by adult participants. Three adults (14.3%) used mouthwash daily. Interdental aids were used much less often, with none using them daily.
Figure 19 - Frequency of use of interdental aids and mouthwash by adult participants

Participants talked about using toothbrushes, interdental aids and mouthrinse, including using sodium bicarbonate mouth rinses. Participants had a number of different explanations for their oral hygiene practices; these could be grouped into ‘reward’, ‘routine’, ‘symptom’ and ‘fear’ categories.

Some people carried out oral hygiene because of the good way it made them feel.

And I find that I actually don’t feel good if I don’t floss. [3]

I just did it [tooth-brushing] because I wanted a fresh mouth. That’s why I did it. And it made my teeth look nicer too, you know, doing them quite vigorously every day. I had a nice pair of ivories you know. [24]

I sort of just floss because it feels better once you’ve got a good.....[clean] [39]
Those who spoke about the importance of routine were almost all talking about tooth-brushing.

Every day I clean my teeth, without fail. It’s a ritual. Shave, teeth. [35]

Like it is just a habit to brush my teeth in the morning and at night, like, I don’t get lazy with that ‘cos it’s just a habit. I just do it before I go to bed every night, anyway. [39]

6: Half and half. I’ve got the morning down, I just don’t have the night. [for toothbrushing]

EH: Right. And is that because you vary what you do in the evening?
6: Yeah. In the mornings it’s usually the same thing over and over. [6]

Some individuals, including all those who spoke about using interdental aids other than floss, only used them when they had particular symptoms.

Oh, I would say – if I’ve got something stuck in my teeth, that’s when I get the odd toothpick out. Let’s say, it might be say like once a month. [17]

Occasionally my wife will say “oh, your breath’s a bit off” so I gargle with [mouthwash], it’s blue, but very seldom though ’cos I don’t think I’m too bad. [35]

For some, the fear of what might happen if they did not keep using oral hygiene products was enough to keep them using them.
I mean, I have been flossing for a very long time because it was always drummed into me again how important it is to floss around that area so that I don’t get an infection. [11]

Yeah cos it’s [using mouthwash] better than losing your teeth. [5]

For those who explained why they did not practise certain oral hygiene behaviours the reasons fell into three groups: material difficulties, ill-effects and lack of priority.

The types of material difficulties encountered with carrying out oral hygiene were either linked to a lack of knowledge or education, or to physical difficulties with actually carrying out the act.

I don’t even know how to do it. [flossing] [7]

Just a round out hard thing to do. [flossing] [6]

Like, the only setback is, like, I was going to say before if, like, I have a bleed that can kind of throw everything out of whack, and um, you know, just depending on how severe it is. Like, if it’s this - ‘Cos I am right-handed, but I can brush my teeth with my left and, like, I try to, but if it’s really sore then kinda everything goes out the window and I don’t do anything until, like, it feels better. [19]

Many individuals gave examples of the ill-effects they had experienced after carrying out oral hygiene procedures; the majority were associated with bleeding after flossing.
I tried dental floss until I discovered that caused bleeding, and I tried mouthwash until I read a whole lot of articles that said that may not be the greatest idea. [13]

Maybe floss more, but then I also heard that flossing is not necessarily good for your gums. Especially maybe as a haemophiliac. [Because] it causes bleeding. [39]

Because my teeth are really close together and I find it [flossing] actually quite painful, and it does make my gums bleed. They are really sensitive and I think it is that whole haemophilia thing that they'll bleed at the drop of a hat and I just don't like it so I don't do it. I know that I should do it more, but I don't. I don't like the blood in my mouth. [5]

By far the largest group of reasons given for not practising oral hygiene were those showing that it was a low priority. This came across as a lack of time to carry out oral hygiene by many, although some had the self-awareness to identify it internally as laziness or to describe it as a low-priority activity.

I normally end up being pretty busy after work, that would be about it. [so no time to brush teeth] [10]

I don’t know [why I don’t floss]. Just laziness. Rushing out the door. Brushing your teeth and then going “I’ve got five minutes to get somewhere.” [11]

Lazy, I guess. And I just felt like - I didn’t put it high on my priority list, to be fair [19]
Can’t be bothered \textit{to brush teeth twice a day} [17]

When talking about what would help them practise oral hygiene behaviours more often, the responses fell into the three categories of external factors, access factors and internal factors, with the latter being a very strong theme.

External factors included both ‘sticks’ (sanctions) and ‘carrots’ (rewards), with ‘carrots’ being important in those interviews where the individual with haemophilia was a child.

The thing that would definitely make me do it is if you guys paid me. [1]

Brush your teeth more and the way I want, \textit{then I will} borrow something for you. Or maybe this week we can er, go somewhere he like to go, like the swimming pool or, you know, that’s like a favour. If he does my favourite, \textit{brushing}, I can do his favourite. [28]

Aspects of improving access included specific actions that the individual could do to make practising oral hygiene easier.

If the floss was – actually if I kept it in my pocket I’d floss more. [36]

You know, I don’t actually bring like a toothbrush to work. Maybe I should, you know. [26]

Internal factors, such as overcoming laziness or taking responsibility, were commonly mentioned. Many individuals felt that it was their failing that was stopping them looking after their teeth as they should and only they could make it happen.
Well, you know, I mean, if you’re talking about my oral health then, you know, it’s basically up to me to sort that out, you know. [26]

I mean I just need to do it [flossing]. [11]

And I just felt like - I didn’t put it high on my priority list, to be fair, um, so once I started taking responsibility – like it’s my mouth, they’re my teeth, nobody else is going to look after them. Um. You know, it’s just like managing haemophilia. [19]

3.15 Smoking

All adult participants were asked about their smoking habits. Current smokers were considered to be those who had smoked at least 100 cigarettes in their lifetime and had smoked in the previous year. Ex-smokers were considered to be those who had smoked at least 100 cigarettes but had not smoked in the previous year. Non-smokers were those who had smoked less than 100 cigarettes. Eleven participants (44.0%) were non-smokers, seven (28.0%) were ex-smokers, and seven (28.0%) were current smokers.

3.16 Summary of Results

The questionnaire, clinical examination and interview data presented in this section paint a picture of a sample who have experienced oral disease and have varied perceptions of oral health. They acknowledge the importance of oral health but do not always carry out behaviours conducive to oral health, with many different barriers being described.
4 Discussion

This section triangulates data from different sources within this study and interprets them within the context of the strengths and limitations of the study, other New Zealand data, and other studies investigating the oral health of those living with haemophilia world-wide.

4.1 Key findings

Thirty-three participants took part in the study. One in six adult participants were edentulous, which was significantly associated with being dentally anxious; not completing formal education; and living further away from the hospital. One-third of dentate adults had untreated caries. The mean DMFT for adults was 8.6. Nearly three-quarters of adult participants had one or more teeth with 4mm attachment loss, one quarter with 5mm attachment loss, and one in twenty with 6mm attachment loss. Three in five had one or more teeth with a pocket of 4mm, one in five with one or more 5mm pocket and no participants had any pockets of 6mm or more. A quarter of all sites had bleeding on probing.

Six out of eight children (80%) with deciduous teeth were caries-free. In the deciduous dentition the mean dmft was 1.2 and there was no untreated caries. The mean DMFT in the permanent dentition was 2.1, with nearly three-quarters of children with permanent teeth being caries-free and one in seven having untreated caries. Overall, three in five children were caries-free and the mean dmft/DMFT was 2.6.

Nearly three-quarters of adult participants (72%) reported no impact from oral health although one third of adults (one in seven children) thought they currently needed dental treatment. One quarter of both adults and children had had oro-facial pain in the previous four weeks. Participants described problems with oral health as either functional or social problems.
Participants were able to articulate what oral health meant to them and had had a variety of dental experiences. Unpleasant experiences were often related to their having haemophilia. Participants were also able to describe why oral health was important.

Over half of adults and six out of seven children visited a dental professional in the previous year with the majority of these appointments being at the hospital. Hospital dental appointments were described as easy/streamlined, safe, and cheap. The reasons given for not attending appointments were categorised as intrinsic, extrinsic or haemophilia-related.

Nearly half of all participants brushed their teeth twice a day or more. Oral hygiene activities were carried out because of reward, routine, symptoms, or fear; while barriers to performing oral hygiene were categorised into material difficulties, ill-effects, or lack of priority.

4.2 Strengths and limitations of this study

As with any research, critiquing the strengths and weaknesses of a study allows the conclusions to be put into context. This section critically examines the study approach.

4.2.1 Study design

This study used a mixed methods approach; this is a research methodology with the central premise that the use of quantitative and qualitative approaches in combination provides a better understanding of research problems than either approach alone (Creswell and Plano Clark, 2007). Describing the oral health of a population and investigating perceptions surrounding oral health and barriers to oral health are questions that require two different data-sets, a scenario best served by using different methods (Bower and Scambler, 2007). In this study, quantitative data were collected on the oral health and oral health behaviours of individuals with haemophilia. Qualitative
methods were used to explore the perceptions of oral health; the importance of oral health; and barriers to oral health. Mixing the two data-sets forms a more complete picture of the problem than using them alone and allows some triangulation of data to find areas of agreement and discord.

4.2.2 Quantitative data

The quantitative data were collected using a cross-sectional survey design involving (1) a clinical examination to obtain data about the oral health of participants, and (2) a questionnaire to obtain data on oral health behaviours and self-perceived oral health. This type of study is very useful in getting a “dental snapshot” of a population. Because these data were taken from a single point in time, no temporal relationship can be determined between any associated factors. Care must be taken then, when considering any associations found in the data.

Questionnaires can demand socially acceptable answers from participants and may be subject to recall bias. The questionnaire used was largely taken from that used in the New Zealand Oral Health Survey (NZOHS) so the questions had been previously piloted and validated. Use of the same questions allows comparison of findings with this survey. Responses were entered into a spreadsheet at the time of administration of the questionnaire which will have eliminated transcription error but provides no method for cross-checking responses.

All clinical examinations were completed by the same examiner, which would eliminate inter-examiner error, and conducted within a dental surgery, which has been shown to reduce error with identifying fillings in root surfaces (Fairhall et al., 2009). Caries was recorded at the level of cavitation, which underreports caries but reduces examiner bias. This is also the threshold used by most national surveys.

The treatment of crowned teeth during analysis may also add to the under- or over-reporting of caries (Pitts, 1997). Deciduous crowns were given an allowance of three
filled surfaces during analysis. This acknowledges that crowns on deciduous teeth are usually placed to treat caries although they may also be placed on non-caries hypoplastic teeth for protection or to reduce sensitivity. Therefore, caries may be over-reported in deciduous teeth.

Crowns on permanent teeth are placed for a wider variety of reasons including after trauma, or for cosmetic reasons. Crowns and bridge abutments on secondary teeth have therefore been excluded from the analysis which may have led to under-reporting of caries. Some 2.1% of permanent teeth and 5.1% of deciduous teeth had been crowned.

Periodontal data were collected from three sites (mesio-buccal, mid-buccal, and disto-lingual) per tooth. This has been shown to be comparable to the gold-standard method of six sites per tooth unless periodontal disease is severe (CAL ≥8mm) (Susin et al., 2005). In this sample, the majority of individuals had attachment loss less than 6mm and the use of three sites per tooth can be justified.

