An investigation into the understanding of basic genetic inheritance amongst amaXhosa caregivers of patients with haemophilia

by

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Declaration

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Map of the study area

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Abstract

This study sought to explore the level of understanding of basic genetic inheritance among isiXhosa-speaking caregivers of patients with the genetic bleeding disorder haemophilia. Haemophilia A and B are X-linked recessive inherited, lifelong bleeding disorders that are caused by deficiencies in blood clotting factors. The condition predominantly affects males, while females are carriers and usually unaffected. These bleeding disorders have a profound impact on the daily life of the affected individual, on carrier mothers and close family. Education is vital to enable women to appreciate the implications of being a carrier and to fully inform them and their partners of the implications for a prospective child. Socio-economic and language issues in South Africa are a major barrier to communication and an obstacle to good medical care for first-language Xhosa speakers. In order to provide culturally-sensitive, effective genetic counselling and to improve communication between health care service providers and first-language Xhosa speakers, it is important to explore the intrinsic knowledge and basic understanding of this cultural group. The study used an exploratory qualitative research design. Ten participants were recruited amongst first-language Xhosa speaking mothers or caregivers of patients with haemophilia residing in townships near Cape Town. Qualitative data were generated from transcribed and translated audio-records of ten semi-structured interviews, conducted in Xhosa by an interpreter as well as from participant observation notes by the investigator. Results suggest that the participants had a very limited understanding of the clinical management and cause of haemophilia. Information given by health care providers did not appear to be assimilated and participants remained unsure as to the implications of haemophilia. In spite of frequent visits to clinics there appeared to be limited understanding of the medical treatment and genetic consequences of haemophilia, which suggests that communication between health care providers and participants was inadequate. While treatment and care by health care service providers was fully accepted, several participants believe that traditional practices would provide more satisfactory explanations regarding the cause of the condition. Awareness by all role players of

different cultural beliefs and of how illness is interpreted by first-language Xhosa speakers might improve communication between health care service providers and isiXhosa speakers.

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Glossary of abbreviations and terms

AIDS	acquired immunodeficiency syndrome
Amniocentesis	a procedure for obtaining amniotic fluid from a pregnant woman, by inserting a hollow needle through the abdominal wall and into the amniotic sac
arthropathy	a pathology or abnormality of a joint
bilateral kinship system	inheritance from both maternal and paternal lines; both sides having equal status
C	Caregiver, e.g. C1 = Caregiver 1
cognition	the mental process of acquiring knowledge through reasoning, intuition, or perception
DNA	deoxyribonucleic acid
de novo mutation	new gene mutation
EC	Eastern Cape
exons	sequence of a gene's DNA that codes information for protein synthesis
exogamy	marriage to a person belonging to a tribe or group other than your own as required by custom or law
GSH	Groote Schuur Hospital
germline	pertaining to the cells from which gametes are derived. The cells of the germline bridge the gaps between generations.

HP	Haemophilia
haemarthrosis	bleeding into joint spaces
hemizygous	having only one copy of a chromosome rather than the usual two
HIV	human immunodeficiency virus
ICH	intracranial haemorrhage
kb	kilo base pair
Khaya ROCK	Khaya Reach Out Centre for Kids
matrilineal	tracing descent through the female line
morbidity	presence of illness or disease
mortality	death rate
NHLS	National Health Laboratory Services
P	Patient, e.g. P1 = Patient 1
patrilineal	tracing descent through the male line
pathogenesis	the origin of a disease and the chain of events leading to that diesease
RNA	ribonucleic acid
mRNA	messenger RNA
RCCMH	Red Cross Children's Memorial Hospital
SAHFMAC	South African haemophilia foundation medical advisory committee
Sangoma	traditional diviner and diagnostician who is believed to be in contact with ancestral spirits

synovitis	inflammation of the synovial membrane that lines a synovial joint; results in pain and swelling
township	peri-urban low-economic suburb comprising formal and informal housing and lacking infrastructure and other essential services. People living in the townships referred to in this study are almost all Black Africans whose first language is Xhosa
USA	United States of America
WHO	World Health Organisation

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Chapter 1: Introduction

1.1 Introduction

Haemophilia is a life-long chronic bleeding disorder caused by genetic defects that result in severe to mild bleeding and bruising. It is an X-linked recessive inherited disorder that has a profound impact on the daily life of the affected individual, carrier mothers and close family members (Goldstein and Kenet, 2002). Without treatment, haemophilia can lead to spontaneous internal bleeding or excessive bleeding following injuries or surgery.

Haemophilia A is the most common form of haemophilia and occurs in 1 in 5,000–10,000 male births, while haemophilia B occurs in 1 in 20,000–34,000. The worldwide incidence of Haemophilia A and B may, however, be even higher, because affected patients are not always diagnosed in developing countries (National Hemophilia Foundation [USA], 2009). In South Africa, for example, the total number of people with haemophilia on the South African Haemophilia Foundation register falls substantially short of the expected 1 in 10 000 number of cases (South African Government Information, 2009).

Patients with haemophilia in Cape Town have access to dedicated clinics at three tertiary referral hospitals, Groote Schuur Hospital (GSH), Red Cross War Memorial Children's Hospital (RCWMCH) and Tygerberg Hospital for diagnosis and management. Tygerberg hospital has adult and paediatric haemophilia care services in the same centre while GSH and the RCWMCH treat adult and paediatric haemophiliacs respectively. Treatment for confirmed or suspected acute bleeding events consists of intravenously administered factor VIII or IX concentrate. Patients will either be treated at the clinic or, if trained in home treatment, will be issued with factor concentrate by the clinic.

This study was undertaken in Cape Town amongst isiXhosa caregivers of patients with haemophilia. Cape Town is the capital of the Western Cape, where 29% of the inhabitants speak isiXhosa at home (City of Cape Town, 2009**).** Unfortunately, socio-

economic and language issues are still major barriers to communication in South Africa and are an obstacle to good medical care for isiXhosa speakers (Levin, 2005; Levin, 2006).

1.2 Motivation for research

The researcher was an observer in over 90 genetic counselling sessions in 2006, most of which were cross-cultural interactions. The counsellors were always White and either English or Afrikaans speaking, while the counselees where either Black African or of mixed ancestry. During these observations the researcher became aware of the problems associated with cross-cultural communication.

The counsellors were all well equipped with biological knowledge and the tools that allowed them to verify the biological system. They were, however, ill equipped with tools to verify what their clients' belief systems were and had no reference as to how this knowledge is imparted within, for example, the Xhosa culture. The counsellors generally used the western 'written, verbal and diagrammatic' approach to explain complex genetic inheritance and clients were 'spoken at' in terms of covering the counsellor's agenda. When asked for feed-back or to make a decision, clients were often fatalistic and preferred to leave the matter in God's hands. It was thus not always possible to get dynamic feed-back and the counsellors were ill equipped with 'cultural' or 'linguistic tools' to determine whether the client had understood or not.

These observations motivated the researcher to explore the baseline knowledge of genetic inheritance amongst isiXhosa speakers living in urban areas of the Western Cape in order to explore the possibility of developing effective linguistic tools as well as culturally sensitive explanatory models.

1.3 Significance of Study

Caregivers of children with haemophilia in South Africa and elsewhere feel restricted and insecure because of the physical vulnerability of the affected children. In addition, families often bear great financial and social burdens. Several studies have shown that effective genetic counselling is important and beneficial for families as it helps raise awareness of possible carrier status and provides options to prevent unwanted recurrence of the disease (Banis et al., 1999; Kelley and Narváez, 2006; Lin et al., 2008). A counsellor is a health care worker who helps equip patients and families with strategies to live with the problem. Genetic counsellors, amongst others, advise patients or relatives at risk of an inherited disorder of the consequences and nature of the disorder, the probability of developing or transmitting it, and the ways in which this may be prevented, avoided or ameliorated. Genetic testing has both benefits and limitations and the decision about whether to be tested is a personal and complex one. Counsellors can assist by providing information about the pros and cons of the test and discussing the social and emotional implications of testing. Ideally patients and/or relatives meet with a genetic counsellor before any genetic testing is done.

In order to improve their competence and effectiveness counsellors should be aware of their clients' knowledge base in connection with genetics and be able to recognize misconceptions and possible alternative beliefs. An understanding of the intrinsic knowledge and basic understanding of genetic inheritance is central to provide effective, culturally-sensitive counselling in order to fully inform carriers and their partners of the implications for a prospective child. Mahlangu (2009) in a report on haemophilia care in South Africa between 2004 and 2007 highlights that a general lack of genetic services in the country is currently the biggest challenge for patients and their families. currently there is only one functioning genetic unit in Johannesburg. The Cape Town unit is presently not equipped to regularly service the three haemophilia treatment centres in the area. Patients can, however, receive counselling on request from the Division of Human Genetics at the University of Cape Town.

It has been shown in the western world that a widespread lay knowledge of inheritance often conflicts with scientific explanations. Richards and Ponder (1996) propose that this lay knowledge is derived from concepts of kinship ties that are formed and sustained by everyday social activities and relationships. These concepts tend to be resistant to change and may impede the assimilation of scientific explanations (Richards and Ponder, 1996). They found that lay accounts of inheritance differ from Mendelian explanations and generally lack concepts of genotype and phenotype and the idea of a developmental process that links these. In some lay accounts there is mention of "genes", "DNA" or "chromosomes", but most do not specify how these may be related to inherited characteristics and there is generally no clear idea of what exactly is transmitted between generations. This, of course, makes explanation of gene expression and penetrance very difficult for a counsellor. A study by Chapple et al. (1995) similarly suggests that many lay people lack knowledge of genetics and inheritance, and that there is considerable stigma attached to genetic disease. Misconceptions and fears may not always be recognized or resolved during the process of genetic counselling and it is important, especially in the cross-cultural setting, that implications of such misunderstandings and beliefs are considered. While several studies have assessed lay understanding of inheritance in the general population in western countries, minimal published data are available on other cultures where kinship patterns differ (Richards, 1998). Port et al. (2008) report on the processes undertaken to provide a culturally-sensitive service for the Maori people of New Zealand. They found that most of the information generated to date on the interpretation of mutational analysis has been within a Eurocentric model, favouring individual autonomy. This western model of individual autonomy could not be integrated into the hierarchical tribal structure of the Maori which involves inherent rights of the collective. Similarly to the Maori, the amaXhosa are a population with an hierarchical tribal structure. Furthermore, as opposed to western cultures, the amaXhosa are a patrilineal society in which the line of kinship and descent is traced through the paternal line. A man and his brother, their children and their son's children are considered to be members of the same descent group.

Counsellors have the difficult task of interpreting complex genetic information to lay clients. Layton et al. (1993) have long been advocating that, especially with scientific explanations, it is important to take note of previous knowledge and to build on such knowledge. They call this approach from the "bottom up", while the traditional "top down" approach tends to begin with the scientific position and attempts to provide a simplified account of this, but is unlikely to achieve widespread scientific understanding. The problem is compounded amongst isiXhosa-speakers because of language problems as well as a lack of indigenous words describing terms like "genes" or "chromosome". Even if interpreters are available, they have difficulties in translating genetic concepts and tend to depend on words borrowed from English.

Awareness, by all role players, of cultural beliefs and of how illness is interpreted will help improve communication between health care service providers and isiXhosa-speakers. To date very little research has been published that explores the basic understanding of genetic inheritance amongst isiXhosa-speakers in the Western Cape.

1.4 Aims and Objectives

Aims

The aims of this research were to:

- explore the level of understanding of basic genetic concepts among Xhosa-speaking mothers or caregivers of patients with haemophilia.
- investigate the cultural background of the participants.
- explore the participant's understanding of genetic inheritance and its implications for genetic counselling.
- describe how caregivers of patients with haemophilia experience living with this condition.

Objectives

The specific objectives of this study were to explore the possibility of:

- developing strategies to improve the ability of isiXhosa-speaking families affected with genetic disorders to make informed decisions;
- improving the general genetic management of haemophilia families in South Africa;
- developing effective strategies for health care workers to explain genetic terminology to the Xhosa-speaking population of South Africa; and
- developing strategies for interpreters to effectively communicate genetic concepts to Xhosa-speaking individuals in South Africa.

1.5 Outline of research design and methodology

An exploratory qualitative research design was adopted. Participants included first-language Xhosa speaking mothers or caregivers of patients with haemophilia residing in townships near Cape Town. Qualitative data was generated from transcribed and translated audio-recordings of ten semi-structured interviews, conducted in a local day hospital or in the homes of the participants. The semi structured interview schedule allowed for the addition, exclusion or variation of the wording of particular questions, in response to the participants, albeit without changing the meaning of the questions (Murray and Rhodes, 2005; Pope and Mays, 2000).

Because the researcher was not familiar with isiXhosa or the cultural life of the study group, it was necessary to employ an interpreter to conduct the interviews. The interpreter was a first-language Xhosa speaker who was familiar with the culture and community in which the research was conducted. The researcher was present as an observer and gained valuable insights into Xhosa culture through personal experience and frequent discussions with the interpreter.

A phenomenological cross-sectional approach was used in order to gain a deeper understanding of how this sample of caregivers experienced their world and what it was like for them to take care of a child with haemophilia. Phenomenology comes from two Greek words: *phainomenon*, meaning ‘appearance’ and *logos*, which denotes ‘reason’.

According to Seymor and Clark (1998): 'Contemporary phenomenology revolves around the problem of how we make sense of the everyday world. It provides opportunities to tease out some of the concepts, ideas, frameworks and structures of meaning which are found in the day to day world of human interaction.'

1.5 Organisation of the study

Chapter 1: Introduction to the study

In this chapter the significance of the study, aims, perspective and motivation for methodology are discussed.

Chapter 2: Literature review

The literature review provides an overview of clinical aspects of Haemophilia A and B, the influence of language and culture in the clinical setting, the role of the interpreter in the cross-cultural consultation, lay understanding of genetics, the impact of language and cultural issues on multicultural counselling, and interviewing techniques in cross-cultural interviews.

Chapter 3: Methodology

Chapter three provides a description of the research design and setting and defines the study population. A detailed account is given of the process of data collection, the instruments used and of the processes of the study. Methods of data analysis are described and finally ethical considerations, trustworthiness and limitations of the study are discussed.

Chapter 4: Findings and discussion

In this chapter the findings of the data is presented and the findings are discussed. Findings are presented under main headings representing themes that emerged from the data. Each theme is introduced with a table that provides an overview of the categories, subcategories and codes that were formulated after thorough analysis of the data.