4.2.2.1 External validity

External validity is the ability to generalise from the study population to other populations. In this case, the ability to generalise to all individuals in New Zealand with moderate or severe haemophilia. This is dependent on the internal validity of the study (discussed above) and the sample being randomly selected from the population under study. In this study the population being investigated was defined as all individuals with moderate or severe haemophilia, aged over two years, and residing in the geographical area covered by the Capital & Coast, Hutt Valley, and Wairarapa District Health Boards. Those with mild haemophilia were excluded because they can live for many years without even being diagnosed (Roberts et al., 2010).

Potential participants were identified from patient lists at the two nearest regional centres because a national database is not yet fully functioning. One individual was identified by both DHBs. Theoretically, an eligible individual could have been missed.
if he only ever attended a centre further away, but this is unlikely due to the regular administration of clotting factors and advice these individuals require.

The incidence of haemophilia is 1/10,000 live births. Using this estimate, the population of these three DHBs, being about 478,400\(^8\), should have yielded 48 individuals, higher than the 38 identified. However, this incidence includes those with mild haemophilia, who were not included in this study. The total eligible population was small (38 individuals), so all were invited to participate, eliminating selection bias.

The overall participation rate was 86.8%, including a 100% participation rate for those aged under eighteen. This compares well with recent cross-sectional clinical surveys with the national and state oral health surveys of New Zealand, Australia and the United Kingdom having participation rates ranging between 33.2% to 57.5% (AIHW Dental Statistics and Research Unit, 2008; Ministry of Health, 2010b; Steele and Cooke, 2011). The Dunedin Multidisciplinary Health and Disability Study had a higher participation rate at age 32 (for those dentally examined) of 96% (Thomson et al., 2010) but this is more correctly considered a longitudinal cohort study.

Some socio-demographic data for non-participants were not collected, so there is no ability to compare these factors. Otherwise, participants and non-participants were fairly similar with non-participants more likely to have moderate haemophilia, be Maori, live in a city and live closer to Wellington Hospital than participants.

With very little possibility of selection bias and a high response rate, it is likely that any findings are representative of individuals with moderate or severe haemophilia living in the Wellington region. The ability to generalise these data to all New Zealanders living with haemophilia is discussed later in this chapter.

4.2.3 Qualitative data

Very little is known about the perceptions of oral health of PWH, so the use of open-ended questions, giving a rich data-set, is appropriate (Stewart et al., 2008). The qualitative data were collected by the use of semi-structured interviews, which were then transcribed, checked and analysed for themes. An inductive method was used for analysis, meaning that there was no preconceived framework. However, this method is interpretive in nature, and the researcher’s views and values can have an impact on the data collection and analysis; the relationship between the researcher and participant may also affect the data (Bower and Scambler, 2007; Masood et al., 2010). In this study, the researcher was an experienced hospital dentist previously known to several participants; indeed, the researcher had provided dental treatment to some participants in the past. An existing rapport with participants may have allowed a more frank discussion of the participants’ beliefs and understanding of oral health. Conversely, the dentist-patient relationship may have encouraged desirable answers in the questionnaire. Use of both a questionnaire and interview gives more validity when multiple methods give similar findings.

There is still some debate over the process and methods of validating thematic content analysis (Burnard et al., 2008; Masood et al., 2010). Where possible, the results of the thematic analysis were considered alongside data collected by other methods. The advantage of this type of presentation, particularly for this small sample, is that comparisons of findings from different methods can be more easily compared. Instances were findings lead to the same or to different conclusions are more easily identified and the validity of the findings can be more readily assessed.

The total number of those completing the interview was determined by those available rather than saturation being reached. However, very few new themes were being found from the interviews carried out later in the study, so it is likely that the study achieved data saturation.
4.3 Oral health status

To provide some context, the data from this sample have been compared with those reported in the 2009 New Zealand Oral Health Survey (NZOHS) (Ministry of Health, 2010b) and other New Zealand studies, where available. Where gender-specific data concerning males is reported, this is also presented for comparison here because the study population comprises males only. The sample was too small for any meaningful statistical analysis, but a broad-brush overview can be seen.

Ethnicity and deprivation have both been shown to be associated with oral health status. In the sample, the highest educational level reached was recorded as a proxy for deprivation. Table 20 summarises the ethnicity and highest educational level achieved for those with haemophilia and those without haemophilia in the three DHBs participating in this study. That Maori were over-represented in the haemophilia population may be related to haemophilia being an X-linked condition and therefore found in clusters of families.

Table 20 - Comparison of ethnicity and highest education level attained by those with haemophilia and the general population within the same geographic region

<table>
<thead>
<tr>
<th>Ethnicity</th>
<th>Participants % (N)</th>
<th>Non-participants % (N)</th>
<th>Total % (N)</th>
<th>General population % (N)</th>
</tr>
</thead>
<tbody>
<tr>
<td>NZ European</td>
<td>60.6% (20)</td>
<td>60.0% (3)</td>
<td>60.5% (23)</td>
<td>66.5%</td>
</tr>
<tr>
<td>Maori</td>
<td>21.2% (7)</td>
<td>40.0% (2)</td>
<td>23.7% (9)</td>
<td>12.1%</td>
</tr>
<tr>
<td>PI</td>
<td>6.1% (2)</td>
<td>0%</td>
<td>5.3% (2)</td>
<td>8.0%</td>
</tr>
<tr>
<td>Other</td>
<td>12.1% (4)</td>
<td>0%</td>
<td>10.5% (4)</td>
<td>13.5%</td>
</tr>
</tbody>
</table>

Highest educational level attained²

<table>
<thead>
<tr>
<th>Level</th>
<th>Participants % (N)</th>
<th>General population % (N)</th>
</tr>
</thead>
<tbody>
<tr>
<td>None</td>
<td>12.1% (4)</td>
<td>19.6%</td>
</tr>
<tr>
<td>High school</td>
<td>33.3% (11)</td>
<td>47.6%</td>
</tr>
<tr>
<td>Diploma</td>
<td>36.4% (12)</td>
<td>9.6%</td>
</tr>
<tr>
<td>University</td>
<td>18.2% (6)</td>
<td>23.2%</td>
</tr>
</tbody>
</table>

¹http://www.stats.govt.nz/Census/about-2006-census/district-health-board-area-summary-tables.aspx accessed 7/2/12; ²Not available for non-participants
4.3.1 Caries experience in children

The oral health of children in this sample was generally good, with 62.5% of children being caries-free, a mean DMFT/dmft of 2.6, and only one individual having any untreated caries. The only individual who had untreated caries was also the only child who had not visited a dental professional in the previous year. This same individual had a DMFT score of 14, illustrating the very skewed distribution of caries experience in this population.

The estimates of the number of teeth with caries experience in the study population are compared with those reported in the NZOHS in Figure 20. Children with haemophilia had fewer deciduous teeth that have experienced caries and more permanent teeth with caries experience than other children. This pattern is replicated when comparing untreated caries (Figure 21), with there being less untreated caries in deciduous teeth and more untreated caries in permanent teeth (albeit only one surface in one individual). However, the caries distribution is usually highly skewed in developed countries (Burt, 1998) and the proportion of children who are caries-free should also be considered when investigating oral health. This comparison is shown in Figure 22. For each subgroup (primary teeth, permanent teeth, and all teeth), a higher proportion of those with haemophilia were caries-free.

Figure 20 - Comparison of caries experience in children
When considered together, these three figures indicate that the caries experience in the sample of children with haemophilia is highly skewed, maybe more so than in the general population. To paraphrase Kaste (1996), 63% of the children were caries-free,
25% of the children had 33% of all carious lesions, and 13% of the children had 66% of all the carious lesions.

Caries is a multifactorial disease with risk indicators that include age, gender, ethnicity and education (Ministry of Health, 2010b). The small numbers in the study group make it difficult to control for these indicators. One way to compare this study group with the general population while controlling for these risk indicators would be to weight individual subgroups of NZOHS to match the haemophilia population, as was done by Boyd in Northern Ireland (Boyd and Kinirons, 1997). This would be likely to introduce less bias than a case-control comparison where the selection of a comparison group is problematic. This weighted approach would be appropriate in New Zealand as the sampling method for the NZOHS was designed to give robust population estimates for subgroups. However, even with the UK national survey data, Boyd et al were unable to determine weighted means for the numbers of missing teeth.

However, whatever its composition, the sample had a highly skewed distribution of caries and generally had less caries experience than the general population. This difference is more obvious when considering the deciduous teeth. This conclusion is reflected in the parents’ perception of their child’s dental care in the qualitative results (page 57).

4.3.2 Edentulism and number of teeth remaining in adults

When compared with NZOHS data for dentate people, those with haemophilia were less likely to have a tooth missing and also more likely to have 21 or more teeth; of those who were dentate no-one had less than 19 teeth. Conversely, the edentulousness rate was higher in those with haemophilia, at 16.0%, than in New Zealand males, of whom 8.8% were edentulous (Ministry of Health, 2010b).

The occurrence of edentulism does not bear a close relationship with disease prevalence, and the decision to undergo the transition to an edentulous state involves
many factors (Cohen, 1978; Sussex, 2008). Treatment decisions made by dentists are not just about the extent of pathology (Kay and Blinkhorn, 1996) and these data may be an indication that different clinical pathways have occurred for those edentulous individuals with haemophilia than those in the rest of the population.

Qualitative data from the four participants who had had all their teeth extracted suggested that, while the condition of the dentition (usually the gums) was one factor, other considerations (such as the presence of dental anxiety, the impact of dental sepsis on other interventions, and pragmatism about the haematology support required for dental treatment) were also important.

The, um, the health of my teeth was pretty bad. And, when I was given the option to get them out, I chose that because I have a real fear of dentistry. I hate it with a passion. So given the option of having false teeth and never have to visit a dentist again, sort of thing, was very pleasing. [15]

But they were saying these [teeth] need to come out, you know. I was waking up every morning with blood all over the place, you know. [24]

That’s why I was wasn’t going to tell the doctor up in Palmerston for a while, but he says “how’s your general...”, you know, and I says “oh, my teeth are coming loose, you know” and of course lucky I told him otherwise I most likely wouldn’t be here now [as the health of my mouth was severely affecting my general health] [29]

But I’ve got a feeling, I’m not sure on this one either, but the doctors, you know, if you’re going to take a couple of teeth out from a haemophiliac, oh, you know, take them all out, sort of thing, because it’s uh [just as easy to take them all]...... but also I was scheduled for a
month or two later for a knee replacement, and an operation here \[indicates\ \text{groin}\], and I’ve got a feeling the doctors, the health of my mouth, teeth, with the other operations, you know, and by then the trouble I was having with the abscesses I think \[the\ \text{doctors thought it was better to have all my teeth taken out in one procedure}\] [27]

Extraction of a tooth in someone with moderate or severe haemophilia is not straightforward. It requires liaison between dental and haematology services, additional monitoring and, in some circumstances, hospital admission (Brewer and Correa, 2006). It is, then, not unsurprising that fewer people with haemophilia have missing teeth or that they are more likely to have more than twenty-one teeth. However, the data suggest that, while the decision to remove teeth is slow, it appears that, once a critical stage (either in disease severity or number of teeth left) is reached, the decision to extract all remaining teeth may be made much more quickly than for someone without haemophilia.

Those who were edentulous in this sample were significantly more likely to have not finished high-school education, to live further away from the hospital, and to be dentally anxious. These factors have all been shown to be associated with edentulousness, in varying degrees, in other populations (Sussex, 2008) and are further discussed later in this chapter.