Chapter 5: Conclusions and recommendations

This chapter provides a summary of the findings and presents recommendations for future research.

Chapter 2: Literature Review

2.1 Introduction

A large number of international studies have dealt with how differences in language and culture affect clinical care, especially in the United States of America (USA) where the effects of cross-cultural communication in the non-English speaking minority groups have been examined in depth. Few studies have focussed on the consequences of cross-cultural communication between patients and health professionals in South Africa. Swartz and Drennan (2000) maintain that the slow progress towards achieving greater access to mental health services in South Africa has its roots in language difficulties (Swartz and Drennan, 2000). Levin addressed these communication issues by assessing the effect of language barriers in a South African paediatric teaching hospital. In particular he highlighted difficulties experienced by Xhosa-speaking parents in understanding English terms used by doctors (Levin, 2005; Levin, 2006). Interpretation is an obvious and important factor in cross-cultural communication and various studies in the USA have explored problems linked to interpretation. In a South African study, A. Crawford has comprehensively documented problems experienced by nurses who have to take on the role as interpreters in South African hospitals (Crawford, 1999).

Lay understanding of genetics has also been well researched in the western world, especially pertaining to bilateral kinship systems, in which kinship ties are traced through both parents. Minimal research has, however, focused on patrilineal descent systems, where kinship is traced through the paternal parent, which is common in many Asian, Middle Eastern and African cultures. This dearth of research also applies to the impact of language and cultural issues on multicultural counselling, although some research in South Africa has focussed on HIV/AIDS and on counselling psychology.

2.2 Haemophilia

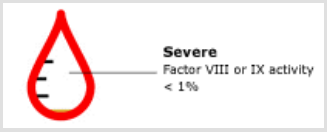
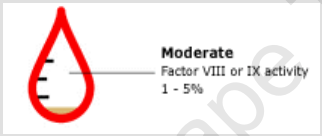
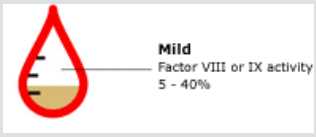
Haemophilia A and B are bleeding disorders, caused by genetic defects that result in low concentrations of specific blood coagulation factors (Talaulikar et al., 2006). The earliest written references to what may have been human haemophilia are attributed to Jewish writings of the 2nd century AD. A familial bleeding disorder was recognized and discussed in the Talmud and other Hebrew writings (Ingram, 1976; Pierce and Rosner, 1994). The first written records describe how Rabbi Judah, the patriarch, decreed that a woman's third son may not be circumcised if his two elder brothers had died of bleeding after circumcision. There also appears to have been an understanding that the disorder is passed on by mothers to their sons and Rabbi Simon ben Gamaliel forbade a boy to be circumcised because the sons of his mother's three elder sisters had died after circumcision (Ingram, 1976). The explanation offered in the ancient writings is that members of some families have "loose blood," whereas normally the blood is "held fast" (meaning it coagulates) (Pierce and Rosner, 1994).

Haemophilia A is the most common form of haemophilia and occurs in 1 in 5,000–10,000 male births, while haemophilia B occurs in 1 in 20,000–34,000 (National Haemophilia Foundation [USA], 2006). The worldwide incidence of Haemophilia A and B may, however, be even higher, because affected patients are not always diagnosed in developing countries. Blood coagulation is a highly regulated process that depends on the interaction of blood vessels, blood platelets and coagulation ('clotting') factors (Ramasamy, 2004). Clotting factors are multiple plasma proteins that cause 'cascades' of enzymatic reactions in blood plasma. Patients with haemophilia A or B have a mutation (an abnormality) of the gene coding for coagulation factor VIII or IX respectively, leading to abnormally low plasma levels of these clotting factors (Nuss et al., 2002). Bleeding occurs because of failure of secondary haemostasis (the arrest of bleeding). Primary haemostasis, which involves the formation of the platelet plug, occurs normally, but the stabilisation of the plug by fibrin is defective because inadequate amounts of thrombin are generated (Bolton-Maggs and Pasi, 2003). Patients with haemophilia do not bleed faster than usual, but continue bleeding for a longer period of time than unaffected individuals.

2.2.1 Clinical manifestations

The prevalence of haemophilia does not vary significantly between populations and a family history is known in about two thirds of cases. The remaining cases are caused by de novo mutations (new gene mutations) where the mother has an 85-90% chance of being a carrier (Tizzano et al., 2003; Graw et al., 2005). Haemophilia is clinically classified as severe, moderate, or mild according to the levels of circulating factor VIII or IX which are directly related to the frequency of bleeding (Table 1). Classification ranges from < 1% factor as severe to >5% factor as mild.

Table 1: Clinical classifications of persons with either haemophilia A or B

SEVERE	MODERATE	MILD
		
Generally 0-1% factor	Generally 1-5% factor	Generally 5-40% factor
May bleed spontaneously from early infancy	Bleeding generally follows trauma or surgery. Occasional spontaneous haemorrhage	Bleed only after severe trauma or surgery. May never have a bleeding problem
May require factor 1-2 times per week	May required factor 1 time per month	May never require factor
Characterized by joint bleeding (hemarthrosis)	May have joint bleeding	Rarely has joint bleeding

(World Federation of Haemophilia, 2009)

Patients with haemophilia with moderate or severe factor VIII or IX deficiency are treated with intravenous factor VIII or IX concentrate to control the bleeding. Unfortunately, approximately 30% of people with haemophilia A develop an inhibitor or antibody to the anti-haemophilic drugs, which render the drugs ineffective (Oldenburg et al., 2002; Graw et al., 2005). The development of inhibitors is a life-threatening complication for people with haemophilia A and is influenced by haemophilia severity and mutation type. Overall, inhibitors appear in 20-25% of patients with severe haemophilia A (Lorenzo et al., 2001). It has also been shown that

the development of risk factors is more frequent in black African patients. Inhibitors are much less common in haemophilia B (SAHFMAC, 2000).

The most frequent symptoms in haemophilia are haemarthrosis (bleeding into a joint causing swelling and pain) and haematomas (collection of blood inside a body cavity or soft tissue resulting in swelling, pain, and bruising) especially in children under school and of school age. The replacement therapy with factor concentrates can stop the haemorrhages, but not reduce synovitis (inflammation of the joint lining which can eventually lead to surface cartilage destruction and joint stiffness) and muscle disorders. Intracranial haemorrhage (ICH) is a feared complication and a common cause of morbidity (illness) and mortality (death) of patients with haemophilia all over the world (Ghosh, 2004)

2.2.2 Gene defects

Haemophilia A is caused by mutations in the *FVIII* gene. The *FVIII* gene spans 186 kb (kilo base pairs), contains 26 exons, and is located on the long arm of the X chromosome at Xq28. This gene is unusual because it contains two additional genes, *F8A* and *F8B*, within intron 22 (Lakich et al., 1993). The functions of the *F8A* and *F8B* transcripts and their potential translated products are unknown, although a number of features suggest that they may function as housekeeping genes (Soares et al., 2003; Colman et al., 2006). Mutations in *FVIII* result in significantly reduced plasma levels of coagulation factor VIII. The gene has a high mutational heterogeneity and defects include point mutations, deletions, duplications and inversions (Table 2). As of Jan 2007, 943 unique mutations, of which 797 were unique point mutations, had been reported on the comprehensive haemophilia *FVIII* mutation database and sequence resource site (HAMSTeRS, http://europium.csc.mrc.ac.uk). Worldwide, point mutations are by far the most prevalent type of defect and may underlie the disease in 90-95% of patients. Point mutations include missense mutations, nonsense mutations and mRNA splice site point-mutations (Bowen, 2000). Deletion mutations account for 5-10% of gene defects, while duplications are very rare. An inversion in the *FVIII* gene is the most common mutation

causing severe haemophilia and results in the disruption of intron 22, and, more rarely, of intron 1. The same types of defects are found in the *FIX* gene for haemophilia B, except that in this gene virtually all are unique mutations and no two unrelated families tend to have the same mutation (Mahlangu and Gilham, 2007).

Table 2: Molecular pathology of haemophilia A

Molecular pathology	Effect of mutations	Incidence
Point mutations:		Collectively these mutations may underlie the disease in 90-95% of patients
• Missense mutations	A codon is changed so that a different amino acid is encoded	
• Nonsense mutations	an amino acid codon is changed into a 'stop' codon	
• mRNA splice site mutations	An mRNA splice site is corrupted or a novel one is created	
Deletion mutations	a sequence of DNA is missing	Collectively deletions, duplications and inversions account for about 5-10% of gene defects. However, across populations, Intron 22 inversions account for 40-50% of severe haemophilia cases.
Insertion mutations	a sequence of DNA is inserted	
Inversions	a nucleotide sequence has been reversed; intron 1 and 22 are inversion hotspots	

Up to 30% of new cases are sporadic (occur for the first time in a family) and result from de novo mutations. Oldenburg et al. (2002) propose that a significant proportion of these mutations can be attributed to a germline or somatic mosaic originating from a mutation during early embryogenesis. Routine mutation analysis is not able to identify all causative mutations and especially somatic mosaicism may remain undetected (Leuer et al., 2001). It is thus important, especially during risk assessment in genetic counselling, that the possibility of somatic mosaicism be considered in families with sporadic cases of haemophilia.

2.2.3 Genetic inheritance

Haemophilia A and B are inherited in an X-linked recessive pattern where males are generally affected and females are either unaffected or have mild symptoms. Males

possess only a single X chromosome and are thus hemizygous for X-linked loci, and the mutant allele is fully expressed in this sex. Females have two copies of the X chromosome and even if they carry the faulty gene copy they have a "back up" correct copy on the other X chromosome and are usually unaffected by the condition. The mechanism of dosage compensation through random X-inactivation ensures that females and males have the same amount of most X-linked gene products. Random X-inactivation results in approximately equal proportions of somatic cells, in which either the normal X or the mutated X chromosome is active. In some instances however, when X-inactivation is non-random, symptoms are also visible in females (Newall et al., 2001; Lin et al., 2008).

2.2.3.1 Genetic testing

Genetics is a branch of biology that, amongst others, deals with heredity, especially the mechanisms of hereditary transmission. Genetic testing identifies changes in chromosomes, genes, or proteins and is generally used to confirm or rule out a suspected genetic condition or help determine a person's chance of developing or passing on a genetic disorder.

In some cases of haemophilia A and haemophilia B, a mutation cannot be identified. However, it may be possible to use indirect or linkage tests to determine the gene carrier status of females by tracking the gene in the family. For linkage testing, linked markers are used to identify the high risk X chromosome. These markers are typically close to the gene and track with the disease in families (Pulst, 2003).

In South Africa DNA testing for haemophilia A is done by the National Health Laboratory Services (NHLS) at the University of Witwatersrand's Division of Human Genetics. Testing is routinely undertaken to determine if the disease-causing mutation is the inversion mutation in intron 22, as this mutation accounts for 40-50% of severe cases. If the common inversion mutation is not detected, linked marker analysis can be undertaken for prenatal diagnosis and to attempt to determine carrier status of family members.

2.2.4 Management of haemophilia

The types of bleeding in haemophilia are divided into major and minor bleeding episodes. Major bleeding episodes may cause death or disability and appropriate factor replacement has to be started urgently and hospitalisation is usually required to maintain adequate factor levels. Minor bleeding episodes, although considered minor, can nonetheless lead to complications and have to be treated early to avoid long term complications (SAHFMAC, 2000).

Major bleeding episodes include:

- Bleeding in the central nervous system
- Severe bleeding from the gastro-intestinal tract
- Severe bleeding from injury
- Advanced muscular/joint bleeds

Minor bleeding episodes include:

- Early muscular/joint bleeds
- Muscle/soft tissue bleeds
- Mouth/gum bleeds

Consequences of repeated acute or chronic bleeding include:

- Anaemia
- Chronic haemophilic arthropathy
- Contractures, caused by repeated joint bleeds and abnormal postures
- Unequal length of limbs, because of repeated joint bleeds resulting in varus or valgus deformity (Ghosh, 2004).

Acute bleeding events are, as mentioned previously, treated with intravenous factor VIII or IX concentrate. The half life's for factor VIII and IX are 8-12 hours and 16-24 hours respectively, after which the infusion has to be repeated if necessary. Patients with mild factor VIII deficiency may receive a synthetic compound, DDAVP (Desmopressin), which is administered if they are proven responsive to it (Nuss et al., 2002, SAHFMAC,

2000). For patients who have developed inhibitors to the factor concentrates, treatment options include the very expensive recombinant factor VIIIa (a genetically engineered clotting factor) or activated prothrombin complex concentrates which have been developed to bypass factor VIII and factor IX in the coagulation cascade (Negrier et al., 2008).

Management of haemophilia is, unfortunately, still hampered by mismanagement, ignorance and poverty, especially in developing countries. Studies conducted in Brazil and New Zealand highlight that limited clinical awareness often impedes the identification of persons with haemophilia (Park and York, 2008; Fontes et al., 2003). Patients and caregivers in New Zealand feel that it is often up to them to decide whether the doctor has adequate experience with haemophilia and, especially during major bleeding episodes, they find themselves in situations where they have to make ad hoc decisions to resolve a crisis (Park and York, 2008).

2.2.5 Caring for a child with haemophilia

Caregivers raising a child with a bleeding disorder have many issues to address. They need to learn how to deal with the child's emotional responses to living with a chronic health problem and also be sensitive to the emotional responses of siblings (Evatt et al., 2004). They need support and guidance to learn to live with this chronic condition and to deal with it adequately in order to ensure an all-round healthy development of the child and his/her siblings (Canclini et al., 2003). The physical vulnerability in particular may cause anxiety and insecurity and caregivers have to learn to balance the desire to protect their child's wellbeing while allowing the child to lead as active and normal a life as possible (Barlow et al., 2007). In addition, they and their families may bear a great financial and social burden. Barlow et al. (2007) explored the experience of living with bleeding disorders in the United Kingdom (UK) and reported that many participants commented on the financial impact these disorders had on their families. Several researchers have emphasized the importance of effective genetic counselling for these

families, to raise awareness of possible carrier status and to prevent unwanted recurrence of the disease (Banis, 1999; Kelley and Narváez, 2006; Lin et al., 2008).

The impact of such a disease and adjustment to it will vary for each family, depending upon family type and characteristics of the family (Goldstein and Kenet, 2002). There are several haemophilia support groups in the Cape Metropol. These are run or initiated by treatment centres and allow people with bleeding disorders to discuss the issues they confront in their daily lives. Support groups provide the opportunity for all members to share common experiences, to exchange practical tips on preventing injuries and to obtain advice on what to expect at different ages (National Haemophilia Foundation [USA], 2006). Members are able to meet people who share their concerns and listen while they voice their anger, worries, and frustrations. Above all, support groups can help families to realize that they are not alone (Kelley and Narváez, 2006).

2.2.6 Genetic counselling

"Genetic counselling is the process of helping people understand and adapt to the medical, psychological and familial implications of genetic contributions to disease. This process integrates the following:

- Interpretation of family and medical histories to assess the chance of disease occurrence or recurrence.
- Education about inheritance, testing, management, prevention, resources and research.
- Counselling to promote informed choices and adaptation to the risk or condition" (Resta et al 2006:77)

Generally genetic counsellors provide their clients with information on the various options available, listen to their clients' feelings and beliefs and act as a sounding board while clients make their own decisions. As part of genetic counselling, a physician or genetic counsellor will take a family history and draw a family tree, called a pedigree.

The pedigree will help identify family members of patients with haemophilia who could be carriers of the faulty gene.

2.2.6.1 Genetic counselling and haemophilia

Perceptions of haemophilia can vary and are influenced by personal beliefs as well as religious and cultural traditions. Park and York (2008) reported on two sisters, who had featured in the news media in New Zealand because of their determination not to have boys with haemophilia. The sisters' position was based on their experience of the suffering that their late father had undergone as a result of haemophilia. Yet several other participants in the same study commented that, while it was true that men who would now be in their 40s to 60s had had a particularly difficult time it was different for children born now. Today the situation has changed because haemophilia has become a manageable condition with improving prospects (Park and York, 2008). Perceptions and beliefs vary and often depend on personal experiences. Counsellors should always explore these beliefs with patients and family members as they can have a major impact on the choices people make about having a child that may be affected (Mannucci, 1993). Women related to a male with haemophilia, such as a mother, sister, aunt, or cousin on the mother's side, can be carriers (Ross, 2000), and as a recent study in the United Kingdom (UK) has highlighted, communication of carrier status to daughters of families with haemophilia is deemed very important (Gregory et al., 2007). Knowledge of their status helps prepare them for decisions they may have to make concerning future reproductive issues. The daughter of a father with haemophilia will definitely carry the gene for haemophilia and is an obligate carrier. If one of her brothers has haemophilia however, she would have to undergo genetic testing to determine whether she has inherited the mutated gene. Several factors can influence a women's decision about carrier testing or how she chooses to use that information, including the severity of the haemophilia that occurs in her family, her own desires and beliefs as well as those of her partner. Daughters confirmed to be carriers, may have problems deciding whom to tell and when and may have to deal with the guilt of passing on haemophilia to their children (Ross, 2000). Carrier daughters may also be afraid that their marriage chances will be

affected and may, thus, be reluctant to disclose their status. Ideally carriers should be counselled and educated about the possibility of inheriting or passing on haemophilia before or when marriage takes place or before any children are conceived (Miller, 1999).

Counsellors need to be aware of their client's perceptions and beliefs in order to resolve misconceptions, to encourage their clients to accept responsibility and to provide them with information that can help them make decisions that will affect their future and the future of their offspring (Lanie et al., 2004; Verwey and Crystal, 1998). In order to negotiate these complex issues counsellors should have a good understanding of the genetic knowledge base of participants (Vermette et al., 2001), be able to communication at an appropriate level, and provide counselling that is culturally sensitive (Mannucci, 1993).

2.2.6.2 Multicultural counselling

Empathy is the cornerstone of counselling and is demonstrated through the use of paraphrasing, summarization, and interpretation of client statements (Sue, 1997). As Ibrahim (1991) notes, "The ability to convey empathy in a culturally consistent and meaningful manner may be the crucial variable to engage the client".

To meet the needs of families who carry genetic disorders counsellors must take into account their client's lay knowledge of inheritance as well as understand family processes that govern family relationships and reproduction (Richards, 1996). As mentioned earlier, however, there is very little information available to counsellors regarding the understanding of inheritance within the Xhosa patrilineal kinship system in South Africa. This raises the question of how effective cross cultural genetic counselling can be in South Africa. An audit of the University of the Witwatersrand's Division of Human Genetics' data on black African women, who were offered an invasive procedure (amniocentesis), revealed a low uptake rate compared to other studies (Penn et al., in press).

Traditional counselling has been criticized as Eurocentric or ethnocentric (Sue, 2001; Tsang et al., 2003) and there is a worldwide growing awareness of how cultural issues impact on the counselling relationship (Robinson et al., 2000). Critics claim that traditional counselling theory and practice is based exclusively on white culture and that counsellors fail to meet the needs of a culturally diverse client population (Katz, 1985; Port et al., 2008). Studies in America suggest that early termination from counselling by non English-speaking minority groups often stem from the fact that client and counsellor are not culturally compatible (Sue, 1977), and that early termination from counselling may stem from mistrust of white counsellors by culturally diverse clients (Watkins and Terrell, 1988). Richardson and Molinaro stress that to ensure multi-cultural competence, counsellors should be taught "

(a) knowledge about the nature and uniqueness of cultural groups and understanding of the socio-political experiences of those cultural groups,

(b) skill acquisition focused on the quality and appropriateness of interactions or interventions between the counsellor and diverse clients, and

(c) self-awareness of the role of culture in one's own experiences" (Richardson and Molinaro, 1996).

Similarly Carter (1991) stresses that in addition to learning about others, there is a need to learn about oneself. To develop multicultural competence in counsellors, critical self awareness factors must be identified and incorporated into multi-cultural training (Richardson and Molinaro, 1996). Ancis and Szymonski emphasise that self-awareness includes admission of ones own racist attitudes and beliefs, and in the case of white counsellors, knowledge of "how they may have directly or indirectly benefited from individual, institutional, and cultural racism" (Ancis and Szymanski, 2001, Sue et al., 1992). The privileges of being white are often not expressed or scrutinised by those who benefit and unless white privilege, ethnocentrism and unintentional racism are addressed, progress towards developing multicultural competence will be delayed (Ancis and Szymanski, 2001). Counsellors must thus be aware of their client's as well as their own cultural values and beliefs and how these may influence attitudes and behaviour (Randall-David, 1989).

Beliefs around the cause of a condition are often culturally determined, and without understanding the needs of a cultural group there can be no adequate delivery of inclusive health care services (Cohen et al., 1998). A recent study in New Zealand reports on the processes undertaken to provide culturally-sensitive genetic services for the Maori people (Port et al., 2008). Western models of genetic service are based on the philosophy of informed choice and consent as well as independence and empowerment of the individual. Central to this model is the genetic counselling service. Port et al., (2008) established that the hierarchical tribal structure of the Maori includes inherent rights of the collective and that the western model of individual autonomy cannot be integrated into this structure. Similarly to the Maori, the amaXhosa are a patrilineal population with a hierarchical tribal structure, in which the line of kinship and descent is traced through the paternal line. Particularly in the context of traditional healing, the unity of a person within the amaXhosa community is paramount (Louw and Pretorius, 1995).

2.2.6.3 Communication about genetic disorders within families

Communication about haemophilia within families fulfils a very different role from communicating with health care providers. Families can serve as a forum to discuss the everyday management of symptoms, sharing of duties and being available during emergencies. Open communication and encouragement to share concerns is also crucial to help identify every family member's needs. To promote meaningful communication within the family concerning risks and carrier status it is thus important that parents are aware of inheritance patterns, carrier testing and prenatal testing services (National Haemophilia Foundation [USA], 2006).

2.2.7 Stigma and Haemophilia

HIV infection in haemophilia was reported in 1981, and in the UK alone more than 1200 individuals were infected by blood-product infusions between 1979 and 1985. There have however been no new cases of HIV in the UK since blood has been virally

inactivated in 1986 (Bolton-Maggs and Pasi, 2003). In a recent study undertaken by Barlow et al. (2007) in the UK, participants reported that because of contaminated blood products in the past, there were still misconceptions about people with bleeding disorders. The mistaken belief that individuals with haemophilia must be HIV positive and that all men who have haemophilia must be gay still persist.

Another issue faced by caregivers of children with haemophilia is the sudden, unexplained bruising in infants or young children. Bruising is common and often lumpy and may be sufficiently extensive to raise the suspicion of child abuse. Park and York (2008) reported on parents of boys with haemophilia in New Zealand who had to fend off accusations of abuse because of spontaneous bruising. Similarly, Cassis (2007) reported that, especially with no known family history of haemophilia, parents often had to endure questions about domestic violence and child abuse.

2.2.8 The South African Health System

South Africa's health system consists of a large public sector and a smaller but fast-growing private sector. Health care varies from the most basic primary health care, offered free by the state, to highly specialised hi-tech health services available in the private sector for those who can afford it. The private system attracts most of the country's health professionals and has been rated as excellent. The United Nations ranked South Africa's private system 39 out of 162 nations for technological innovation and achievement. (South Africa Info: The official Gateway, 2009).

The public sector on the other hand, which delivers services to ~80% of the population is under-resourced. Health care standards within the public sector vary according to location with large, urban teaching hospitals offering good, if clogged-up, service while many rural hospitals are run-down with broken equipment, worn out ambulances and a shortage of doctors and basic medicine. The public sector is overused because of a high incidence of poverty (71% in rural areas and 50% overall), and unemployment (at least 38%) which make it almost impossible for the majority of people to pay for health

services (Mansoor and Dowse, 2007; South Africa Info: The official Gateway, 2009). An additional strain is put on the public sector because of the high rates of infection with the human immunodeficiency virus (HIV) and full-blown AIDS, which require increased human and financial resources (Benatar, 2004).

Traditional healers in South Africa also play an important part in the South African health sector. They have existed in South Africa for many centuries and have continued to flourish in the face of competition from modern medicine. It is estimated that between 150 000 and 200 000 traditional healers practice in South Africa, compared with 25 000 doctors in modern medicine and that up to 80% of the black population use the services of traditional healers. Traditional healers are enshrined in the minds of the people, are respected in their community, and are often its opinion leaders (Kale, 1995). The Traditional Health Practitioners Act No. 35 recognises and regulates the practice of South Africa's traditional healers and was approved by Parliament in 2004.

The act serves to:

> "establish the Interim Traditional Health Practitioners Council of South Africa; to provide for a regulatory framework to ensure the efficacy, safety and quality of
> traditional health care services; to provide for the management and control over the registration, training and conduct of practitioners, students and specified categories in the traditional health practitioners profession; and to provide for matters connected therewith" (Republic of South Africa: Traditional Health Practioners Act No. 35, 2004)

2.2.9 Haemophilia health care

Haemophilia is one of 26 diseases in the list of chronic conditions for which care cannot be refused in South Africa (Mahlangu, 2009). However, medical care of patients with haemophilia does not enjoy the same urgency as the AIDS pandemic, malaria

infestation, malnutrition and poverty alleviation, all of which are national health priorities (Jones, 1999; Benatar and Fleischer, 2000). There is however a functional National Haemophilia Programme which is driven and supported by the South African Haemophilia Foundation, the Medical and Scientific Advisory Council and the South African National Department of Health (Mahlangu, 2009).

Delivery of haemophilia health care in South Africa is through a network of haemophilia treatment centres, which function in the context of the National Department of Health. They work in close partnership with specialist haemophilia health care workers of the Medical and Scientific Council and the South African Haemophilia Foundation. Unfortunately haemophilia care faces a number of challenges which include limited diagnostic facilities, inadequate number of treatment centres and lack of haemophilia specialists. Despite these challenges the existing haemophilia treatment centres are able to deliver high quality of care, improving the quality of life and reducing mortality due to bleeding in patients (Mahlangu, 2009).

Most of the participants in this study had migrated to Cape Town from the Eastern Cape. The Eastern Cape Province has amongst the worst health and socioeconomic indicators in the country (Xoli et al., 2001). Health services, predominantly providing primary health care, are provided by a network of 920 public health facilities spread across the Province. Access to first level care is through clinics, mobile services and satellite clinics that provide services only on certain days (Xoli et al., 2001). This poses significant difficulties for patients with haemophilia who need timely and effective treatment when a bleed occurs. Because haemophilia is a relatively rare disorder, rural hospitals or clinics often do not know how to diagnose or treat patients (Park and York, 2008; Beijing Aizhixing Institute, 2006). In addition, because of lack of awareness of hospital staff, patients often have to wait until the bleeding has reached a critical state (Beijing Aizhixing Institute, 2006).

2.3 South African Demographics

South Africa is a nation of over 47 million people of diverse origins, cultures, languages, and beliefs. The 2006 estimated figures for the various ethnic categories were Black African 79.5%, Caucasian 9.2%, Mixed Ancestry 8.9%, and Indian or Asian 2.5% (Fig. 1) (Statistics South Africa, 2008). Despite the fact that the major part of the population classifies itself as African or Black, it is neither culturally nor linguistically homogeneous with 11 official languages recognised in the constitution of the Republic of South Africa.

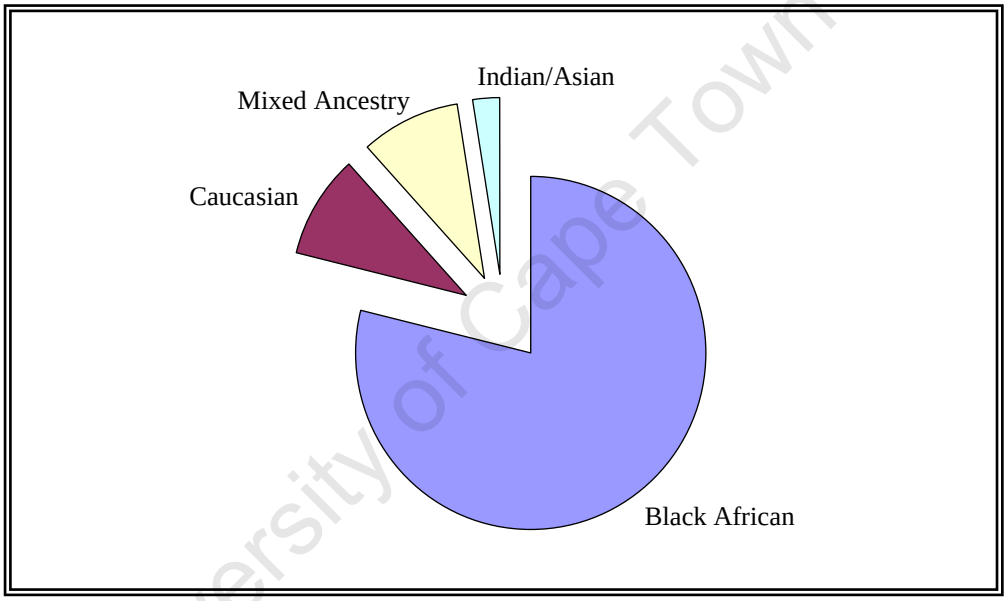


Figure 1: South African demographics in percent (Statistics South Africa, 2008)

This study focussed on caregivers of Xhosa patients with haemophilia in the Cape Metropole, South Africa. Haemophilia affects every ethnic group equally and the haemophilia patient population is thus culturally and socio-economically as diverse as the general population.