4.3.2.1 Dental anxiety

Dental anxiety in this study was measured by the well validated Corah Dental Anxiety Scale (DAS) (Corah, 1969; Corah et al., 1978) and was found to be significantly associated with being edentulous. Overall, about a quarter of the sample were dentally anxious. Fiske et al (2002) found that 50\% of those with bleeding disorders in the UK were reluctant to visit the dentist due to fear, although this category included both general dental anxiety and fear that the dentist did not know enough about treating those with haemophilia. Kalsi et al, also in the UK, found 17\% of those with a bleeding disorder felt apprehensive about visiting the dentist (Kalsi et al., 2011). Neither of these
studies used the DAS to measure dental anxiety and it is therefore difficult to compare the findings with those of the current study.

Edentulism has been found, by other authors, to be associated with both high dental anxiety (Locker and Liddell, 1992) and low dental fear (Armfield et al., 2006). Those with high dental anxiety are less likely to attend routine dental appointments (Locker et al., 1991; Thomson et al., 2000) and it has been hypothesised that delayed presentation by those with dental anxiety is more likely to result in tooth loss (Sussex, 2008). It is possible that the traumatic event of being rendered edentulous creates dental anxiety. However, the qualitative data in this study suggest otherwise.

Other authors have investigated dental anxiety in specific New Zealand populations. The severity of dental anxiety in the sample (DAS score 9.1) is very similar to that found on the West Coast of New Zealand (9.8) (Thomson et al., 1999) and in Dunedin 26- and 32-year-old males (9.2 and 9.0 respectively) (Thomson et al., 2009). The prevalence of dental anxiety in the sample (24.0%) is also similar to that reported in the West Coast (20.5%) (Thomson et al., 1999) and in 26- and 32-year-olds from Dunedin (20.8% and 18.4% respectively) (Thomson et al., 2009). This suggests that dental anxiety on its own does not lead to a higher proportion of edentulous individuals in the haemophilia population. However, once dental anxiety has delayed the presentation to a dental professional, it is likely that having haemophilia alters the treatment plan.

4.3.3 Caries experience in adults

Lower scores for all caries indices are seen in those with haemophilia than for New Zealand males as reported in the NZOHS. This is particularly striking when considering the numbers of missing teeth, as discussed in the previous section. The sample had a mean of 8.6 teeth with caries experience (13.6 for male NZOHS respondents) comprising 2.1 missing teeth (4.6 for male NZOHS respondents), 5.8 filled teeth (8.0 for male NZOHS respondents), and 0.7 decayed teeth (1.0 for male NZOHS respondents). It is possible that those with more caries have been rendered edentulous
(as discussed in the previous section), although the qualitative data suggest periodontal disease is a more likely cause for the teeth to be extracted.

4.3.4 Periodontal health in adults

This is the only study that has reported periodontal disease in PWH by full mouth probing. Ugolotti et al (2004) partially probed pockets in PWH (with no significant difference between study and controls) but did not report findings or what was recorded. Other studies investigating periodontal disease in PWH have used gingival inflammation indices (Albayrak et al., 2006; Azhar et al., 2006; Hanagavadi, 2006; Sonbol et al., 2001; Ugolotti et al., 2004) or estimated bone loss from radiographs (Ziebolz et al., 2011).

Periodontal condition cannot be adequately assessed without probing. Periodontal disease is a collection of chronic and acute inflammatory diseases, caused by bacteria, affecting the periodontal supporting structures of the teeth. There are many modifying factors, and the presence of inflammation does not necessarily lead to periodontal destruction. It is important that separate measures of both inflammation and attachment loss are made to assess periodontal disease (Beck and Loe, 1993).

No studies identified why they did not assess periodontal condition by probing when this is the accepted method. It may have been avoided because of the risk of causing periodontal bleeding. The NZOHS excluded from periodontal probing any participants who declared a bleeding disorder because of this risk, although the number of participants excluded for this reason (if any) is not known⁹. The WHO guidelines for oral care and dental treatment (Brewer and Correa, 2006; Scully et al., 2008) do not mention probing. They do, however, state that ‘non-surgical dental treatment’ can be carried out for mild-to-moderate haemophilia under anti-fibrinolytic cover. The lack of mention of periodontal probing suggests that it is considered a low-risk activity and does not require additional haemostatic cover. The decision to use periodontal probing in this study was made after a consideration of the benefits (accurate assessment of

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⁹ Personal communication
periodontal disease) and the risks (considered low by the haemophilia team). Probing in this study was carried out in a hospital setting with the knowledge of the haemophilia nurse. Patients were fully informed, were able to decline probing, and were offered tranexamic acid. No participants declined probing and, while all but one experienced bleeding on probing, this was short-lasting (less than five minutes) and no participants required additional medical intervention subsequent to the examination.

Figure 23 shows the comparison of CAL and pocket depths at different thresholds of periodontal disease of the sample and NZOHS.

![Figure 23 - Comparison of prevalence of periodontal disease at different thresholds](image)

Lower prevalence estimates with more stringent case definition thresholds were observed in all groups. This gradient, however, appears to be steeper for those with haemophilia, demonstrated by a higher prevalence in those with haemophilia for more mild disease and a lower prevalence for more severe disease. This suggests either that periodontal disease progresses differently in those with haemophilia, or that something different happens to those who develop severe periodontal disease to ‘cure’ the disease.
It is likely that those with deep pockets (>6mm) experience more, or even spontaneous, bleeding. For someone with haemophilia, this bleeding will be prolonged and these teeth will become problematic. Indeed, several participants described such scenarios (page 45). Qualitative data from the edentulous participants (page 93) suggest that gingival bleeding is a factor in the decision to move to edentulousness. Therefore, anyone with haemophilia who develops severe periodontal disease is likely to have all teeth removed and no longer have periodontal disease.

Periodontitis is a multifactorial disease caused by the periodontal tissue response to plaque (Bagg et al., 2005). For attachment loss to occur, then, there must be bacteria present and an underlying predisposition. Disease progression is also influenced by other factors such as smoking and general health. Differences in any of these may help explain why PWH appear to have more periodontal disease.

4.3.4.1 Oral hygiene

Adults in this study were less likely to brush their teeth twice a day or more than those without haemophilia as reported in the NZOHS (57.0% and 52.4% respectively, among males). This difference was more apparent among children, of whom only a quarter (25.0%) brushed their teeth twice a day or more, far fewer than the 58.7% of New Zealand boys (Ministry of Health, 2010b) and 66.8% of Auckland 6-year-olds (Paterson et al., 2011) reported to brush twice a day or more.

In studies in Turkey and Pakistan, those without haemophilia were more likely to be regular brushers (more than once a day) than those with the condition (Alpkilic Baskirt et al., 2009; Azhar et al., 2006). Kabil et al (2007) found that fewer than half of their participants with haemophilia used any form of oral hygiene and only 16.7% used toothbrush and toothpaste, although there was no comparison group used in this Egyptian study(Kabil et al., 2007).
When considering other oral hygiene practices, 44.2% of 32-year-olds living in Dunedin reported that they floss daily (Broadbent et al., 2011), far more than the 4.8% of adults with haemophilia reporting the same.

These findings, together with the qualitative findings (page 79), support the hypothesis of Azhar et al (2006) that those with haemophilia are less likely to practise oral hygiene due to the risk of bleeding (Azhar et al., 2006). For those with severe periodontal disease, experiencing spontaneous and prolonged gingival bleeding (as illustrated in the qualitative data), there is a real impact on quality of life and, depending on the duration, may involve significant blood loss. However, bleeding associated with oral hygiene, similarly to that associated with periodontal probing, is usually of short duration and does not present any real danger. International guidelines concerning oral health in those with haemophilia stress the importance of good oral hygiene practices (Brewer and Correa, 2006; Scully et al., 2008).

Good oral hygiene is an important factor contributing to good oral health. Harmful bacteria are removed and medicaments such as fluoride and antimicrobials are introduced through toothpaste. Participants, especially children, in this study reported brushing less than the general population. The low number of child participants brushing their teeth twice a day is worrying as a relatively stable pattern of toothbrushing is established during childhood and adolescence (Kasila et al., 2006) and beliefs about the importance of toothbrushing change little over time (Broadbent et al., 2006).

4.3.4.2 Gingival inflammation

Gingival inflammation, characterised by bleeding on probing, does not directly result in attachment loss but is often used as a predictor of disease progression. Having haemophilia does not increase the risk of bleeding, just the ability to stop bleeding once it has started. However, gingival bleeding may be more noticeable in PWH because the bleeding lasts longer.
All but one participant experienced gingival bleeding on probing, although only 24% had experienced it in the previous four weeks. No studies have been found that report bleeding index in the general New Zealand population. Azhar et al, in Pakistan, found that 17% of participants with bleeding disorders (but only 12% of a comparison group) reported bleeding on brushing (Azhar et al., 2006). Research in children has suggested that there may be differences in the underlying susceptibility to gingival inflammation between those with and without haemophilia (Albayrak et al., 2006). This was not seen in the current study; however, it had low numbers and a dichotomous scale was used for the severity of haemophilia.

4.3.4.3 Smoking

Participants in this study were slightly more likely to be or to have been a smoker (56.0%) than the New Zealand population (41.5%) (Ministry of Health, 2010c), or males living in the same geographical area (41.1%)\textsuperscript{10}. Smoking is a well documented cause of periodontal disease (Gelskey, 1999) and may be a contributing factor to the greater prevalence of mild and moderate periodontal disease. Only one participant mentioned that stopping smoking could help oral health, and this may be an area where a health strategy could be targeted.

In summary, there is a suggestion from the data that those with haemophilia are more likely to have mild periodontal disease but, as their periodontal disease increases in severity, those individuals are more likely to progress to being edentulous. This higher prevalence of periodontal disease may be the result of an artefact, a difference in underlying susceptibility, a higher prevalence of smoking, or less effective oral hygiene, with the oral hygiene hypothesis having more evidence to support it.

4.4 Self-reported dryness

Low saliva flow, which has many causes, will lead to greater caries risk (Kidd and Joyston-Bechal, 1997). Saliva flow was not objectively assessed in the current study, but respondents were asked a single question about how often their mouth felt dry (xerostomia). The prevalence of xerostomia has not been reported in other haemophilia populations, but has been reported in New Zealand in a study of 32-year-olds in Dunedin (Thomson et al., 2006). The Dunedin study respondents were more likely than the sample to never have dry mouth (21.2% and 12.5% respectively) but this is likely to be age-related because they were aged only 32 in this study and the prevalence of xerostomia is considered to rise with age. While many drugs have xerostomia as a reported side-effect, drugs which may be more commonly taken by PWH (such as tranexamic acid, clotting factors, and anti-retrovirals) do not (MIMS, 2012).

4.5 Self-reported oral health

All participants were asked to rate their oral health on a five-point categorical scale. A higher proportion of adults than children rated their oral health as poor or fair (29.2% and 12.5% respectively). These proportions were similar to those reported in the NZOHS.