2.4 ama-Xhosa

Modern Xhosa are South Africa's second largest ethnic group, with approximately 7 million people. They are an African population that form part of the Nguni language

group. Nguni peoples are classified into three large subgroups: Northern Nguni, Southern Nguni, and Ndebele. The Xhosa group make up the largest Southern Nguni society (Hammond-Tooke, 1998).

2.4.1 History of clans

The following paragraph aims to give a brief insight into kinship organization amongst the southern Nguni. Traditional societies are often organized into descent groups in which the population is divided into tribes, the tribe into clans and the clans into lineages (Fig. 2) (Chaix et al., 2004; Mtuze, 2004). A lineage can be defined as a corporate descent group of kin who are able to trace their genealogy to a common ancestor, who is traced back through the male or female line. Patrilineal populations, in which the line of kinship and descent is traced through the paternal line, are roughly twice as frequent as matrilineal populations (Burton et al., 1996). Members of such a group may hold property, assign status within the group and regulate inter-group relationships (Llobera, 2003). Over time the lineage group will grow too large and become unmanageable, leading to splits and the formation of new lineages (Fig. 2). These new groups will however still acknowledge a certain type of link to each other, which leads to the formation of a non-corporate clan (Llobera, 2003). Members of a clan usually claim descent from a named but distant and often mythical ancestor. The main function of a clan is ceremonial but can also regulate marriage through exogamy (the custom of marrying outside the clan or other social unit) (Llobera, 2003; Hammond-Tooke 1998).

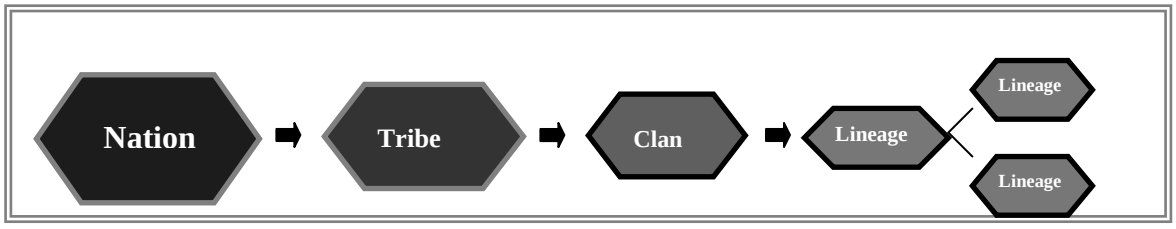


Figure 2: Nations divided into tribes, tribes into clans and clans into lineages (Mtuze, 2004)

In the past all Nguni had patrilineal exogamous clans who shared a common clan name and were believed to be descendants of a common ancient clan founder (Hammond-Tooke, 1998). Under patriliny, the lines of descent and authority are traced through

fathers: a man and his brothers, their children, and their sons' children are counted as members of the same descent group.

Hammond-Tooke (1998) maintains that historically it has been difficult to establish relative status within a clan because no written records existed. It was thus not possible to prove descent from a clan founder nor was it possible to prove relationship to other clan members. Within the clans there were different units of people that formed lineages and these could indeed demonstrate genealogical relationship to each other on a family tree. These relationships were mostly not more than about six generations, as, in the absence of writing, genealogies can usually only be committed to memory no further back than grand- or great-grandfather. He argues that a Nguni social group beyond the family would have consisted of the segment of a lineage that shared a particular location and formed the congregation of the ancestor cult. The ancestors were the spirits of the deceased senior males of the patrilineal lineage or clan (Magubane, 1998, Hammond-Tooke, 1993). The Nguni typically defined the group of ancestral spirits as all the dead of the clan and worshipped this group as a collective. This great group was believed to be present, also as a collective, at all rituals performed by their descendants. Appeals to them were always made by a senior male of the clan or lineage (Hammond-Tooke, 1993). Even today the clan name is considered to be more important than a surname. The clan name is the strongest way of identifying someone, even stronger than a father's or grandfather's surname because it identifies a person's whole family group and ancestors. (Pinnock and Schonstein, 1988).

One of the main functions of the clans was to define marriage through the exogamy rule (marriage partners were sought outside the kin group) (Magubane, 1998). Marriage inside the Nguni patrilineal clan was considered a heinous offence and strictly

prohibited. The Xhosa were known to be particularly fastidious in this regard and marriage within the clans of all four grandparents was taboo (Magubane, 1998).

2.4.2 Traditional healing

Traditional healers are established community-based health care workers. Even today, between 60 and 80% of South Africans still use traditional healers as their first call for medical advice. Traditional healers are generally very caring people who are highly skilled in psychotherapy and counselling (Kale, 1995). They treat their patients holistically and consider not only the physical, but also the psychosocial aspects (Buehrmann, 1984; Pretorius, 1999)

Traditional medicine is defined in the Traditional Health Practitioners Act No. 35 as

> "… an object or substance used in traditional health practice for –
>
> (a) the diagnosis, treatment or prevention of a physical or mental illness; or
>
> (b) any curative or therapeutic purpose, including the maintenance or restoration of physical or mental health or well-being in human beings, but does not include a dependence-producing or dangerous substance or drug" (Republic of South Africa: Traditional Health Practioners Act No. 35, 2004).

Diviners and herbalists play a significant part in Xhosa culture and it is necessary to note the important distinctions between the two. A diviner's (Xhosa: *igqirha,* Zulu*: isangoma*) vocation stems from a special 'calling' from the ancestors and he is believed to work with divine assistance (Hammond-Tooke, 1974). He has the power to interpret the cause of misfortune, establish the cause of serious illnesses, failure of crops, epidemics of disease in human beings and cattle, to ask favours of the ancestral spirits and more (Mtuze, 2004). The herbalists (Xhosa: *ixhwele*) are ordinary people without divine calling and are masters of medicines with a vast knowledge of plants, roots and other substances. They are rather like modern pharmacists, whereas diviners are similar to medical doctors (Hammond-Tooke, 1993; Dagher and Ross, 2004).

The practice of traditional healing continues to be widespread amongst black South Africans, regardless of their inclination and many modern Xhosa continue to place a high importance on ancestors and ancestral spirits (Ashforth, 2005). Traditional beliefs are still very much intertwined in daily life. This also applies to pregnancies which are

seen as a delicate time when women have lots of problems. Findings of a study conducted in and around Cape Town by Abrahams et al. (2002) show that the majority of isiXhosa speaking women follow indigenous healing practices for both themselves and their babies. Interviews with indigenous healers revealed that problems during pregnancy are particularly unpredictable and could harm both mother and child. Women and their babies are said to be vulnerable to "evil" sent by third parties or just to `spells' which a woman might step over in the environment (Jewkes and Wood, 1998). Women especially look to indigenous practices with concerns related to the need of strengthening the womb against sorcery and the prevention of childhood illnesses. The fact that they follow indigenous healing practices does, however, not mean that they do not visit clinics regularly, they simply visit both. A study by Sifunda et al. (2007) demonstrated that health care-seeking behaviour among isiXhosa speakers very often involves the consultation of the dual system of Western-based medicine and traditional healers. While they will accept western medicine and advice they will, at the same time, turn to traditional healers who provide herbal remedies, potions and spiritual counselling (Sifunda et al., 2007).

These healers believe in traditional herbs and roots and ask for guidance from their ancestors to cure illness, to relieve a streak of bad luck or bless a child (Jewkes and Wood, 1998). Spirit and body are seen as a whole and illness is conceived in terms of a breakdown of human relationships. The living community is seen as an extension of the ancestral community and life on earth and beyond are continuous and interdependent. The spirits of the ancestors protect the living community and must be honoured through the teaching of traditions and the practice of appropriate rituals; otherwise their spirits will cause problems that can manifest in poor health, misfortune or social disruption (Wessells and Monteiro, 2004; Hammond-Tooke, 1993).

> *"It is as if the world were a spider's web in which any touch of a single thread reverberates throughout the entire structure"* (Tempels, 1965 in Wessells and Monteiro, 2004:325).

Traditional healing uses a holistic approach and aims to restore a harmonious social life, and treatment of illness consists of coming to grips with relationships with ancestors or, between close kin and neighbours (Hammond-Tooke, 1985; Nyika, 2006). Louw and Pretorius (1995) highlight some fundamental differences between approaches taken by traditional healers versus Western Psychotherapy and counselling (Table 3).

Table 3: Differences between approaches taken by traditional healers versus Western psychotherapy and counselling (Louw and Pretorius, 1995).

African approach	Western approach
African approach is symbolic and intuitive.	Western is based largely on scientific and logical principles
Traditional healers act as a mouthpiece of the ancestors, are directive and give advice to their clients. Guidance is therefore considered to come from the superior wisdom of the ancestors, not from the healer.	Western counselling is generally non-directive, giving clients information to enable them to take responsibility and make their own decisions
The unity of a person within a community is emphasised in traditional healing.	Western healing places more emphasis on the individual.

It is also relevant to note here that different cultural beliefs and practices give rise to different expectations. Traditionally, a Xhosa person would have consulted an African diviner whose duty it is to diagnose the disease and cause of the disease without asking the patient from what he/she is suffering. Diviners appeal to their ancestors for help in diagnosing problems as well as for prescribing remedies. They would also routinely check to see if illnesses are caused by their clients having violated cultural norms and traditions. Consequently, as opposed to western expectations, when consulting a doctor this person would similarly expect the doctor to diagnose his/her sickness without having to describe the symptoms (Mtuze, 2004; Nattrass, 2006). The concepts of ancestor cult and traditional healing are deeply entrenched in Xhosa society and it is, therefore, reasonable to infer that this traditional model of illness will have a significant effect on care-seeking behaviour within this society.

2.4.3 Circumcision

Rituals are a means for society members to communicate values and ways of living, and to reinforce a particular collective identity. In South Africa, traditional rituals (*amasiko*) are performed by the ama-Xhosa at virtually every stage of the life-cycle, from birth through puberty, marriage and menopause to death. Rituals are still widely performed today, both in the rural areas and in the suburbs of South African towns and cities. African culture, like other cultures is dynamic and adapts itself to external circumstances and events and rituals may today be performed in considerably altered and moderated form (Magubane, 1998). Male initiation however, remains an intact and nationally acceptable ritual and is an important facet in a Xhosa boy's life. The ritual is practiced by Xhosa society on a large scale and is essential to mark the initiation of young boys into manhood (van Vuuren and de Jongh, 1999). Men are recognised as the gatekeepers and custodians of social customs, and this status demands clear demarcation between manhood and boyhood (Mtuze, 2004). Until the young boys have gone through this initiation process, they are regarded as nobodies, are not allowed to address their elders and are generally not taken seriously (Mkoka et al., 2003; Mtuze, 2004). Circumcision is an important, integral part of this male initiation ritual, and it is important for any Xhosa male to undergo this process in order to have status and be regarded as a man by their community.

The male circumcision ritual has long been used by various cultures for religious and cultural reasons as one of the important ritual junctures to provide and reinforce group identity (Heyns and Krieger, 2006). In the Jewish culture for example, circumcision rites were first documented as oral law in the Talmud in the fourth century, but more than likely the practice of the rites precede it by centuries (Kavakli and Aledort, 1998; Ziegler, 1999). Ritual circumcision has been practiced by Jews throughout the centuries, and was later also performed by Muslims (Kavakli and Aledort, 1998) and has become the mark of initiation for these two major religions. In addition to these faiths, ritual

circumcision is practiced by Black Africans, Australian and Polynesian aborigines and some South American aborigines (Kaplan, 1977 in Kavakli and Aledort, 1998).

2.4.3.1 Circumcision and Haemophilia

In many societies where circumcision is a religious, social or cultural practice, people with haemophilia and their families view being uncircumcised as socially unacceptable (Zulfikar et al., 2003). In Xhosa culture the circumcision is performed by an experienced 'traditional surgeon' (*ingcibi*) who nonetheless has no form of medical training. Characteristically he uses non-medical instruments under sometimes unhygienic conditions. Unfortunately, traditional beliefs that only traditional circumcision as opposed to medically directed operations can endow real manhood prevail, in spite of serious problems associated with the practice (Mayatula and Mavundla, 1997). Because patients with haemophilia have a high risk of excessive and prolonged bleeding after any surgical procedure it is essential that patients, caretakers and traditional surgeons fully understand the risks involved (Karaman et al., 2004).

2.4.4 Urbanisation

Many Xhosa-speaking inhabitants of the Cape Metropole today live in one of the townships that have developed on the outskirts of Cape Town. Historically the amaXhosa were cattle and crop farmers on the plains in the Eastern Cape region of South Africa (Anderson et al., 1999). During the mid-20th century, poverty, unemployment and other factors initiated the move of many Xhosa from rural areas to cities, such as Cape Town (Thompson, 1990). These cities were for the most part strange and hostile environments for rural migrants. Urban centres had not grown and developed around local industry or farming, but were driven by foreigners who introduced their own traditions and experiences to urban life. Towns thus developed according to foreign western norms and required rural people, who had different values and whose social structures were organised around kin-based groups, to adapt and to fit in (Mitchell, 1996). The system of "apartheid" ("separateness" in Afrikaans) was a

cornerstone of South African economic development and political policies from 1948 until 1994. Apartheid legislation and specifically the Group Areas Act 41 of 1950 resulted in the creation of townships on the outskirts of Cape Town. These townships were situated far away from city centres or business districts and commuters had to spend hours each day commuting to their places of work (Chapman and Rubenstein, 1998). People from rural areas migrated to these townships, while at the same time they kept strong ties with home areas through regular visits. Research in South Africa has indicated that rural migrants tend to view their stay in urban areas as a transitional phase which helps them cope with the strange and hostile environment. Within this environment they are inclined to trust and have dealings with people from their rural home areas and otherwise choose to keep to themselves (Magubane, 1973; Gugler, 2002).

Apartheid laws obviously also dramatically affected Xhosa family life. Xhosa men for instance, would often only be able to come and work in the cities on one-year contracts and would not be allowed to bring their wives and children with them (Burman and van der Spuy 1996). People from formerly cohesive communities were thus relocated and dispersed, which produced negative effects on household organisation, cultural practices and children's education.