One quarter of parents of a child with haemophilia (25%) reported that their child had experienced pain in the previous twelve months. This is higher than the 7.0% who reported having pain in the previous year in the NZOHS (Ministry of Health, 2010b) and higher than the 17.0% of 6-year-old Pacific Island children in Auckland reporting toothache in the previous year (Paterson et al., 2011). Similar numbers of adults, both with haemophilia and in NZOHS respondents, reported pain in the previous four weeks (24.0% and 25.4% respectively) – more than reported in a New Zealand study which found that, in the previous four weeks, 6.1% of respondents had had toothache and 7.3% had had discomfort (Dixon et al., 1999).
Experience of pain was not necessarily associated with the perceived need for treatment. Five study respondents reported pain but did not think they currently needed treatment. This could suggest that pain is not thought to be an indicator of poor oral health or treatment need. It also suggests that, although those with haemophilia may be more likely to report pain (as shown in children), it does not necessarily lead to their seeking treatment. The presence or absence of pain was rarely described as a factor when describing their own oral health (page 67); conversely, pain was the sub-domain with the highest mean OHIP score.

It may be that those with haemophilia (and particularly parents of children with haemophilia) are more attuned to recognising and remembering pain, which, in those with haemophilia, may be more readily attributed to causes other than dental.

Well, the problem is that with haemophilia that you don’t always know whether pain is a cramp, a bleed, or something more serious. So I tend to step on pains, any pain around my face I step on straightaway. Um, and if it goes away within 24 hours then I know it was most likely a bleed. I think I’m possibly more attuned [to anything that might be wrong], because I analyse every little pain and cramp that I get, 'cos if I don’t step on it fast if it is a problem, it turns into a bigger problem. [13]

Generally, fewer adults with haemophilia thought that they currently needed dental treatment than adults in the NZOHS (36.0% and 45.9% respectively; 14.3% of children in the haemophilia population felt that they currently needed dental treatment but the corresponding data was not reported in the NZOHS). This was also found to be the case in Turkey, where there was a significantly higher proportion in the comparison group who thought they currently needed treatment than in the haemophilia group (Alpkilic Baskirt et al., 2009).
Twenty-eight percent of adult study participants reported one or more impact on the OHIP scale. This is higher than reported in other New Zealand studies where 12.4% of male NZOHS respondents (Ministry of Health, 2010b) and 23.4% of 32-year-olds in Dunedin (Lawrence et al., 2008) reported one or more impact. This is consistent with the finding in children that those with haemophilia were more likely to report pain.

The mean OHIP score in the sample was lower than found in 32-year-olds in Dunedin (5.2 and 8.0 respectively) (Lawrence et al., 2008). However, those with haemophilia in Turkey perceived their oral-health-related quality of life to be worse than those without haemophilia using both the OHIP and a visual analogue scale (Alpkilic Baskirt et al., 2009). In addition, those with haemophilia also consistently give lower general health-related quality of life scores than controls without haemophilia (Alpkilic Baskirt et al., 2009; Barlow et al., 2007; Beeton et al., 2005).

When using qualitative methods to investigate why quality of life is perceived to be lower by those with haemophilia, Beeton et al (2005) found that those who recognised the benefit of access to adequate clotting factor product (together with those who could compare life ‘pre-factor’) perceived better quality of life. Elements of these themes were seen in this study.

No, so, the Factor cover, the Factor’s better now. It’s more accessible. The cover’s - it’s easier to take. It’s not a big bag of sloppy yellow stuff that you’ve got to try and sieve through and is ineffective. [21]

But as I say, it’s getting easier and easier now with the, with the treatment. We never had any when we were kids. We were just sick and putting up with it. Your knee got bigger and bigger and bigger and bigger and you got one in the groin and it doubled you over you couldn’t move or stand up. Things like that, but now it’s all gone. Now I see my grandkid and I’ve got one grandchild that’s got it, haemophilia, out of the boys um, and he’s good, so good. [14]
Nowadays, er, *dental treatment's* a breeze...... whack the Factor 8 and away you go. Wish I’d had it when I was in my teens or early 20s even. [12]

In summary, when asked in general terms to rate their own oral health, those with haemophilia gave similar responses as the general population. However, when asked more objectively about pain (children) or impacts (adults), those with haemophilia were more likely to indicate that their oral health has an impact on their life. This finding is supported by other literature. It is not clear whether this is due to PWH being more aware of their oral health or of the consequences of poor oral health, or whether they have been conditioned to give ‘bad’ answers. Either way, the presence of an impact or pain does not necessarily drive them to seek care or lead them to believe that they need treatment.

4.6 Oral health in summary

The sample was small, and statistical tests were therefore inappropriate. The available data, from multiple sources, suggest that those with haemophilia have better oral health than the general population, with less caries experience, less severe periodontal disease (but more mild periodontal disease, possibly related to oral hygiene), and are less likely to perceive that they need treatment. However, when oral disease is present, it has more of an impact on the individual, with children more likely to report pain and adults more likely to report OHIP impacts.

4.7 Facilitators and barriers to oral health

Many oral diseases are preventable. However, knowledge and understanding of desired health behaviour is not enough on its own to encourage that behaviour (Ajzen, 1991). This was demonstrated in this study where participants had a good understanding of
behaviours that improved or damaged oral health but did not always practise them. This was discussed with respect to oral hygiene previously (page 98).

Prophylaxis is a preventive health behaviour for those with severe haemophilia whereby clotting factor is administered regularly, in the absence of acute symptoms, to prevent the arthropathies associated with recurrent joint bleeds. Damage to joints from recurrent bleeds can be painful and debilitating. Despite this, young people with haemophilia often have poor compliance with prophylaxis. Forty-one percent of adolescents on preventive prophylaxis did not follow the prescribed treatment regimens despite 60% of them giving additional clotting factor prior to sports indicating that they believed prophylaxis to work (Lindvall et al., 2006). Conversely, Park found that more PWH exercised to keep fit (another health-promoting behaviour) than in the general population (Park et al., 1995).

4.7.1 Barriers to performing oral hygiene

Fewer individuals with haemophilia perform regular oral hygiene procedures than in the general population, and this may be contributing to a higher prevalence of mild and moderate periodontal disease. Participants offered four main reasons why oral hygiene is carried out; these were reward, symptoms, fear, and routine. This is supported by other research investigating the psychosocial determinants influencing oral hygiene practices which has found that children need adults to remind them and provide routine for toothbrushing (Gill et al., 2011) and parents need high self-efficacy in order to set up good toothbrushing routines in their children (Huebner and Riedy, 2010). Parents who are worried about causing bleeding when brushing may have less self-efficacy, and joint bleeds may disrupt routine. Visiting the dentist more often may remove barriers and increase enablers to performing oral hygiene (Syrjala et al., 2001; Syrjala et al., 1992a) and this may be an important tool. Indeed, Kabil et al showed an improvement in oral hygiene after eight months of a targeted prevention programme in children with haemophilia (Kabil et al., 2007). This suggests that additional dental visits (including for the parents of children with haemophilia) would be beneficial in improving oral hygiene.
4.7.2 Dental visiting habits

4.7.2.1 Dental visiting habits in adults

Figure 24 compares the visiting habits of the sample and the New Zealand population. The sample were more likely to visit a dentist and more likely to attend for a check-up than the general population.

Figure 24 - Comparison of visiting habits of adults

Regularity of dental visiting by those with haemophilia was investigated in Turkey, but no statistically significant difference was found between the study group and the controls (Alpkilic Baskirt et al., 2009). In the UK, 94% of children and 71% of adults with bleeding disorders reported receiving regular dental care (Fiske et al., 2000). Another UK study found that 68% of those with bleeding disorders had visited a dentist in the previous year (Kalsi et al., 2011). These estimates are higher than those reported for the general UK population (Donaldson et al., 2008) suggesting that either oral care
is prioritised or barriers to care are reduced for these individuals within the UK health-care system.

Nearly half of adult participants in this study had not visited a dental professional in the previous year, although the WHF guidelines (Scully et al., 2008) recommend yearly visits at a minimum (acknowledging their special needs); this is clearly a target which is not yet being met by this group. These data are unlikely to be the result of participants giving desirable answers because they had already given undesirable answers to questions about the frequency of oral hygiene practices. Moreover, it is not likely to be due to disinterest, because two out of five participants expressed a belief that they should attend more often.

4.7.2.2 Dental visiting habits of children

Five out of six children (85.7%) of children had visited a dental professional in the previous year. This is very similar to the NZOHS results (81.2%). When broken down into age groups, both samples display a similar pattern, with those aged 5-11 most likely to have visited a dental professional, and the lowest attendance rate in preschoolers. This is likely to be due to the universal nature of the school dental service as more children visit a dental professional than adults.

4.7.2.3 Barriers to dental visits

Dental visiting has been shown to confer oral health benefits (Thomson et al., 2010) yet individuals with haemophilia in this study are not visiting the dentist as often as recommended. Other authors have looked at barriers to dental visiting, both in general populations and in haemophilia populations, in different countries. The wide variety of reasons identified are listed in Table 21.
Table 21 - Barriers to dental visiting identified by different authors

<table>
<thead>
<tr>
<th>Barrier identified</th>
<th>Haemophilia population</th>
<th>General population</th>
</tr>
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<tbody>
<tr>
<td>Lack of confidence in</td>
<td>(Park and York, 2008)</td>
<td>(Fiske et al., 2000)</td>
</tr>
<tr>
<td>health professional</td>
<td></td>
<td>(Kalsi et al., 2011)</td>
</tr>
<tr>
<td>Fear of bleeding</td>
<td>(Alpkilic Baskirt et al., 2009)</td>
<td></td>
</tr>
<tr>
<td>Stigma of HIV/hepatitis</td>
<td>(Fiske et al., 2000)</td>
<td></td>
</tr>
<tr>
<td>Dental anxiety/fear</td>
<td>(Fiske et al., 2000)</td>
<td>(Finch et al., 1988)</td>
</tr>
<tr>
<td>Difficulty of access</td>
<td>(Kalsi et al., 2011)</td>
<td>(Finch et al., 1988)</td>
</tr>
<tr>
<td>Time</td>
<td>(Kalsi et al., 2011)</td>
<td></td>
</tr>
<tr>
<td>Lack of dental</td>
<td>(Finch et al., 1988)</td>
<td>(Syrjala et al., 1992b)</td>
</tr>
<tr>
<td>awareness</td>
<td>(Syrjala et al., 1992b)</td>
<td>(Ministry of Health, 2010b)</td>
</tr>
<tr>
<td>Cost</td>
<td>(Finch et al., 1988)</td>
<td>(Syrjala et al., 1992b)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>(Ministry of Health, 2010b)</td>
</tr>
<tr>
<td>Laziness</td>
<td></td>
<td>(Syrjala et al., 1992b)</td>
</tr>
<tr>
<td>No problems/no need</td>
<td></td>
<td>(Ministry of Health, 2010b)</td>
</tr>
</tbody>
</table>

There is a small amount of overlap but, generally, the perceived barriers to dental visiting are different for the two groups. Dental anxiety is one barrier experienced by both groups and this study found similar proportions experiencing dental anxiety as in the general New Zealand population (page 94).

Adult participants in this study could potentially be asked about barriers to dental visiting in three different ways: in the questionnaire those who thought they did not attend often enough were asked why not and their answers recorded in predetermined categories; those who had not attended in the previous year were asked why not and their answers, again, placed in predetermined categories; and, all participants had the opportunity to discuss barriers to visiting an oral health professional in the interview. These led to different results (Figure 7) which demonstrates the context specific nature of the data.

Those who were asked generally why they did not attend often enough responded with only three answers: ‘no problems’, ‘cost’, and ‘access’ – all barriers identified by those
authors who looked at barriers in the general population (including the NZOHS). These reasons are not supported by other data collected in this study.

Those who were asked more specifically why they did not attend in the previous year gave a much wider range of answers, which differed in several respects to those given to the same question asked in the NZOHS (Figure 25). Those with haemophilia were more likely to respond with ‘no teeth’, ‘too busy’ or ‘not important’ and less likely to respond with ‘cost’, or ‘no problems’. These responses align much better with the clinical and qualitative data.

![Figure 25 - Comparison of reported reasons for not visiting dentist in previous year (adults)](image)

When participants discussed why they (or people like them) did not visit the dentist many more themes were identified. These fell into the broad categories of extrinsic factors (time and cost); intrinsic values or perceptions (fear, additional medical interventions, need not perceived, and laziness); and reasons related to their haemophilia (joint bleeds and unwilling dentist), illustrating again, the complex nature of this subject.
4.7.3 The setting for dental visits

Some differences in the reasons given maybe explained by the difference in how those with and without haemophilia access dental care. In New Zealand, the majority of dental care for adults is provided through private practitioners. Hospital dental services must be provided by each District Health Board for medically compromised individuals who cannot be seen in general dental practice. These hospital dental services are provided at a highly subsidized cost to the individual. The majority of respondents in this study received their dental care from such a hospital service based in Wellington Hospital. This is in keeping with the Haemophilia Foundation of New Zealand Needs Assessment which found that ‘people who lived close to a hospital with a haematology centre..... tended to use hospital dental services exclusively for their dental needs’ (Haemophilia Foundation of New Zealand Inc., 2009). It also aligns with the national recommendation that people with haemophilia receive specialist dental care from comprehensive, hospital-based, care centres (Medical Advisory Committee of the Haemophilia Foundation of New Zealand, 2005).

When asked, as part of the questionnaire, to give the reason for attending at the hospital respondents’ most popular response was that it provided the specialist services required. When this issue was investigated more deeply in interviews, three main reasons appeared: it was more streamlined, safer, and cheaper.

4.7.3.1 Streamlining treatment

Streamlining of treatment is important to overcome the identified barrier of additional medical interventions. Individuals with haemophilia already feel the burden of medical interventions, either as part of home-care, or within a hospital (Park et al., 1995). Dental procedures may require additional haemostatic treatment or even a hospital stay (Brewer and Correa, 2006) and Fiske suggested that there is a reluctance to undertake this additional treatment (Fiske et al., 2002). Many participants in this study spoke about the additional procedures involved in receiving dental care as an individual with haemophilia. A recent haemophilia needs assessment found that ‘those in major centres felt [informing practitioners outside haematology teams about haemophilia] was less of an issue as the majority of their care was administered through the hospital, including
dental care’ (Haemophilia Foundation of New Zealand Inc., 2009) indicating that the use of hospital services makes receiving this dental care easier for these individuals.

4.7.3.2 Safer treatment

In the UK, 45% of those with bleeding disorders did not feel confident in the GDPs’ ability to treat those with bleeding disorders (Kalsi et al., 2011). This lack of confidence may be justified as the same study found some UK GDPs would be happy to carry out extractions in general practice, which is possibly not appropriate (Brewer and Correa, 2006). Dental treatment provided within the hospital was generally perceived as safer by study participants because the dentists were perceived to be more experienced with haemophilia, and the haematology department was closer.

4.7.3.3 Cost of dental care

The majority of dental care for adults in New Zealand is provided within the private sector, with all costs being met by the patient. The cost of dental care has been suggested as a potential barrier to receiving dental care. Petelo et al (2004) found one-third (33.9%) of Pacific Islanders living in Christchurch had not visited a dentist in the previous year due to the cost and more than one in five respondents (22.8%) in a New Zealand health survey had deferred seeing the dentist at least once in the previous twelve months due to cost (Jatrana and Crampton, 2009). Some authors have argued that cost is over-emphasised as a barrier to receiving dental care (Dixon et al., 1999; Grytten et al., 1996), but a large-scale experimental study manipulating the cost of dentistry has shown that the removal or reduction of costs associated with dental care can influence decisions to seek or access dental care (Keeler and Rolph, 1988; Manning et al., 1985).

When considering the study findings, there seems to be a difference between the perception of the cost of dentistry as a general issue (considered to be a problem) and the cost of dentistry to the participant as an individual (not really an issue). This is seen in both the qualitative and the quantitative findings (page 59).
While half of respondents said they would visit more if the cost was lower, cost was rarely reported as a reason for not attending in the previous year (Figure 25). Figure 26 shows a comparison of treatment avoidance due to cost in the sample and the NZOHS. Far fewer study participants reported avoiding visiting a dental professional or refusing dental treatment due to the cost than NZOHS respondents.

![Figure 26 - Comparison of avoidance of dental care due to cost](image)

It could be assumed that, for these adults, the ability to access dental care through Wellington Hospital, where treatment is highly subsidised, reduces costs. In reality, of the thirteen participants who had spent money on dental care in the previous twelve months, the average spend in private practice was $250 (two participants) and the average spend in the hospital was $312 (eleven participants). No data is available about what treatment these patients received for their money but the perception of hospital dental care incurring lower costs remains.

### 4.7.4 Dental awareness

Dental awareness is an important precursor to oral health behaviours. Gregory et al, after qualitatively investigating the relevance of dental health to attenders and non-attenders, concluded that many factors, such as beliefs, perceptions, and judgements, create a ‘margin of relevance’ around oral health, oral disease, and oral care providers
(Gregory et al., 2007). If this margin is small then oral health is not considered relevant and it becomes a low priority.

Participants in this study were asked about the importance of oral health (in the questionnaire) and invited to discuss the importance of oral health (in the semi-structured interview). The majority responded that oral health was important (Table 19) and were able describe why it was important; only one person vocalised that oral health was not important in the context of general health. However, when asked about how government money should be spent, most thought most other areas of healthcare were more important than dental health (except for non-urgent surgery) (Figure 17). In addition, low priority was identified from the qualitative data as a barrier to both dental visiting and oral hygiene practices. This may indicate that in reality oral health is not as important as participants say it is. Indeed, participants in this study were more likely to use ‘not important’ as a reason for not going to the dentist in the previous year than NZOHS respondents (Figure 25).

Kabil et al (2007) postulated a lack of awareness of the importance of dental care as a reason for poor oral health in those with haemophilia (Kabil et al., 2007). The World Hemophilia Federation guidelines emphasise the importance of preventive dental care (Brewer and Correa, 2006; Scully et al., 2008). However, haemophilia treatment guidelines in New Zealand do not, unfortunately, mention preventive dental care (Medical Advisory Committee of the Haemophilia Foundation of New Zealand, 2005) and the New Zealand Haemophilia Foundation website\(^\text{11}\) contains no advice or information about dental care.

Therefore, dentistry does not have a high profile within the sector in New Zealand and it is difficult to see why those with haemophilia should think dental care and prevention is important if the haemophilia community as a whole in New Zealand does not. However, it must be acknowledged that one of the factors in achieving a high participation rate in this study was the input of the local haemophilia nurse who has a great interest in oral health.

\(^\text{11}\) [www.haemophilia.org.nz](http://www.haemophilia.org.nz) accessed 31/10/11
4.7.5 Access to dental care

Access to dental care is not always about geographical location or distance. Twenty percent of those with bleeding disorders in the UK have experienced being refused treatment by a dentist (Kalsi et al., 2011); the same percentage as in this study. Just as patients with haemophilia are not always confident in general dentists, general dentists have also been shown to be reluctant to treat patients with haemophilia (Fiske et al., 2000; Kalsi et al., 2011; Parry and Khan, 2000; Roberts et al., 1978), probably due to the risk of bleeding.

Dentists have also been shown to be reluctant to treat patients with blood-borne viruses (Robinson et al., 1994). This may be relevant because untreated early blood products in New Zealand led to 28 people with haemophilia contracting HIV and the majority of those with severe haemophilia contracting Hepatitis C (Harper et al., 2003). Those with haemophilia have reported feeling stigmatised by blood-borne viruses, even if they themselves do not carry any (Barlow et al., 2007; Park et al., 1995). Fortunately, this issue was not identified in this study because withdrawal of care for this reason would be contrary to the New Zealand Dental Association Code of Practice (New Zealand Dental Association, 2007).

Lack of access was not identified as an issue in this study, probably because all participants are eligible to attend the hospital dental service and the majority do so.

4.7.6 Time required for dental care

The time taken to get to a dental appointment is commonly used as a proxy for access. Dixon et al investigated dental visits in the West Coast, where travel time is longer due to the remoteness. They found that geographical location had no effect on dental visiting (Dixon et al., 1999).
Those with haemophilia may have similar problems with increased distances to access a dentist because the majority of them are seen in a hospital setting. Living further away from the hospital was associated with higher caries experience and significantly associated with being edentulous. However, although four out of ten respondents travelled more than half an hour to get to their dental appointment, very few felt that this was too long to travel. It may be accepted that travel to a specialist centre is a just part of having haemophilia.

But then we were also told when he was born that we should always consider living near a main centre, because of his haemophilia. [2]

Respondents were much more concerned with the need to take time away from work. This may be due to the lack of pay when time is taken off work or due to having to take other time off work due to their haemophilia and the fear of becoming a burden or losing their job

So with work, [and going to dental appointments] I mean, luckily I work in a place that understands about this kind of thing, but work is a problem, I mean, you’ve got to work [11]

If you’re having time off work regular to have your teeth done, you know you’ve got to go one day this week and one day next week and of course the boss gets a bit uppity about it. Some bosses do. [14]

Barlow et al found that respondents reported a negative impact of haemophilia on their work life due to bleeds, time taken off, and fear of their colleagues viewing them unfairly (Barlow et al., 2007). PWH may not want to worsen this prejudice by taking additional time off for dental appointments. In the UK, Kalsi et al found that 30% of those with inherited bleeding disorders had not visited in the previous year due to a lack of time (Kalsi et al., 2011).
Nineteen percent of adult participants had taken time off during the previous year to attend the dentist. This is higher than the 11.8% of men in the NZOHS who had done the same; this may reflect the more limited opening hours that hospital dental services have than private dentists. This is an issue that may need to be investigated further if dental visiting within the hospital sector is to be increased.

4.7.7 Joint bleeds

Despite the use of prophylactic treatment, repeated bleeding into joints still occurs, and can lead to pain and lack of mobility (Park et al., 1995). Stairs, lack of nearby parking, and policies about late cancellations of appointments will all impact on the ability of those with joint problems to visit the dentist. This was a real issue for some participants who frequently had to cancel appointments, which may affect the dentist-patient relationship.

Severity of haemophilia was not associated with any difference in oral health. This may indicate that haemophilia per se does not affect oral health or that those with more severe haemophilia take prophylactic Factor so differences due to bleeding are minimised.

4.7.8 Other factors

Grytten et al concluded that getting a reminder from the dentist as a prompt to go was more important a factor in actually attending the dentist than any other factor, even a cost subsidy (Grytten et al., 1996). This did not emerge as a theme in this study, although one individual stated during the questionnaire that he had not attended the dentist in the previous year because his reminder had gone to the wrong address.
4.8 Model of oral health facilitation

Utilizing the data discussed previously, a model is proposed in Figure 27 for the sample who appear to have better oral health than the general population and visit a dentist more often, usually through the hospital dental service.
Figure 27 - Model of oral health facilitation in the sample
4.9 Access to hospital dental services

Figure 27 suggests that the underlying reason for better oral health in the sample is the widespread access to hospital dental services for those with haemophilia. This is plausible because it has also been suggested that the decrease in inequalities in oral health seen in 5-11-year-olds in New Zealand is due to universal access to dental care (National Advisory Committee on Health and Disability, 2003). Indeed, any difference in oral health was harder to identify for this age group than for adults in this study.