2.5 English language proficiency

Every reflection we make takes place in language which is our distinctive way of being human (Maturana and Varela, 1992). O'Neil (2007) further argues that language is one of the most important components of culture and that it is impossible to understand the subtle nuances and deep meanings of another culture without knowing its language well.

The dominant languages in South Africa have, since colonial days, been English and Afrikaans, and it has been up to the indigenous people to master these languages and familiarize themselves with western culture (Mbatha and Plueddemann, 2004). Very few English speakers have, however, mastered the African languages and even today are

surprisingly ignorant of indigenous cultures. The African language speakers, albeit demographically dominant, were forced to adopt English, because it was the economically dominant language (de Klerk, 2000). Speaking English became essential for economic and social advancement, but, because education was inferior, many black South Africans were not able to get access to adequate English schooling (de Klerk, 1999). During the apartheid era the education system was based on racial and linguistic divisions, where 'Black' institutions were under-funded and a rudimentary curriculum was instructed in African languages (Davies, 1994). African-language schools, including Xhosa schools in the Eastern and Western Cape, were in poor condition and, unfortunately still are as little has changed since the end of Apartheid (de Klerk, 2000). Even today, English competence amongst the amaXhosa ranges from complete fluency to being limited to a few rudimentary phrases (de Klerk, 2003).

2.5.1 Cross-cultural communication

As mentioned previously, South Africa is a nation made up of many ethnic, racial, religious and cultural groups and it has been recognised that this diversity lends strength and uniqueness to our society (Randall-David, 1989). In order to draw the most benefit from this diversity every effort should be made to understand our cultural differences as well as our cultural similarities. Cultural groups can be seen as a collection of individuals who share a common cultural heritage (Randall-David, 1989). Such a group may be defined as an ethnic community that shares language, culture, traditions and history as well as common beliefs, attitudes, values and behaviours (Flores, 2000). At the same time Randall-David (1989) warns that it would be dangerous to suggest that members of particular ethnic groups behave in characteristic ways, as this would lead to over-generalizing or stereotyping. Ethnic communities interact with each other and are exposed to influences from other cultural systems. This process can lead to the transformation of culture and even generate new ethnic communities. Moreover, individuals of an ethnic community may reflect differences in how they adopt or internalize their own culture (Tsang et al., 2003).

The culture you are born into determines your language patterns, behavioural pattern, patterns according to which you interact and how you acquire knowledge. Culture not only influences cognition, the mental process of acquiring knowledge, but also the means by which knowledge is conveyed (Nisbett et al, 2001). The culture of an ethnic community also has a fundamental influence on the perceptions of issues such as health and disease, as well as on the beliefs and practices around these issues (Weil, 2001). In South Africa especially, there are great differences in beliefs and behaviours between physicians and patients, who are often separated by a wide gulf of social class, race and language (Crawford, 1999). Research in the USA has shown that as barriers to health care, socio-economic status and ethnicity are critically important, while the inability to speak English in particular, has been associated with less care-seeking and diminished success (Flores et al., 1998; Woloshin et al., 1997). Various studies have further shown that effective communication between patients and physicians exerts a positive influence on the general management of patients and also on their emotional health (Stewart, 1995). Haffner (1992), a professional medical interpreter in the United States, describes unique obstacles that must be overcome in the medical setting when caregivers and patients experience difficulties in communication. In her experience with Hispanic patients, good health care was often affected by miscommunication and differences in attitudes about health care. She reported problems with proper history taking, because for many Hispanic women only a successful pregnancy counts, while miscarriages or a stillborn child might not be reported. Other problems encountered by her evolved around the modesty of many Latino women and their reluctance to reveal personal or private problems when family members were used as interpreters. She concluded that, to effectively help their patients, health care professionals must be acutely aware of just how different cultural attitudes and beliefs can be.

Generally patients are not equal partners in the medical process. Health care workers usually dominate the medical process, simply because they possess knowledge and skills that patients lack (Fisher and Groce, 1985). As a result most patients are not, or do not feel equal to the health care workers and Waitzkin and Waterman (1974) allege that the gap widens at the lower end of the socioeconomic spectrum. These socio-cultural

divergences between physicians and patient may not only lead to problems in communication, but often also result in relationship barriers (Pachter, 1994). In order to overcome these barriers it would be essential to engage in any cross-cultural encounter with insightful empathy, curiosity and respect while striving to understand how the socio-cultural background of individuals affects their health beliefs and behaviours (Carrillo, 1999).

Medical providers in South Africa face the challenge of caring for patients from many cultures with different languages, socioeconomic status and unique ways of understanding illness and health care (Carrillo et al., 1999; Levin, 2005). But language barriers and cultural differences between participants cause difficulties with communication and, ultimately, result in a major obstruction to health care. (Levin, 2006). Healthcare workers in most major hospitals in the Cape Town region mainly communicate in English or Afrikaans, while many patients speak Xhosa as their first language (Levin, 2006[b]). Numerous first language Xhosa-speaking patients thus face the difficulty of trying to communicate with doctors or counsellors in a foreign language. Not only do they find it difficult to understand the language, but they are further handicapped by not always being able to ask appropriate questions (Levin, 2005, Levin, 2006[b]). Lambert et al. (1997) maintain that while attention has been given to how doctors may improve communication, little or no research has been done on patient competence to communicate. This situation has hardly changed to date. While patients may be more or less competent at answering doctors' direct questions they may be less competent to provide unsolicited information that is relevant to the consultation (Cegala et al., 1996).

Effective communication is a two-way process involving listening just as much as talking and ideally, to better serve their patients, all health care workers, including genetic counsellors, would need to understand as much as possible about the socio-cultural aspects of the community they are trying to reach. Patients make health decisions based on their health beliefs, relationships with medical personnel, and their experiences with health professionals (Vanderford et al., 1997). Communication is

perhaps the most important ingredient in effective health care (Kaplan, 1997). It is essential in the provision of social support (by health care providers, participants in formal and informal support groups, family and friends) and is also vital in co-ordinating health care treatment and activities of various providers and consumers (Kreps et al., 1994). During the communication process, health care providers not only need to provide patients with the necessary information to make health care decisions, but they also need to encourage patients to accept responsibility and to make decisions about their health (Verwey and Crystal, 1998).

The Zulu–speaking people of South Africa have a saying

‘*akubuyelwa nganxanye Kungemanz* ’

That, translated, means that people never see the same thing in the same light. Thus, strength can be viewed as a potential weakness, a threat as an opportunity, depending on one’s perspective and how the identified issue is addressed (Watson and Fouche, 2007).

More awareness by all role players of how culture shapes our attitudes, values and practices would, therefore, lead to more culturally-sensitive health care and to more appropriate community outreach (Randall-David, 1989). Although there is a tradition of research on different cultures’ perceptions of disease and on cross-cultural and cross-language communication in medical settings in South Africa, this has been largely limited to the more common diseases like tuberculosis and HIV (Levin, 2005[b]).

2.5.2 Communication through interpreters

South Africa has eleven official languages and the constitution recognizes the right of every individual to use their language and to participate in the cultural life of their choice. Nonetheless, as Alexander (2005) clearly pointed out: ‘for reasons connected to the colonial history of southern Africa, the language of power in post-apartheid South Africa is undoubtedly English’.

This study took place in and around Cape Town, which is the capital of the Western Cape Province. This province has three official languages, namely Afrikaans, English and Xhosa, but the majority of health care workers only speak English or Afrikaans and cannot speak any of the indigenous African languages (Randall-David, 1989). English is still the dominant language in the medical community and is enshrined as the socially powerful language (Levin, 2006). First-language Xhosa speakers who are not in full command of the English language are thus either unable to communicate effectively or, where possible, are dependent on interpreters.

Levin (2005) extensively documented concerns regarding communication between medical staff and patients at a paediatric teaching hospital in the Cape Town region. All doctors were concerned about communication with their Xhosa speaking patients. Some were concerned about the efficacy of available interpreters and others recognised the need to understand their patients' cultural aspect of a particular disorder. He also reports that difficulties with communication affect doctor's ability to take a reliable history and to ultimately counsel the patients in a meaningful way. The only way to communicate is often through the help of interpreters who are frequently chosen from general hospital staff on an ad hoc basis. He established that many doctors were concerned because the dynamics of the interviews shifted when interpreters were used and some doctors felt that they lost control of the consultation. They were also concerned about the accuracy of the interpretations, missing crucial information from the patients as well as the possibility of misunderstandings (Levin, 2005). Chris Ellis (1999), a doctor working in KwaZulu-Natal, notes that one of the most common causes of misinterpretation is that many African languages use metaphors, allusions and euphemisms especially when dealing with illness. Even on the official DACST (Department of Arts, Culture, Science and Technology) webpage, a sexual organ is referred to in African languages as *the secret member*: ilungu langasese (Xhosa), isitho sangasese (Zulu), setho sa bong (Sotho). Sexual parts are also referred to as *in front* or *the things in front*: ngaphambili (Xhosa/Zulu) and dikapeli (Sotho) (Ellis, 1999).

Although interpreting requires a great amount of skill, interpreters are frequently chosen indiscriminately from amongst whoever is bilingual and available at the time (Penn, 2007; Baker et al., 1998; Putsch, 1985). They are neither formally trained nor paid for this duty and typically have to interrupt their regular duties when required to interpret (Penn, 2007). Baker et al. (1998) determined that compared to patients with no language barriers, patients who communicated through an ad hoc interpreter often perceived their examiner to be less friendly, less respectful and less concerned for them as a person.

Crawford (1999) has likewise identified significant problems in interpreting situations, including loss of meaning, lack of equivalent terms and emphasis that has been added or deleted. Other studies of non-medical interpreters have further revealed that frequently variable message changes occur because of bad paraphrasing and frank omissions (Putsch, 1985). Time constraints can furthermore motivate interpreters to summarize the patient's account, focussing on the key complaint without being aware that the censored information may have a bearing on the consultation. Many doctors are aware of this censoring and see it as a major problem, since what seems irrelevant to the interpreter may be of vital importance to them (Crawford, 1999). One English-speaking doctor at a South African hospital, who is also fluent in Xhosa, said that he has become extremely aware of the extent of mistranslation, loss of meaning and consequent misunderstanding that occurred on a daily basis between doctors, nurses and patients (Crawford, 1999). Putsch (1985) further warns that in interactions involving interpreters often too little attention is paid to the verbal and nonverbal interactions between the interpreter and the patient. He believes that the voiced word accounts for only a small fraction of emotional expression and that the majority of an emotional message is often conveyed non-verbally.

Ideally interpreters need training in specific linguistic skills and counselling techniques. It must also be empathised that it is undoubtedly difficult to achieve immediate and efficient translation, because medical interpretation requires a firm grasp of two different and complex languages and the ability to communicate in each language at many different educational levels (Haffner, 1992). Successful communication in these settings

clearly requires more than mechanical translation between two languages. It also requires that the interpreter and the health care worker are at all times aware of the implications of both, the difference in language and in culture (Haffner, 1992; Putsch, 1985). Studies in the USA have shown that even when trained interpreters are available, there is often no standardization in interpreter training nor are health care providers adequately instructed in how to use an interpreter (Baker et al., 1998). If not taught how to use an interpreter they may, amongst others, inadvertently look exclusively at the interpreter rather than at the patient, decreasing the patients' sense of connection to their provider (Slomski, 1993). Many doctors, when working with an interpreter, unfortunately also do not use their normal strategies such as reflecting and paraphrasing to improve communication (Levin, 2005).

It is further generally recognized that having to rely on an interpreter can be frustrating. The interview will take much longer, communication can be awkward and the health care worker has less control (Haffner, 1992). There are practical issues as well, as there is often a wait before the interpreter arrives and there always seems to be a lack of interpreters because of budgetary constraints, especially in South Africa.

Baker et al. (1998) further highlight the need to define comparative benefits of professional versus ad hoc interpreters and to investigate whether proper training of interpreters will improve the patient-provider relationship. The presence of an interpreter, even if optimally trained, may always negatively affect the relationship between provider and patient, because the usual sense of privacy and intimacy during a consultation is disturbed. Hornberger et al. (1996) found that both patients and providers preferred simultaneous interpretation using headphones to the usual practice of consecutive interpretation with the interpreter present in the exam space.

2.5.3 Translation issues

Issues concerning interpretation and translation are not always the same. While interpretation usually involves face-to-face interaction, translators work with written texts (Temple and Edwards, 2002). The literature points out that it is nearly impossible

to transfer literal meaning from one language to another and that translators usually do not find solutions in dictionaries (Wilson, 2001). There are frequently no exact matches, word for word, in different languages and there is no one correct translation (Temple and Edwards, 2002). Translators need to be familiar with the two different language worlds they inhabit and evaluate the degree to which these worlds are the same (Simon, 1996). Ideally a translator should be a native speaker of the target language, or at the very least an expert (Translation Working Group, www.eurolight-online.eu). The term "target language" is applied to the language that a source text is being translated into. Simon (1996) points out that the translator is actually involved in discussing concepts rather than just words, and that context ultimately decides equivalence or difference in meaning. Understanding a language does not presuppose the ability to translate it. Translation requires the mapping from one language to another language while understanding is something that is internal to a person (Lakoff, 1985). The complexity of translation is emphasized by Gordon (2004) who maintains that studying or learning a second language, particularly if the second language is of a totally different linguistic origin (for example English and isiXhosa), involves learning to understand a different worldview (without necessarily adopting it). The concept of worldview is borrowed from the German word 'Weltanschauung'. 'Welt' is the German word for *world* and 'Anschauung' is the German word for *view* or *outlook*. Weltanschauung is a concept fundamental to German philosophy and refers to a *wide world perception*. Weltanschauung of a people originates from the unique world experience of a people, which they experience over several millennia. Hiebert (2008) suggests that "… 'worldview' is the fundamental cognitive, affective, and evaluative presuppositions a group of people make about the nature of things, and which they use to order their lives."