This model is also supported by other data from this study. This population is less likely to brush teeth than the general population but has less caries. This suggests that other factors that prevent caries are present. Non-invasive prevention interventions such as fluoride applications or fissure sealants are more likely to be placed in patients who visit the dentist. In addition, the data also show that the mean DMFT is higher in those participants who live further away from the hospital.

Regular dental attendance can improve oral health by allowing prevention to be targeted. As caries distribution changes, methods of prevention should change, and, because caries is seen in smaller subgroups of the population, preventive strategies should be targeted to these high-risk groups in order to prevent diminishing returns (Burt, 1998).

The dental treatment of those with haemophilia is easier than in the past because the availability of recombinant factor allows dental treatment to be carried out easily and relatively safely (‘it takes the haemophilia away’). However, for those who have developed inhibitors, dental treatment still comes with risks (Brewer, 2008). The treatment of those with haemophilia can also incur other costs. In 2011, 26 individuals with haemophilia attended a total of 76 dental appointments within CCDHB dental service. The cost of additional Factor to cover treatment required at these out-patient appointments was just over $57,000 (spread over 15 appointments)\textsuperscript{12}. This additional

\textsuperscript{12} Personal communication
cost does not include the cost of hospital admission of two patients for monitoring after dental extractions.

With this in mind then, it is appropriate to consider those with haemophilia a high risk group, not because of any greater risk of caries, but because of the greater risk and cost of the consequences of oral disease.

Participants were generally supportive of the hospital dental service, feeling that it met their needs better than a private practitioner might. However, while three-quarters of all respondents had made their last dental visit to a hospital department, only sixty percent would prefer to attend the hospital department. It may be that those with haemophilia are being forced into the hospital dental system by haematology health professionals and a reluctance by private dentists to provide treatment. There are only six haemophilia centres in New Zealand, and these may not be easily accessible for all, possibly leaving some individuals unsure about how to access dental care.

This is not the case in all countries; in the UK, those with bleeding disorders see a GDP for care and are subsequently referred to hospital or community dental service for specific treatment (Fiske et al., 2000). The WHF dental treatment guidelines do not recommend all treatment needs to be carried out in hospital but suggest that dental surgery, extractions, inferior dental blocks, and lingual infiltrations often require prophylaxis and as such may be more appropriately performed in the hospital setting (Brewer and Correa, 2006). Table 22 compares the usual setting of dental visit between respondents in this study and those with bleeding disorders in the UK.

<table>
<thead>
<tr>
<th>Table 22 - Comparison of usual setting of dental visit between those with haemophilia in the UK and this study</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Sample</strong></td>
</tr>
<tr>
<td>Usually see private dentist</td>
</tr>
<tr>
<td>Usually see hospital dentist</td>
</tr>
<tr>
<td>Usually see other provider</td>
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</tbody>
</table>

*(Kalsi et al., 2011)*
It must be remembered that, in the UK, NHS dentistry is subsidised, and free for those on a low income, meaning that cost may be much less of an issue there; it was not mentioned as barrier at all in Kalsi’s study of barriers to dental care for those with bleeding disorders (Kalsi et al., 2011).

While hospital services are perceived as cheaper in New Zealand, the use of hospital dental services comes with opportunity costs to the dental service. Interestingly, while acknowledging that oral health is important and that cost is an issue, participants did not feel that oral care should be subsidised for everyone but that it should be prioritised, mostly to those on a low income. A few participants felt that those with severe health issues should also received subsidized care but it is not clear if they count themselves in category (Figure 9).

No real cost-benefit analysis has been carried out to determine whether the greater costs associated with regular hospital dental service visits are outweighed by the lower costs of providing additional clotting factor to facilitate dental treatment when needed, but one participant was in no doubt that providing preventive dental care for those with haemophilia benefited the country as a whole.

And from a, from a kind of wider population point of view it does have an impact on everybody from sort of the pragmatic thing [because] haemophilia is so expensive to treat that by making the case that, you know, by treating this population really well you’re actually making huge savings over here for other, you know, a whole bunch of people could have elective surgery if you cut this, you know. [11]

4.10 Generalisability of findings

This study covered only those individuals with haemophilia who resided in the region covered by three adjacent District Health Boards. It was found that these individuals
probably had better oral health than those without haemophilia in the general population and a model explaining this has been suggested.

No national database of those with haemophilia is kept in New Zealand. Limited demographic data are compiled nationally as part of a project analysing the usage of clotting factors across New Zealand. However, the data are incomplete and were unavailable for comparison with this study.

The CCDHB allows those with haemophilia using the haemophilia centre in Wellington to have regular dental examinations within the hospital. Regular dental attendance has been shown to influence oral health in other populations (Thomson et al., 2010) and this is postulated to be the main driver to better oral health in this sample. This is not the case at all DHBs, with some accepting only referrals for extractions and assuming that all other treatment is being accessed through private GDPs\textsuperscript{13}, much like the UK model. This may result in oral health in these individuals being worse because they would not have the benefit of receiving what is perceived to be streamlined, safe and cheap dental care, thus creating barriers.

A much larger study incorporating other areas in New Zealand, including those DHBs where access to dental care is less readily available, would be needed to confirm these findings.

\textsuperscript{13} Personal communication
5 Conclusions

In order to help improve the provision of oral healthcare to individuals with haemophilia, the aims of this research were to:

- describe the oral health and oral health behaviours of those with haemophilia;
- compare these findings with estimates from the New Zealand population;
- explore the perceptions of oral health and its importance expressed by those with haemophilia; and
- identify the barriers to oral health perceived by those with haemophilia.

Data were collected through different methods and combined to give a picture that supports the following conclusions.

1. The decision-making process to render edentulous an individual with haemophilia is different to that for individuals without haemophilia.

2. Poor oral health has more impact on those with haemophilia.

3. Those with haemophilia have more mild periodontal disease, which may be related to a reluctance to perform oral hygiene, especially flossing.

4. Access to hospital dentistry leads to better oral health (less caries experience and lower prevalence of severe periodontal disease) and a model has been proposed to show how this interaction occurs.
5.1 Recommendations for further research

This study should be repeated on a larger scale, incorporating those DHBs where access to hospital dental care is less readily available. This should support the hypothesis that access to hospital dental care in this population improves oral health.

A cost-benefit analysis should be carried out to consider the greater costs of providing widespread hospital preventive dental services to this population (assuming this leads to lower treatment need) versus the cost of the additional clotting factor and hospital admission required to cover the increased treatment need if this preventive dental care is not provided. This will determine whether hospital dental service provision to those with haemophilia can be justified within a publicly-funded health care system.
References


Brewer A (2008). Dental management of patients with inhibitors to Factor VIII or Factor IX. Quebec: World Federation of Hemophilia.


Cutress TW, Hunter PB, Davis PB, Beck JD, Croxson LJ (1979). Adult oral health and attitudes to dentistry in New Zealand 1976: An epidemiological survey of adult oral conditions and treatment needs, oral behaviour, and attitudes to oral health, disease, and services. Wellington: Dental Research Unit, Medical Research Council of New Zealand.


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Appendix 1 - Letter of approval from Central Region Ethics committee
9 August 2010

Professor W Murray Thomson
Dept of Oral Sciences
School of Dentistry
University of Otago
PO Box 647, Dunedin

Dear Professor Thomson

Re: Ethics ref: CEN/10/05/015 (please quote in all correspondence)
Study title: The Oral Health of Individuals with Haemophilia: a Mixed Methods Investigation
Investigators: Professor W Murray Thomson

This study was given ethical approval by the Central Regional Ethics Committee on 9th August 2010. A list of members of the Committee is attached.

Approved Documents
- Information sheet for adult participants version 3
- Information sheet for participants aged 15 years version 3
- Information sheet for participants aged 14 years and under version 3
- Information sheet for parents of participants version 3
- Consent form for adult participants
- Consent form for participants aged 15 years version 2
- Consent form for participants aged 14 years and under
- Consent form for parents of participants
- Oral health of individuals with haemophilia – adult questionnaire
- Oral health of individuals with haemophilia – child questionnaire

This approval is valid until the 1st of October 2011, provided that Annual Progress Reports are submitted (see below).

Amendments and Protocol Deviations
All significant amendments to this proposal must receive prior approval from the Committee. Significant amendments include (but are not limited to) changes to:
- the researcher responsible for the conduct of the study at a study site
- the addition of an extra study site
- the design or duration of the study
- the method of recruitment
— information sheets and informed consent procedures.

Significant deviations from the approved protocol must be reported to the Committee as soon as possible.

**Annual Progress Reports and Final Reports**
The first Annual Progress Report for this study is due to the Committee by 9 August 2011. The Annual Report Form that should be used is available at www.ethicscommittees.health.govt.nz. Please note that if you do not provide a progress report by this date, ethical approval may be withdrawn.

A Final Report is also required at the conclusion of the study. The Final Report Form is also available at www.ethicscommittees.health.govt.nz.

We wish you all the best with your study.

Yours sincerely

[Signature]

Jacqi Bartlett
Administrator
Central Regional Ethics Committee
Email: jacqibartlett@moh.govt.nz
Appendix 2 – Letter of approval from Research Advisory Group – Maori (RAG-M)
16 June 2010

Dr Elizabeth Hitchings
Dental and oral health department
Wellington Regional Hospital
Private Bag 7902
Wellington South

RAG-M 2010/89 — Letter of Endorsement

Tena koe Dr Hitchings

On behalf of the Research Advisory Group Maori I write in relation to your study entitled The oral health of haemophiliacs: a mixed methods investigation.

You have supplied a RAG-M cover sheet, ethics application and questionnaires, information sheets and consent forms.

Our reading of your proposal characterises the research as:

- A mixed method study of the oral health of people with moderate and severe haemophilia identified from a register of patients with inherited bleeding disorders living in the lower North Island.
- A study involving a questionnaire, a clinical examination conducted at a dental surgery at Wellington Hospital, and a semi-structured interview conducted with a subsample.
- A study involving around 45 children and adults of whom approximately 13 may be Maori.

We note that you will are seeking approval from the Central Ethics Committee and that you are registering the study with the Clinical Research Centre.
Given the possibility that Maori patients may be participants, we would expect that you would:
- Ensure any Maori who participates is well informed and supported, including their whanau as appropriate;
- Recognise any cultural expectation and seek to meet these expectations responsibly;
- Report any results on Maori participants in a culturally safe manner.

We have notification from Whanau Care Services that you have a support agreement with them.

We would also expect the results of this project to be disseminated appropriately to Maori communities, as a way of raising awareness about eye disease and treatment options.

Given the importance of oral health to Maori, and the potentially higher barriers to care for Maori with haemophilia, we hope that this study will contribute towards better outcomes for Maori patients and we wish you well in your study.

On confirmation that the expectations specified above are understood and accepted by you we will be happy to endorse your research proposal. Please confirm these details with the RAG-M secretary, Ms Vanessa Mili by email to raqm@ccdhb.org.nz.

Upon completion of your study, where relevant it would be useful if a copy of your findings was sent to RAG-M.

Naku noa na

Jack Rikihana
Chair RAG-M
Appendix 3 – Letter of approval from Ngai Tahu
Ngāi Tahu Research Consultation Committee
Te Komiti Rakahau ki Kai Tahu

23/02/2010 - 37
Tuesday, 23 February 2010

Professor Thomson
Oral Sciences
Dunedin

Tēnā koe Professor Thomson

Title: The oral health of haemophiliacs: a mixed methods investigation.

The Ngāi Tahu Research Consultation Committee (The Committee) met on Tuesday, 23 February 2010 to discuss your research proposition.

By way of introduction, this response from the Committee is provided as part of the Memorandum of Understanding between Te Rūnanga o Ngāi Tahu and the University. In the statement of principles of the memorandum, it states "Ngāi Tahu acknowledges that the consultation process outlined in this policy provides no power of veto by Ngāi Tahu to research undertaken at the University of Otago". As such, this response is not "approval" or "mandate" for the research, rather it is a mandated response from a Ngāi Tahu appointed committee. This process is part of a number of requirements for researchers to undertake and does not cover other issues relating to ethics, including methodology; they are separate requirements with other committees, for example the Human Ethics Committee, etc.

Within the context of the Policy for Research Consultation with Māori, Consultation is defined according to the definition of Justice McGechan:

"Consultation does not mean negotiation or agreement. It means: setting out a proposal not fully decided upon; adequately informing a party about relevant information upon which the proposal is based; listening to what the others have to say with an open mind (in that there is room to be persuaded against the proposal); undertaking that task in a genuine and not cosmetic manner. Reaching a decision that may or may not alter the original proposal."

The Committee considers the research to be of importance to Māori health.

As this study involves human participants, the Committee strongly encourage that ethnicity data be collected as part of the research project. That is the questions on self-identified ethnicity and descent, these questions are contained in the 2006 census.

The Committee notes the researchers have identified that, "Approximately one-third of haemophiliacs in the Wellington region are of Māori descent", and ask what the reference for this statistic is.

The Ngāi Tahu Research Consultation Committee has membership from:
Te Rūnanga o Oākāra Incorporated
Katiki Haurapa Rūnanga ki Puketaraki
Te Rūnanga o Moeraki
NGĀI TAIU RESEARCH CONSULTATION COMMITTEE
TE KOMITI RAKAHAU KI KĀI TAHU

The Committee suggests dissemination of the findings to relevant Māori health organisations, for example the National Māori Organisation for Dental Health, Oranga Niho and to Associate Professor John Broughton, who is involved in Māori Dental Health, University of Otago.

We wish you every success in your research and the Committee also requests a copy of the research findings.

The recommendations and suggestions above are provided on your proposal submitted through the consultation website process. These recommendations and suggestions do not necessarily relate to ethical issues with the research, including methodology. Other committees may also provide feedback in these areas.

Nāhaku noa, nā

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Facilitator Research Māori
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Web: www.otago.ac.nz

The Ngāi Tahu Research Consultation Committee has membership from:
Te Rūnanga o Otākou Incorporated
Kāti Huirapa Rānake ki Poutereaki
Te Rūnanga o Moeraki
Appendix 4 - Letter of support from the Haemophilia Foundation of New Zealand
Tuesday 30th March 2010

Liz Hitchings
52a Connaught Terrace,
Brooklyn, Wellington 6021

22 March 2010

Dear Liz,

Thank you for the copy of your research protocol The oral health of haemophiliacs: a mixed methods investigation.

As the sole community agency providing specific education, care and support for people with haemophilia and other inherited bleeding disorders in New Zealand we are very supportive of research that promotes the health of people with haemophilia.

After considering your protocol, HFNZ would like to extend their support to your research into the oral health of people with haemophilia.

People with haemophilia or congenital bleeding tendencies are a priority group for dental and oral health care since bleeding after dental treatment may cause severe or even fatal complications. Maintenance of a healthy mouth and prevention of dental problems is of great importance, not only to quality of life but also to avoid the dangers of surgery and costs to healthcare systems of the clotting factor replacement products then required.

Please contact us if we may be of assistance with your research.

Kind regards

[Signature]

Belinda Burnett
Chief Executive Officer
Haemophilia Foundation of New Zealand Inc
Appendix 5 – Participant information sheets
The oral health of individuals with haemophilia

Information sheet for participants aged 14 years and under

Thank you for showing an interest in my project. Please read this information sheet carefully before deciding whether or not to take part. If you decide to participate I thank you. If you decide not to take part that is fine and I thank you for considering my request.

I am a dentist working at Wellington Hospital and I am doing this project as part of my study for the Masters in Community Dentistry degree from the University of Otago.

What is the aim of this project?

This project aims to describe the oral health (health of the mouth and teeth) of individuals with haemophilia and to identify possible reasons for any findings.

You have been invited to take part in this project as you have moderate or severe haemophilia and live in either the Wellington, Hutt or Waiarapa region.

What happens if I take part?

If you agree to take part in this project, three things will happen:

- Your parent or guardian will complete a questionnaire with me about your mouth and how you look after it. There are also some questions for you to answer. This should take about 45 minutes and can be done wherever your parent or guardian chooses.
- I will check your teeth in a dental surgery at Wellington Hospital. This will be just like when the dentist or dental therapist checks your teeth. The dental examination is free but there is no reimbursement for travel or other expenses.
- You and your parent or guardian will talk to me about what you think about your teeth, how you look after them and how easy this is. I will tape what we talk about.
Oral health of individuals with haemophilia

This will take about forty-five minutes and can be done wherever you or your parent or guardian choose.

You can choose to do all three parts of this project or just one or two parts.

All information collected as part of this study will be kept secret and I will not tell anyone that you have taken part unless you want me to.

You can choose to:

- Not take part,
- Refuse to answer any particular questions,
- Stop being part of the project at any time without giving a reason.

What happens next?

Please take time to talk about this project with your family as you will need the permission of your parent or guardian to take part. If you or your family have any questions about the project please contact me on the contact details below. If you and your parent or guardian agree to you taking part in this project, please ask them to contact me on the details below and we can arrange a time to start.

If you have any questions please contact:

Liz Hitchings
Dental and Oral Health Department
Wellington Hospital
04 918 6494
Liz.hitchings@ccdhb.org.nz
The oral health of individuals with haemophilia

Information sheet for participants aged 15 years

Thank you for showing an interest in this study. Please read this information sheet carefully before deciding whether or not to participate. If you decide to participate I thank you. If you decide not to take part there will be no disadvantage to you of any kind and I thank you for considering my request.

I am a dentist working at Wellington Hospital and I am doing this project as part of my Masters in Community Dentistry degree from the University of Otago.

What is the aim of this project?

This project aims to describe the oral health of individuals with haemophilia and to identify possible reasons for any findings.

You have been invited to participate in this study as you have moderate or severe haemophilia and live in the Wellington, Hutt or Waiarapa region.

What will participants be asked to do?

Should you agree to take part in this project, you will be asked to do three things.

- To complete a questionnaire about your mouth and how you look after it. This will be completed with me and should take about 45 minutes. This can be done at a place of your choosing.
- To undertake a dental examination, carried out by me. This will take place in a dental surgery at Wellington Hospital. The examination will be similar to that done by a dentist at a routine check-up; each tooth will be checked for decay and your gums will be checked for gum disease. No x-rays will be taken. The dental examination is free but there is no reimbursement for travel or other expenses. After the examination you
Oral health of individuals with haemophilia

will receive an oral health report, telling you what was found. Your haemophilia nurse specialist (BJ) may provide haemostatic cover before this examination.

- To participate in a taped interview with me about what you think about your mouth, how you look after it and how easy this is. This will take about forty-five minutes and can be done at a place of your choosing. You can choose to participate in all three parts or just one or two.

All information collected as part of this study will be kept confidential and anonymous.

You may arrange to have a support person present during any part of the project.

You have the right to:

- Not take part,
- Refuse to answer any particular questions,
- Withdraw from the project at any time without giving a reason.

What happens next?

Please take time to discuss this study with your family as you will need the permission of a parent or guardian to take part. If you or your family have any questions about the project please contact me on the contact details below. If you and your parent or guardian agree to you taking part in this study, please contact me on the details below and we can arrange a time to start.

If you have any questions please contact:

Liz Hitchings

Dental and Oral Health Department

Wellington Hospital

04 918 6494

Liz.hitchings@ccdhb.org.nz
The oral health of individuals with haemophilia

Information sheet for adult participants

Thank you for showing an interest in this study. Please read this information sheet carefully before deciding whether or not to participate. If you decide to participate I thank you. If you decide not to take part there will be no disadvantage to you of any kind and I thank you for considering my request.

I am a qualified dentist working for Capital and Coast DHB and this project is being undertaken as part of the requirements for the Masters in Community Dentistry degree from the University of Otago.

What is the aim of this project?

This project aims to describe the oral health of individuals with haemophilia and to identify possible reasons for any findings.

You have been invited to participate in this study as you have moderate or severe haemophilia and live in the Wellington, Hutt or Waiarapa region.

What will participants be asked to do?

Should you agree to take part in this project, you will be asked to do three things.

- To complete a questionnaire about your mouth and how you look after it. This will be completed with me and should take about 45 minutes. This can be done at a place of your choosing.
- To undertake a dental examination, carried out by me. This will take place in a dental surgery at Wellington Hospital. The examination will be similar to that done by a dentist at a routine check-up; each tooth will be checked for decay and your gums will be checked for gum disease. No x-rays will be taken. The dental examination is free but there is no reimbursement for travel or other expenses. After the examination you
Oral health of individuals with haemophilia

will receive an oral health report, telling you what was found. Your haemophilia nurse specialist (BJ) may provide haemostatic cover before this examination.

• To participate in a taped interview with me about what you think about your mouth, how you look after it and how easy this is. This will take about forty-five minutes and can be done at a place of your choosing. You can choose to participate in all three parts or just one or two.

All information collected as part of this study will be kept confidential and anonymous.

You may arrange to have a support person present during any part of the project.

You have the right to:

• Not take part,
• Refuse to answer any particular questions,
• Withdraw from the project at any time without giving a reason.

What happens next?

If you have any questions about the project please contact me on the contact details below. If you would like to take part in this project, please contact me on the details below and we can arrange a time to start.

If you have any questions please contact:

Liz Hitchings
Dental and Oral Health Department
Wellington Hospital
04 918 6494
Liz.hitchings@ccdhb.org.nz
The oral health of individuals with haemophilia

Information sheet for parents of participants

Thank you for showing an interest in this study. Please read this information sheet carefully before deciding whether or not to allow your child to take part. If you allow them to participate I thank you. If you choose not to allow them to take part there will be no disadvantage to you or them of any kind and I thank you for considering my request.

I am a qualified dentist working for Capital and Coast DHB and this study is being undertaken as part of the requirements for the Masters in Community Dentistry degree from the University of Otago.

What is the aim of this study?

This study aims to describe the oral health of individuals with haemophilia and to identify possible reasons for any findings.

Your child has been invited to participate in this study as he has moderate or severe haemophilia and you live in either the Wellington, Hutt or Waiarapa region.

What happens if I agree that my child can take part?