2.6 Lay understanding of genetics

2.6.1 African society

The aim of this study was to explore the level of understanding of basic genetic concepts among Xhosa-speaking mothers or caregivers of patients with haemophilia. While several studies have assessed lay understanding of inheritance in the general population in western countries, little published data are available on other cultures (Meiser et al., 2001; Richards 1996) As kinship systems vary across cultures it follows that lay knowledge about inheritance is also likely to be subject to cultural variation (Meiser et al., 2001). Ethnographic research has shown that many cultures have a profoundly different understanding of kinship and this may have a substantial impact on the way in which the family history of a genetic condition is understood and presented (Modell, 1997). Barlow-Stewart et al. (2006) recently explored culturally determined attitudes to cancer genetics in Australia using Chinese-Australians as a case in point. They concluded that the level of acculturation does not correlate with holding beliefs about inheritance and kinship that are based on Western traditional concepts and recommend that during the recording of family history, management, and referral to genetic counselling, recognition is made of the patrilineal concept of kinship prevalent in this community. In patrilineal kinship systems, a kindred is defined chiefly by the male line of descent from a common ancestor and a disease "running in the family" may be construed as being derived directly from a common ancestor (Modell, 1997). This belief may have a significant impact on help-seeking behaviour.

2.6.1.1 The classification of kin

In the western world the term “father” is reserved for a man’s biological father and the brothers of both mother and father are referred to as uncles. The Iroquois system (after the well-known North American Indian tribe which also classify in this way) is an anthropological classificatory system that classifies father and father’s brother together as father and uses a separate term for the mother’s brother. This system is found among

all southern Nguni and the related kinship terminology works with precise logic (Hammond-Tooke, 1993)

He describes it as follows:

> "If you call a man 'father' (even if he is in reality your father's brother) you will call his children (your parallel cousins) 'brother' and 'sister', and will be prohibited from marrying them. You will also treat him with respect due to a father."

In fact, the entire father's male kin were regarded as belonging to the same group. Consequently the father's brother's wife was called 'mother' and his children (cousins) 'brother' and 'sister'. The same does not apply to the mother's side of the family. The mother's brother, for instance, was not called father, but addressed by quite a different term, one meaning 'male mother'.

A similar difference existed between the mother's sister and the father's sister. The mother's sister was called 'mother' and her children 'brothers' and 'sisters', whereas the father's sister had a special status, was accorded extreme respect and was called 'female father'. Children of the maternal uncle, however, were again not referred to as brother and sister. Among the Nguni they were called *mzala*, which can be translated as 'cross-cousin'. Among Sotho and Venda, the mother's brother's daughter was a preferred marriage mate, but Nguni prohibited marriage with all maternal and paternal cousins (Hammond-Tooke, 1993).

This can obviously cause confusion with individuals who are not familiar with this concept. Genetic counsellors and doctors who are trying to construct a pedigree should be especially aware of this system of kinship and of possible misunderstandings.

2.6.2 Western society

In 1996, Richards and Ponder proposed that a more general 'genetic literacy' within the public would be valuable for the appropriate use of predictive or diagnostic genetic tests. Studies in western countries have shown that lay understanding of scientific genetics is very limited (Richards and Ponder, 1996).

Human interest in and concern around inheritance is very old and it is reasonable to assume that ideas about physical resemblances and behavioural characteristics common to families are as old as concepts of family and kinship (Richards, 1998). Lay knowledge about inheritance is closely related to ideas about kinship and social relatedness between family members (Richards, 1996) and there has always been a great interest in and good knowledge of inheritance of family characteristics. With any newborn baby there tend to be many family discussions as to who the new baby takes after. This knowledge of inheritance is part of western family culture and Richards and Ponder (1996) showed that the Mendelian explanation may conflict with the lay knowledge of inheritance and that this pre-existing lay knowledge sometimes makes the understanding and acceptance of the scientific account more difficult. Lay knowledge, while it accepts that a condition is inherited, does not usually include the notion that a gene mutation is the cause of the condition. Recessive inheritance is especially puzzling because the condition is not expressed in the parents, so how can it be passed on to the children? (Richards, 1996).

Aspects of inheritance, according to family culture, include physical features, aspects of behaviour, character, personality, mannerisms and habits as well as health and proneness to illness. There is also the belief that we are likely to resemble those that are closest to us in kin terms, which in western kinship will mean our parents and our children. Thus the Mendelian concepts do not always fit in with what we know (Richards and Ponder, 1996). In a study on cultural aspects of cancer genetics, Meiser et al. (2001) have established that there is a general belief amongst families with inherited diseases that the disease may skip a generation; that there will be a resemblance (physical or in character)

between those in the family who are affected and those who are not and that a person is likely to develop the disease at the same age as the affected family member.

In western culture kinship is described as bilateral, which acknowledges inheritance from both maternal and paternal lines with both sides having equal status (Richards, 1996). The public's knowledge and beliefs about inheritance are supposedly organised around these reference points and it is usually assumed that characteristics may be inherited from either side of the family. For a large part these bilateral kinship systems do thus correspond to the descent models of clinical genetics which makes it generally easier for patients from western cultures to assimilate knowledge about genetics (Davison, 1997).

It has previously been mentioned that there is a lack of genetic services for patients with haemophilia in South Africa. The literature indicates that the counselling services, if available, must be culturally-sensitive and adapted to the intrinsic knowledge of the multicultural clients. It must further take into account how different kinship systems may influence, if at all, their clients' understanding and beliefs regarding genetic inheritance.

2.7 HIV/AIDS in South Africa

The human acquired immune deficiency syndrome (AIDS) is caused by an infection with the human immunodeficiency virus (HIV). The HI-virus is a RNA-virus belonging to the retrovirus group. Retroviruses integrate their genetic code into the genotype of the hosts's cells and viral RNA is assimilated into the double-stranded structure of DNA (Kirch, 2008). People who are HIV-positive are initially asymptomatic and on average only develop clinical symptoms after 8-10 years. Because AIDS has a weakening effect on the immune system infected persons have an increased risk of contracting opportunistic infections such as TB or pneumonia especially during the later stages of the disease (Kallings, 2008).

The HI-virus is primarily found in the blood, semen, and/or vaginal fluid of an infected person and is mainly transmitted through having unprotected sex with an infected person, sharing needles, syringes and other equipment with someone infected with HIV, or through being exposed to HIV before or during birth or through breast feeding (UNAIDS, 2009). South Africa is currently experiencing one of the most severe AIDS epidemics in the world, with more people living with HIV than any other country (AVERT, 2009). At the end of 2007, there were approximately 5.7 million people living with HIV in South Africa, and almost 1,000 AIDS deaths occurring every day (UNAIDS, 2009).

The reduction in the workforce, the increased human and financial resources required to provide care for people living with AIDS, and the needs of orphans will profoundly affect all aspects of social and economic development and the lives and well-being of all. (Benatar, 2004).

In previous years a perplexing debate about the cause of AIDS in South Africa as well as a large amount of misinformation has hampered efforts to increase access to treatment, and has also created a climate of confusion in which prejudice towards people living with HIV thrived. A study in 2002 revealed that one in ten respondents who had revealed their HIV-positive status were met with outright hostility and rejection (Avert, 2009). In an effort to challenge the stigma that surrounds HIV infection, Nelson Mandela publicised the cause of his son's death who died of AIDS in 2005:

> *"Let us give publicity to HIV/AIDS and not hide it, because [that is] the only way to make it appear like a normal illness."*

Goffman (1963) defined stigma as "an attribute that is significantly discrediting and a stigmatised person as one who possesses an undesired difference:

> *" Stigma is conceptualised by society on the basis of what constitute difference, and it is applied by the society through rules and sanctions towards the affected individual or group. It is in this context that attitudes and perceptions about HIV*

> *may have been formed and enacted to alienate and discredit people those who are seen as more likely to be affected by the disease than other"* (Goffman, 1963)

Stigmatisation is the result of the fear and anxiety that local South Africans display towards the disease and leads to fear and anxiety amongst those who have been stigmatized (Iboko, 2006).

Chapter 3: Methodology

In this chapter the methodological process is described and issues pertaining to the research are discussed. The reasons for having selected particular methodological choices are provided, potential sources of bias are identified and attempts at minimising these are described in the relevant sections.

3.1 Research Design

A consequence of South Africa's ethnic diversity is the challenge of communication within the medical setting. The purpose of this study is to describe and evaluate the basic understanding of genetic inheritance amongst mothers and caregivers of patients with haemophilia within a cultural subgroup in South Africa in order to heighten awareness of medical workers, counsellors and interpreters.

An exploratory qualitative research design using phenomenological methods was adopted. Qualitative research has been selected because it offers insight into emotional and experiential phenomena in health care (Giacomini and Cook, 2000). It is concerned with the meanings people attach to their experiences of the social world and how people make sense of their world (Pope and Mays, 2000). Qualitative methods permit thorough inquiry into selected issues with careful attention to detail, context and nuance (Patton, 2002). This type of inquiry focuses in depth on relatively small samples, selected purposefully.

In general, qualitative inquiry is an in-depth study that uses face-to-face techniques to collect data from people in their natural setting. Data for this qualitative study were generated from in-depth interviews to elucidate what was experienced, how it was experienced and what meaning the participants assigned to the experiences. All data, including participants' observation and audio-records, were collected in the form of

words rather than, as would be in qualitative research, in numbers (McMillan and Schumacher, 2001; Patton, 2002; Creswell, 2007).

Another key difference between quantitative and qualitative methods is flexibility (Ely et al., 1991). Generally, quantitative methods ask all participants identical, 'close-ended' or fixed questions in the same sequence. This inflexibility allows for meaningful comparison of responses across participants and study sites and allows for measurement and interpretation of the relationship between dependent and independent variables (McMillan and Schumacher, 2001). Qualitative methods however require flexibility. This adaptability permitted an element of spontaneity in the interaction between the interviewer and the study participants and allowed for adaptation of the interview protocol as new information become available. Because the questions were typically open-ended, participants had the opportunity to respond elaborately and in great detail and the interviewer could react by tailoring subsequent questions to the information provided by the participants. As a consequence of this more relaxed interviewing technique, the relationship between the interviewer and the participants was less formal than it would have been in a quantitative research design (Patton, 2002; Ely et al., 1991).

3.1.1 Phenomenological Research

Orlikowski and Baroudi (1991) suggest three research paradigms for qualitative research: positivist, interpretive and critical. A similar three-fold classification has been adopted by Fossey et al. (2002), namely empirico-analytical, interpretive and critical.

This study made use of an interpretive research paradigm where the focus is primarily on understanding and accounting for the meaning of human experiences and actions (Fossey et al., 2002). Interpretive methodologies include ethnography, phenomenology and narrative approaches, with each mode of enquiry addressing the issue of meaning from a slightly different standpoint (Fossey et al., 2002).

As a specific interpretive methodology, phenomenological inquiry was selected, as it

focuses, with a greater degree of attention than other qualitative research, on three distinctions:

a) the non-assuming and non-intervening study of a personally or socially significant phenomenon

b) which is investigated as a natural experience without attempting to generate theories or models

c) with the goal of understanding characteristic and essential themes (Patton, 2002; van Manen, 1990; Ploeg, 1999).

The origins of phenomenology are in philosophy, particularly the works of Husserl, Heidegger, and Merleau-Ponty (van Manen, 1990). The primary aim of this type of enquiry is to gain a deeper understanding of how people experience their world and what it is like for them. Data is mostly collected by means of in-depth interviews to elucidate what was experienced, how it was experienced and what meanings the participants assign to the experiences (McMillan and Schumacher, 2001; Patton, 2002; Creswell, 2007). This type of enquiry allowed the non-assuming and non-intervening study of how this group of isiXhosa-speakers experience living with and caring for patients with haemophilia, how they made sense of their experiences, and how they made sense of, or understood, genetic inheritance.

The central purpose of any qualitative methodology is to explain and understand before attempting to predict and solve problems (McMillan and Schumacher, 2001). For this study, in particular, it was not appropriate to predict results or to prove a specific hypothesis because it was structured around an exploratory framework. Instead the phenomenon of experiences and understanding was described as a natural experience by analysing the words that constituted the narratives (van Manen, 1990; Ploeg, 1999; McMillan and Schumacher, 2001).

3.2 Participants

3.2.1 Study population

The Western Cape is divided into five district municipalities, one of which is the City of Cape Town Metropole. According to the 2001 census the majority of the population of approximately 4.5 million is made up citizens of Mixed Ancestry (also referred to as Coloured), followed by Black Africans, Whites and citizens of Asian/Indian origin (Fig. 3) (City of Cape Town, 2009).

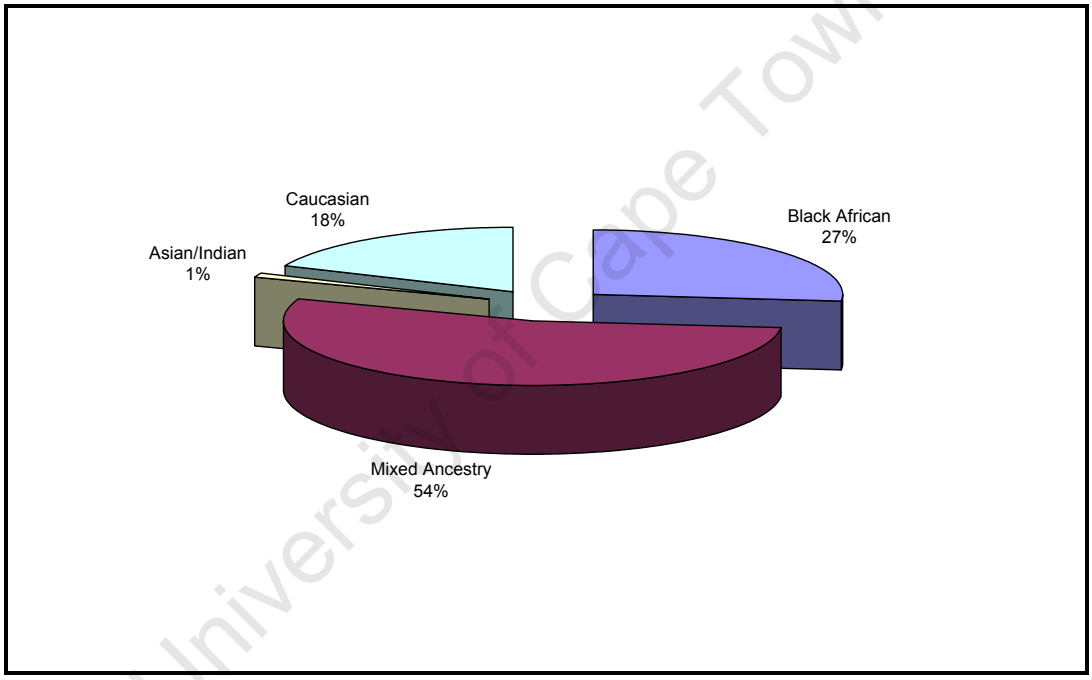


Figure 3: Cape Town's demographic profile in terms of population groups.