If you agree to your child taking part in this project, you and they will be asked to do three things:

- For you to complete a questionnaire with me about your child’s mouth and how it is looked after. This should take about 45 minutes and can be done at a place that you choose. If your child is aged 9 – 14 they will be asked some questions of their own as well.
- Your child will have a dental examination, carried out by me, in a dental surgery at Wellington Hospital. The examination will be similar to that done by the dentist or dental therapist at your child’s routine check-up. You can be present at this
Oral health of individuals with haemophilia

examination. After the examination you will receive an oral health report, telling you what was found. The examination is free, but there is no reimbursement for travel or other expenses.

- I will interview you and your child about what you think about your child’s mouth and teeth, how your child’s mouth is looked after and how easy this is. I would like to tape this interview. This will take about forty-five minutes and can be done wherever you choose.

You can choose to do all three parts of this project or just one or two parts.

All information collected as part of this study will be kept confidential and anonymous.

You or your child can choose to:

- Not take part,
- Refuse to answer any particular questions,
- Stop being part of the project at any time without giving a reason.

What happens next?

If you have any questions about the project please contact me on the contact details at the bottom of the page. If you agree to your child taking part in this project, please contact me on the details below and we can arrange a time to start.

If you have any questions please contact:

Liz Hitchings
Dental and Oral Health Department
Wellington Hospital
04 918 6494
Liz.hitchings@ccdhb.org.nz
Appendix 6 – Participant consent forms
The oral health of individuals with haemophilia

Consent form for participants aged 14 years and under

Request for an interpreter

<table>
<thead>
<tr>
<th>Language</th>
<th>I wish to have an interpreter</th>
<th>Yes</th>
<th>No</th>
</tr>
</thead>
<tbody>
<tr>
<td>English</td>
<td>I wish to have an interpreter</td>
<td>Yes</td>
<td>No</td>
</tr>
<tr>
<td>Deaf</td>
<td>E hiahia ana ahau ki tetahi kaiwhakaha Māori</td>
<td>Ae</td>
<td>Kao</td>
</tr>
<tr>
<td>Māori</td>
<td>Ka inangaro au i tetai tangata uri reo</td>
<td>Ae</td>
<td>Kare</td>
</tr>
<tr>
<td>Cook Island Māori</td>
<td>Au gadreva me dua e vakadewa vosa vei au</td>
<td>Io</td>
<td>Sega</td>
</tr>
<tr>
<td>Fijian</td>
<td>Fia manako au ke fakaaoga e taha tagata fakahokohoko kupu</td>
<td>E</td>
<td>Nakai</td>
</tr>
<tr>
<td>Niuean</td>
<td>Ou te mana’l ia i ai se fa’amatala upu</td>
<td>Ioe</td>
<td>Leai</td>
</tr>
<tr>
<td>Sāmoan</td>
<td>Ko au e fofo’u ki he tino ke fakalilu te gagana Peletania kin a gagana o na motu o te Pahefika</td>
<td>Ioe</td>
<td>Leai</td>
</tr>
<tr>
<td>Tongan</td>
<td>Oku ou fiema’u ha fakatoneulea</td>
<td>Io</td>
<td>Ikai</td>
</tr>
</tbody>
</table>

I have:

- Read and understand the information sheet about this project,
- Talked about this project with my parent(s) or guardian(s) and they have been able to ask any questions that they want to ask,
- Asked all the questions I want to about this project and they have all been answered.

I understand that:

- taking part in this study is voluntary (my choice),
- I can stop being part of the project at any time without giving a reason and that this will in no way affect my future or continuing healthcare,
- my participation in this study is confidential and no material that could identify me will be used in any reports on this study,
- I can ask for more information about the study at any time.
Oral health of individuals with haemophilia Consent form 14 years and under

I consent to participating in:

Oral health questionnaire (survey) Yes ☐ No ☐
Dental examination Yes ☐ No ☐
Interview Yes ☐ No ☐
I consent to my interview being audiotaped Yes ☐ No ☐

Name of participant Signature of participant date

I would like a copy of the results of this study when they are ready (I understand this may be several months) Yes ☐ No ☐

Please send to:

Parent or guardian of participant

I agree to my child or ward taking part in this study as indicated above. I agree to be present during all parts of the study (as below) and to answering questions about my child or ward’s oral health.

Oral health questionnaire (survey) Yes ☐ No ☐
Dental examination Yes ☐ No ☐
Interview Yes ☐ No ☐
I consent to my interview being audiotaped Yes ☐ No ☐

Name of parent/guardian Signature of parent/guardian date

14 years and under consent form April 2010 Version 1
The oral health of individuals with haemophilia

Consent form for participants aged 15 years

Request for an interpreter

<table>
<thead>
<tr>
<th>Language</th>
<th>Request for interpreter</th>
</tr>
</thead>
<tbody>
<tr>
<td>English</td>
<td>I wish to have an interpreter</td>
</tr>
<tr>
<td>Deaf</td>
<td>I wish to have a NZ sign language interpreter</td>
</tr>
<tr>
<td>Māori</td>
<td>E hiahia ana ahau ki tetahi kaiwhaka Māori</td>
</tr>
<tr>
<td>Cook Island</td>
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</tr>
<tr>
<td>Māori</td>
<td></td>
</tr>
<tr>
<td>Fijian</td>
<td>Au gadreva me dua e vakadewa vosa vei au</td>
</tr>
<tr>
<td>Niuean</td>
<td>Fia manako au ke fakaaga e taha tagata fakahokohoko kupu</td>
</tr>
<tr>
<td>Sāmoan</td>
<td>Ou te mana’l ia i ai se fa’amatala upu</td>
</tr>
<tr>
<td>Tokelaun</td>
<td>Ko au e fofo’u ki he tino ke fakalilu te gagana Peletania kin a gagana o na motu o te Pahefika</td>
</tr>
<tr>
<td>Tongan</td>
<td>Oku ou fiema’u ha fakatonulea</td>
</tr>
</tbody>
</table>

I have read and I understand the information sheet (dated July 2010) for volunteers taking part in the study designed to investigate the oral health of individuals with haemophilia. I have had the opportunity to discuss this study and all my questions have been answered to my satisfaction. I have had the opportunity to use whānau support or a friend to help me ask questions and understand the study.

I understand that:

- taking part in this study is voluntary (my choice),
- I am free to withdraw from the study at any time without giving a reason and that this will in no way affect my future or continuing healthcare,
- my participation in this study is confidential and no material that could identify me will be used in any reports on this study,
- I can ask for more information about the study at any time.
I consent to participating in:

Oral health questionnaire (survey)  
Yes ☐ No ☐

Dental examination (I understand that the regional haemophilia nurse specialist may administer haemostatic cover if appropriate)  
Yes ☐ No ☐

Interview  
Yes ☐ No ☐

I consent to my interview being audiotaped  
Yes ☐ No ☐

Name of participant  
Signature of participant  
date

I would like a copy of the results of this study when they are ready (I understand this may be several months)  
Yes ☐ No ☐

Please send to:

Parent or guardian of participant

I agree to my child or ward taking part in this study as indicated above.

Name of parent/guardian  
Signature of parent/guardian  
date
I have read and I understand the information sheet (dated July 2010) for volunteers taking part in the study designed to investigate the oral health of individuals with haemophilia. I have had the opportunity to discuss this study and all my questions have been answered to my satisfaction. I have had the opportunity to use whānau support or a friend to help me ask questions and understand the study.

**I understand that:**

- taking part in this study is voluntary (my choice),
- I am free to withdraw from the study at any time without giving a reason and that this will in no way affect my future or continuing healthcare,
- my participation in this study is confidential and no material that could identify me will be used in any reports on this study,
- I can ask for more information about the study at any time.
I consent to participating in:

Oral health questionnaire (survey) Yes □ No □

Dental examination (I understand that the regional haemophilia nurse specialist may administer haemostatic cover if appropriate) Yes □ No □

Interview Yes □ No □

I consent to my interview being audiotaped Yes □ No □

Name of participant

Signature of participant date

I would like a copy of the results of this study when they are ready (I understand this may be several months) Yes □ No □

Please send to:
The oral health of individuals with haemophilia

Consent form for parents of participants

Request for an interpreter

<table>
<thead>
<tr>
<th>Language</th>
<th>Interpretation Description</th>
<th>Yes</th>
<th>No</th>
</tr>
</thead>
<tbody>
<tr>
<td>English</td>
<td>I wish to have an interpreter</td>
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<td></td>
<td></td>
</tr>
<tr>
<td>Sāmoan</td>
<td>Ou te mana’l ia i ai se fa’amatala upu</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tokelauan</td>
<td>Ko au e fo’ou ki he tino ke fakalilu te gagana Peletania kin a gagana o na motu o te Pahefika</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tongan</td>
<td>Oku ou fiema’u ha fakatonulea</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

I have read and I understand the information sheet (dated July 2010) for volunteers taking part in the study designed to investigate the oral health of individuals with haemophilia. I have had the opportunity to discuss this study and all my questions have been answered to my satisfaction. I have had the opportunity to use whānau support or a friend to help me ask questions and understand the study.

I understand that:

- taking part in this study is voluntary (my choice),
- I am free to withdraw my child from the study at any time without giving a reason and that this will in no way affect his future or continuing healthcare,
- our participation in this study is confidential and no material that could identify me or my child will be used in any reports on this study,
- I can ask for more information about the study at any time.
I consent to participating in:

Oral health questionnaire (survey) about my child’s oral health
Interview
I consent to my interview being audiotaped
I consent to my child being dentally examined

Yes ☐ No ☐
Yes ☐ No ☐
Yes ☐ No ☐
Yes ☐ No ☐

Name of child

Name of parent/guardian Signature of parent/guardian date

I would like a copy of the results of this study when they are ready (I understand this may be several months)

Yes ☐ No ☐

Please send to:
Appendix 7 – Adult questionnaire

To be found on CD at back of book
Appendix 8 - Child questionnaire

To be found on CD at back of book
Appendix 9 – Interview template
Semi-structured interview with individuals with haemophilia

Interview purpose

- Determine attitude towards oral health
- Identify barriers to oral health behaviours

Introduction and welcome

As you know, this interview is part of research about the oral health of individuals with haemophilia and you have been invited to take part as you have haemophilia. This interview is intended to help me find out what you think about oral health, how oral health can be improved and what barriers there are to achieving oral health.

I will be taking notes, but I would also like to record this interview so that I don’t miss anything. Is that alright?

The first few questions will be about oral health and how a mouth can be made healthier. Then we will talk about how easy or difficult it is to improve oral health.

<table>
<thead>
<tr>
<th>Questions</th>
<th>Notes</th>
</tr>
</thead>
<tbody>
<tr>
<td>How would you rate the health of your own mouth? Why?</td>
<td></td>
</tr>
<tr>
<td>How would you tell if someone has a healthy mouth?</td>
<td></td>
</tr>
<tr>
<td>What about an unhealthy mouth?</td>
<td></td>
</tr>
<tr>
<td>Do you think the health of your mouth is important? Why?</td>
<td></td>
</tr>
<tr>
<td>What would you think about a person who has YY? (from question 2 – repeat for each YY)</td>
<td></td>
</tr>
</tbody>
</table>
If a friend or family member asked you how they could improve the health of their mouth, what would you tell them?

Is there anything else that someone could do to improve the health of their mouth?

Is there anything that you think you could do to improve the health of your mouth?

You said that XX would help improve the health of someone’s mouth. Is this something that you do? (repeat for each XX)

How easy is it to do XX?

How could doing XX be made easier?

What might make doing XX harder?

Is there anything else you would like to share with me about oral health, the things you can do to improve oral health or the problems in maintaining oral health?