The predominant home language in the Cape Town Metropole is Afrikaans, followed by isiXhosa and English (Fig. 4) (City of Cape Town, 2009).

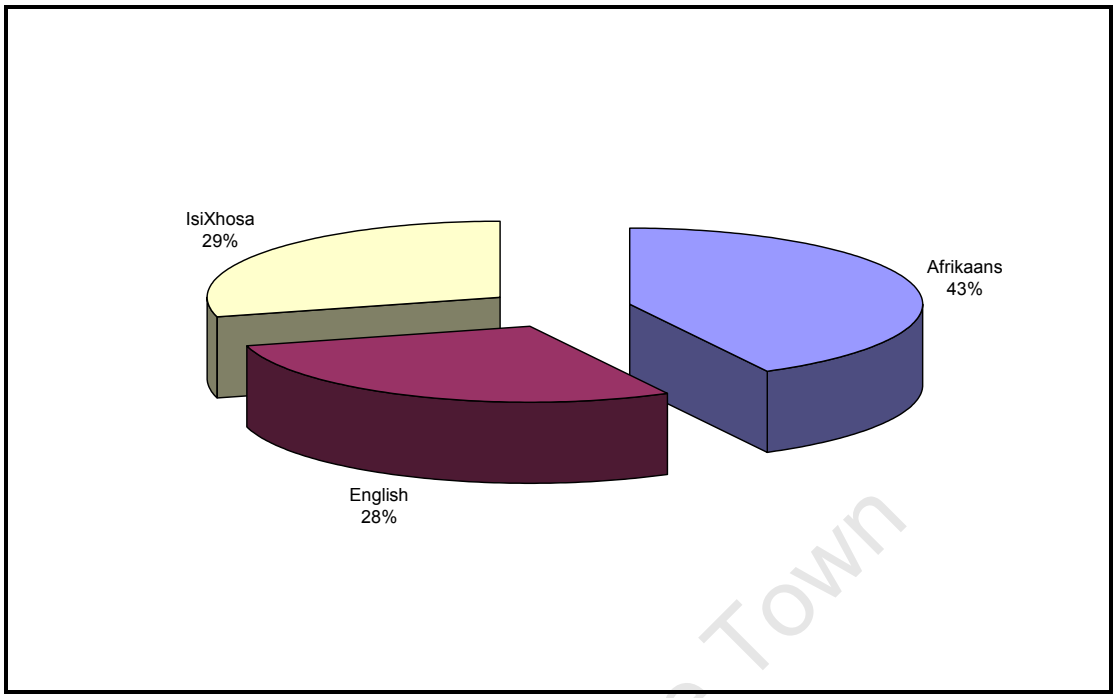


Figure 4: Cape Town's demographic profile in terms of home language distribution

Because this study sought to explore culturally determined beliefs or attitudes towards haemophilia within the study population it was necessary to define the cultural group. Cultural beliefs may change and evolve depending on the circumstances of a particular community. This is well illustrated by the fact that beliefs may be considerably different between Hindus born and educated in Britain and those who recently migrated (Sensky, 1996). The same may be true for isiXhosa speakers living in rural areas and those that are living in and around major cities.

3.2.2 Sampling strategy and sample size

A combination of purposeful and convenience sampling was selected where the purposeful selection of participants represents a key decision point in a phenomenological study (Creswell, 2007; Patton, 2002). Purposeful sampling selects cases for study that are information-rich and that offer useful manifestations of the phenomenon of interest (Patton, 2002). To fulfil these requirements, participants were selected who belonged to the cultural group in question (first-language Xhosa speakers

living in the greater Cape Town area), had knowledge of the phenomenon being researched (taking care of someone with haemophilia), and had the ability to verbalise their experiences of it while being willing to take part in the study.

The following factors were considered when caretakers of patients with haemophilia were selected as the sample group:

- Haemophilia is usually diagnosed at, or shortly after birth, and mothers/caregivers, because of the nature of the condition, will usually visit specialist clinics relatively frequently.
- Regular and frequent contact with clinic staff should ensure that the caregivers have a reasonable understanding of the condition, its cause and its inheritance patterns.
- Because the treatment for haemophilia is normally regular and ongoing, it is reasonable to expect a good understanding of why and how treatment is given at the clinics.
- It can furthermore be assumed that caretakers of patients with haemophilia have over time developed their own understanding or theories of how and why their family members suffer from this condition.

Besides purposeful sampling, this study also employed an element of convenience sampling by approaching caretakers who attend the monthly support group meetings. Convenience sampling involves the selection of the most accessible subjects and is generally regarded as the least rigorous technique (Marshall, 1996), because it involves choosing the nearest and most convenient subjects to act as participants. Individuals who met the sampling criteria were approached at the monthly Haemophilia Support Group meetings in Khayelitsha, which is within easy reach of Cape Town. Support group meetings are generally held on the last Saturday of each month. These meetings are coordinated by a nurse who is employed by the Haemophilia Foundation of South Africa. All other participants are volunteers and are mostly isiXhosa-speakers. Support group meetings in Khayelitsha are usually held in a tearoom at Site B Day Hospital. Volunteers endeavour to contact all patients who attend the haemophilia clinic, and who

are known to the co-ordinating nurse before a meeting. The Co-ordinator always provides snacks (tea, coffee, cool drinks, doughnuts, bread, fruit, potato chips etc) and meetings are scheduled to start at 10 a.m. Unfortunately, attendance is often poor and volunteers become despondent. Poor attendance may be the result of transport costs or the timing of the support group meetings. Meetings are usually scheduled for a Saturday morning, which may be inconvenient for, especially, single working mothers who use that time to do shopping or weekly chores.

Ten caregivers of patients with haemophilia were interviewed. Amongst this group were six mothers, two aunts and two grandmothers of affected boys.

3.2.3 Inclusion and exclusion criteria

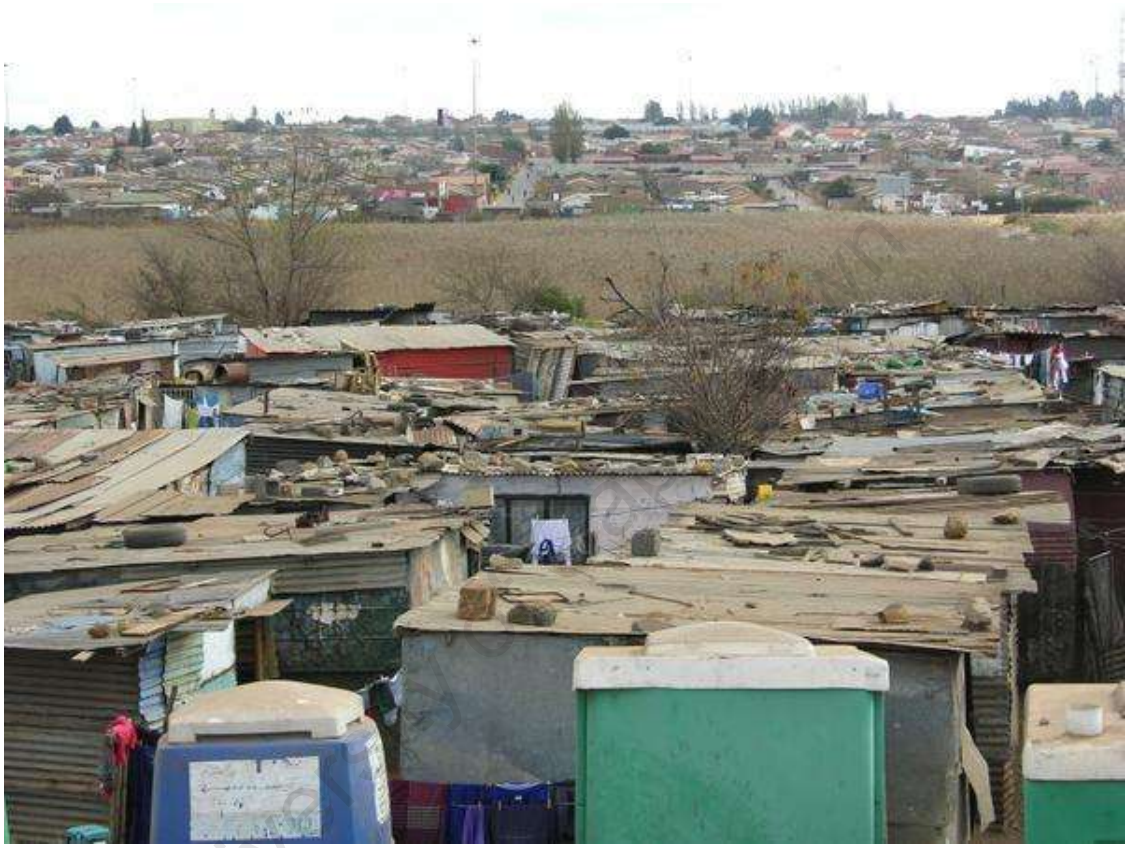
Inclusion criteria

- Only Xhosa-speaking mothers/caregivers of boys with haemophilia A or haemophilia B.
- Mothers/caregivers over the age of 21.
- Mothers/caregivers who attend the Khaya Rock Support Group and/or the regular haemophilia clinics at the Red Cross War Memorial Children's Hospital (RCWMCH) and Groote Schuur Hospital (GSH).
- Mothers/caregivers who were willing to be interviewed by an interpreter, with the investigator being present.
- Mothers/caregivers who were willing to sign informed consent, including that the interview will be tape-recorded.

3.3 Research Setting

The study was undertaken mainly on the Cape Flats in Khayelitsha, one of the biggest black townships in the country. The Cape Flats are a large, flat area on the Cape Peninsula where numerous housing projects were established by the previous apartheid government. Khayelitsha is demarcated as part of the Tygerberg Substructure and falls

under the jurisdiction of the Cape Town Metropolitan Council (McDonald and Smith, 2004). The township is made up of different kinds of dwellings including several areas of informal settlement where people live informally and shelters are generally made of used scrap metal, corrugated metal sheets, and even plastic (Photograph 1).



Photograph 1: Informal settlement

Three interviews were conducted in a tearoom at the Site B Day Hospital and six in the homes of participants in and around Khayelitsha. The interview venues were chosen by the participants depending on what was most convenient to them. Giving the participant the opportunity to choose the site can help them feel more empowered in their interaction with the interviewer and thus facilitate an easier, more relaxed exchange (Elwood and Martin, 2000). The tea room at the Day Hospital was familiar to the participants because regular haemophilia support group meetings are held at this venue (Photograph 2). Permission to use this room for interviews was obtained from the hospital superintendent via a nurse at the hospital who also volunteers at the support

group meetings. The tea room is a large, pleasant room with a comfortable seating area that lends itself well for interviews.

Photograph 2: Tea room at Site B Day Hospital during a support group meeting.

The home interview venues included small, formal houses, informal shacks and a shebeen. Shebeens are a type of drinking establishment that are often run by local people in private houses. The home environment offered more insight into the day-to-day lives of the participants and allowed the researcher to take detailed notes of relationships between family members and their environment. Kvale (2006) reminds that the interview is a hierarchical relationship with an unbalanced power distribution between the interviewer and the participant. The interview serves the interviewer, contains hidden agendas, and can potentially be a manipulative dialogue that gives the interviewer the sole power of interpretation. The home interviews helped balance this skewed power distribution, to some degree, in favour of the participants. Hopefully this made them feel safer and more empowered in their home environment, making it easier for them to discuss difficult and sensitive issues.

3.4 Data collection

3.4.1 Research tool

Interviews were chosen as the means of collecting data. Interviews are the most commonly used method by which data are collected in phenomenological studies as they allow the researcher to capture the perspective of participants and to explore their views, experiences and beliefs (Moustakas, 1994; Kwan-Gett, 1995; Genzuk, 2003). Interviews provided the means of asking the right question in the right ways and helped the researcher to see the world through the eyes of the participants (Westby et al., 2003). Within the phenomenological paradigm, the investigator endeavoured to gather data in ways which were minimally intrusive and allowed the participants to make the biggest contribution. Field observations were used to capture social phenomena directly and without judgement or contribution by the researcher (Seymor and Clark, 1998).

Phenomenological interviewing techniques in this study attempted to uncover how participants experienced living with haemophilia on a day-to-day basis and whether and how culture influenced their means of dealing with haemophilia as well as their understanding of basic genetics (Sorrell and Redmond, 1995). The interpreter/interviewer needed to treat the lay informants as the experts, relinquishing control to the participants and allowing the participants to be the teacher. At the same time the interpreter/interviewer maintained a friendly conversational tone while noting the information being collected (Kwan-Gett, 1995; Westby et al., 2003).

3.4.2 Design of research tool

A semi-structured interview schedule was developed that included close-ended and open-ended questions (Pope and Mays, 2000). Close-ended questions were used to obtain demographic information and data that could be easily categorized. Open-ended questions were formulated to encourage free responses and allow the participants to tell their story. The semi-structured interview schedule allowed for the addition, exclusion or variation of the wording of particular questions, in response to the participants, albeit without changing the meaning of the questions (Murray and Rhodes, 2005).

A list of questions was developed that provided a provisional structure to the schedule. Questions were broadly based on medical anthropologist and psychiatrist Arthur Kleinman's eight questions (Table. 3). He devised these questions to uncover the participant's perspective and explanatory model of a condition (Kleinman, 1980, Kleinman, 1976).

This first draft of questions was reviewed and discussed in a focus group meeting consisting of the investigator, two project supervisors, a lay isiXhosa first-language speaker as well as an isiXhosa first-language speaker with a background in science. The objective of the focus group was to ensure that the questions would be easily understood once translated into isiXhosa and that they would be sensitive to the culture of the participants.

Table 4: Arthur Kleinman's (1976) eight questions

Arthur Kleinman's eight questions:
1. What do you call the problem?
2. What do you think has caused the problem?
3. Why do you think it started when it did?
4. What do you think the sickness does? How does it work?
5. How severe is the sickness? Will it have a short or long course?
6. What kind of treatment do you think the patient should receive? What are the most important results you hope he/she receives from this treatment?
7. What are the chief problems the sickness has caused?
8. What do you fear most about the sickness?"

Structured questions appeared at the beginning of the interview schedule and included demographic data such as name, clan name, age, marital status, relationship to patient, level of education and previous knowledge of haemophilia. The first part of the semi-structured questions sought to explore when the participants first became aware of the condition, what their understanding and concerns were as well as their own interpretation of the condition. The second part focused on how they understood the genetic implications of haemophilia on family history, how they communicated to their extended family and their community, and on inheritance in general.

The questionnaire was initially formulated in English after which it was translated into Xhosa by the interpreter who also conducted the interviews.

3.4.3 Interviews conducted with caregivers

Consent forms and information sheets were either handed to the participants by the researcher during support group meetings, delivered to their home a few days before the interview or, in one case, faxed at the participant's request. Participants were contacted telephonically by the isiXhosa-speaking interpreter to schedule the appointments and to determine what venue would be most convenient to the participants.

3.4.3.1 Interviewing process

The interpreter introduced herself and the researcher. During the introduction the format of the interview was explained and permission to use a tape recorder and notebook were obtained. The approximate duration of the interview was discussed and assurances of confidentiality and anonymity were given. The interpreter/interviewer endeavoured to create a climate in which the participants felt comfortable and free to respond honestly and in detail (Moustakas, 1994). She established rapport through positive verbal and nonverbal encouragement. Good eye contact, smiling and open body language ensured that the participant felt comfortable with the process and sufficiently relaxed to 'talk back'. The consent forms were discussed and the interpreter offered to go through them again or read them out aloud. Participants were reassured that they were free to ask questions or to withdraw from the interview at any time. The interpreter conducted the interview over approximately 45 min to an hour using the interview schedule (Appendix vi).

3.4.3.2 Role of researcher

The researcher does not speak isiXhosa and in order to overcome language barriers and to allow participants to freely converse in their own language, a first language Xhosa-speaking interpreter was employed. The researcher was present at all interviews and

acted as an observer, taking notes of the interview process, of surroundings and, if applicable, interactions between family members. Interviews are not just instruments to gather information but also an opportunity for participant observation (Elwood and Martin, 2000) and additional pertinent information such as nonverbal behaviour or interruptions were noted.

The participants of this study group were all first language Xhosa-speakers. IsiXhosa-speakers residing in and around Cape Town are not necessarily proficient in English and may hold different health concepts, values, and beliefs from the researcher, who is not a member of the studied population and is not proficient in the participants' language and culture (Tsai et al., 2004). Ultimately, the data generated from interviews are words and words can take on a very different meaning in other cultures, while misunderstandings may occur even between persons that speak the same language (Patton, 2002). Researchers are not in the field to judge or change values and norms, but to understand the perspectives of others, which requires special sensitivity to another culture as well as respect for differences (Patton, 2002).

3.4.3.3 Role of interpreter

The interpreter for this study was in her final year of a BPsych degree in Health Sciences and Social Services with specialisation in Counselling Psychology at the University of South Africa (UNISA). She has further completed a six-month theoretical course in HIV/AIDS Care and Counselling with UNISA.

In this study the interpreter played an active role and conducted most of the interview. In order to keep the dialogue between interpreter and participant flowing, it was decided that the interviews be conducted in Xhosa, without interpreting back and forth between the various participants. This gave more responsibility to the interpreter, but it allowed her to engage with and build a better relationship with the participants and allowed the interviews to progress without interruption. A disadvantage was reduced control from the researcher's perspective. It also required the interpreter to quickly become familiar

with the aims of the interview and interview schedule**.** The researcher was always present and as the interpreter became aware of key points she would summarise those points to the researcher to allow for additional questions to be asked**.** The interpreter was empathetic and friendly and had an easy and good relationship with the participants. She also developed an understanding of probing questions.

Pitchforth and van Teijlingen (2005) refer to a passive and an active role for the interpreter. In the passive model the researcher will ask a question (for example in English) and the interpreter will interpret the question to the participant and the response to the researcher. This is very time-consuming and the researchers found that this method does not allow the conversation between participants to flow. The dialogue was constantly interrupted by interpretations and the interviews tended to become disjointed. It has also been shown that interpreters often want to be helpful by summarizing and explaining responses and it can become unclear whose perceptions are actually being presented. On the other hand there are words and ideas that simply cannot be translated directly and some languages lack words, especially for new, scientific concepts like 'DNA', 'genes', 'genome' and 'genetic predisposition' (Patton, 2002).

3.5 Stages of the study

3.5.1 Pilot study

A pilot study was conducted to assess whether the interview questions were clear and unambiguous and to estimate how long it would take to complete the interview (McMillan and Schumacher, 2001). The participant was an isiXhosa speaking caregiver of a patient with haemophilia who was also relatively proficient in English. It became evident during this interview that continuous interruptions by translations into or explanations in English were very disruptive. Even though the participant spoke English well, she preferred to speak in her mother tongue, especially when sensitive issues were broached. The interview was short, lasting only about 30 minutes and it became clear that probing methods had to be improved.

3.5.2 Adjustments to Interview schedule

The interview schedule was developed to ensure that the questions were culturally-sensitive and to allow the participants to tell their story in their own time and sequence. A semi-structured interview schedule not only requires a good understanding of the research aims, but also that decisions be made regarding the need for follow-up questions. Ultimately the decision to decide whether or not a question had been sufficiently answered lay with the interpreter. Prompted by the outcome of the pilot study, researcher and interpreter discussed probing questions and practiced probing through role play.

The style of the interpreter/interviewer could, however, only be judged at a later stage, after reading some of the transcripts. There seemed to be a tendency to rush the interviews and to occasionally ask two questions in quick succession or to ask leading questions. Observations by the researcher further suggested that the interpreter/interviewer sometimes tended to ask the next question too quickly without allowing the participant enough time to reflect before answering. Reading through of the interviews also showed that not enough probing questions were asked. The researcher discussed these issues regularly with the interpreter/interviewer and the interviewing techniques constantly improved. It was, however, challenging for the researcher, who does not speak or understand Xhosa, not to be able to give guidance or to intervene during the interview. To facilitate the interviewing process, the researcher wrote interviewing guidelines (Appendix vii) that emphasised, amongst others, the significance of probing questions and issues relating to conduct. The researcher was aware that each interview should ideally be transcribed and translated as soon as possible in order to adjust questions and the style of interviewing. However, problems with transcriptions, which will be discussed under the next heading, resulted in long delays and backlogs.

3.5.3 Adjustments to the transcription/translation process

As discussed in Chapter 2, a translator should ideally be a native speaker of the target language (Translation Working Group, www.eurolight-online.eu). It was however not possible to find and employ a native English speaker who is fluent in ixiXhosa for this study. Problems associated with the process of translation, including possible loss of meaning, lack of equivalent terms and emphasis that has been added or deleted have also been mentioned earlier (Crawford, 1999). Mkoka (2003) translated a widely used measure of health-related quality of life questionnaire from English into Xhosa, and found that concepts which appeared to be simple, nevertheless proved to be difficult to translate. An important basic requirement for a translation is that it is semantically equivalent and conveys the same meaning as the original.

Limited funding, unfortunately, restricted the use of professional transcribers and translators and these processes were thus performed by lay people. Qualifications of the transcribers involved in this study are listed in Table 5.

Table 5: Qualifications of transcribers (T) involved in the project

Transcriber	Qualifications
T1	BCom, Experience in data capture and technical production of research
T2	B-Tech in Journalism, some experience in translating from English to isiXhosa
T3	BSoc, Postgrad Diploma in Organisation and Management

Transcription of the audio-taped interviews proved to be a protracted process. It was difficult to find part-time, yet reliable and experienced, isiXhosa transcribers. The first person employed to transcribe took over five months to transcribe four interviews. After receiving the first two transcripts it was found that she had not transcribed the isiXhosa text but had translated directly into English. It was consequently only possible to verify the translation while listening to the tapes. Spot-checks by the interpreter revealed that some parts had been shortened and summarized. Before handing over the next two interviews, the researcher wrote transcription guidelines (Appendix viii) to address these problems, to highlight some transcription conventions and to emphasize the importance of transcribing verbatim. These transcription guidelines were discussed with the

transcriber via telephone and via email. When the next two transcriptions were finally finished three months later, the researcher was astonished to find that the transcriber had once again not transcribed the original Xhosa text and had again translated directly into English.

A new part-time transcriber was found who worked much faster, transcribed verbatim into isiXhosa and translated into English as well. To verify the translations of the first transcriber, she transcribed two previously-translated interviews into isiXhosa and translated the transcripts into English. Although the wording was different, no major differences in the content meaning were found. The following two excerpts illustrate the two different methods:

Translation as per transcriber 1:

> "*You know my father is allergic to red meat, but sometimes he eats it and does not react on in. He passed his allergy to everyone at home and am the only one who does not have allergy."*

Translation as per transcriber 2:

> *"You know back home in my family, my dad is allergic to red meat but sometimes he would eat it sometimes and nothing would happen to him (health wise). All my siblings inherited dad's allergy. I am the luckiest one. I've never encountered such problem. I am not allergic (to red meat) but my siblings are."*

The second transcriber did, however, find that the work was too time-consuming and was reluctant to take on any more transcriptions. This made it necessary to find yet another transcriber. The third transcriber was reasonably fast, transcribed the audiotapes and translated the transcripts into English. Unfortunately, her personal circumstances changed and the transcription of the last interview took more than two months.

3.5.4 Scheduling of appointments

Participants were reluctant to speak to the researcher (in English) on the phone and to make commitments to interviews because of language issues. The researcher was thus dependent on the interpreter to make appointments. This turned out to be a lengthy process, firstly because as the interpreter was fully employed, she could only try and contact participants during her lunch hour or after work and, secondly, because many households do not have land-lines and depend on mobile phones. These mobile phones are however sometimes not operating because the owner cannot afford to buy airtime. On other occasions cell phones got stolen and contact numbers were lost altogether. It sometimes took days and weeks to get one firm appointment for an interview and the researcher had to constantly follow up and prompt the interpreter to retry.

3.5.5 Security issues

Most interviews took place in people's homes in townships around Cape Town. Some were in volatile areas and the interpreter felt obliged to bring a male escort who would wait outside and guard the car. One interview took place in the Brown's Farm squatter camp in Philippi where the roads are very narrow and not designed for cars (Photograph 3). In this instance the car had to be left on the outskirts of the settlement and researcher and interpreter went by foot into the settlement.

Photograph 3: Entrance to the Brown's Farm squatter camp in Philippi

3.6 Data capture

All interviews were audio-recorded with the permission of the participants. The researcher tested the equipment before the interviews and ensured that spare batteries and tapes were available. As far as possible, background noise and interruptions during the interviews were avoided. This was not always possible, in two cases because of the nature of some informal dwellings that were built of corrugated sheeting in close proximity to each other, and in one case where the house served as a shebeen and loud music was playing.

Audio-taped recordings were transcribed into isiXhosa and translated by a transcriber. Participants' names were encoded and data from the transcripts were captured on Excel spreadsheets under initial headings representing meaning units.

3.7 Analysis/Interpretation

A basic decision when using content analysis was selecting the unit of analysis (Graneheim and Lundman, 2004). The most suitable unit of analysis for this study was the interviews as these contained the text about experiences and beliefs around haemophilia and inheritance (Graneheim and Lundman, 2004). In the literature the unit of analysis is referred to as individual people, clients, a community, state or nation, programs (Patton, 2002), interviews or interactions under study (Downe-Wamboldt, 1992 in Graneheim and Lundman, 2004). Patton (2002) states that "the key issue in selection and making decisions about the appropriate unit of analysis is to decide what it is you want to be able to say something about at the end of the study".

The interviews were read and re-read to become familiar with the data and to develop a sense of the whole (Graneheim and Lundman, 2004; Patton, 2002). The interview text was sorted into content areas that were broadly related to the interview questions. From these blocks of text meaning units were developed. These were then condensed and labelled with a code (Fig. 5).

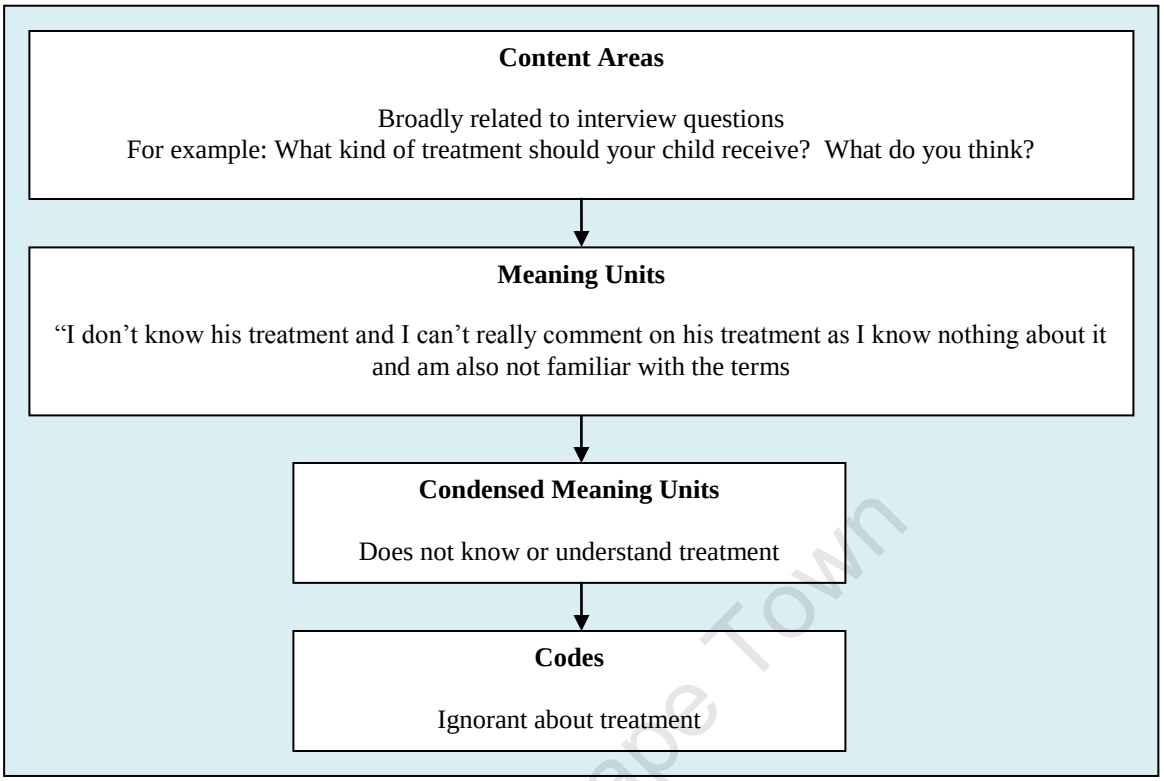


Figure 5: Flow diagram depicting an example of the initial stages involved in data analysis

The codes were sorted into sub-categories and categories from which ultimately themes were formulated (Fig. 6).

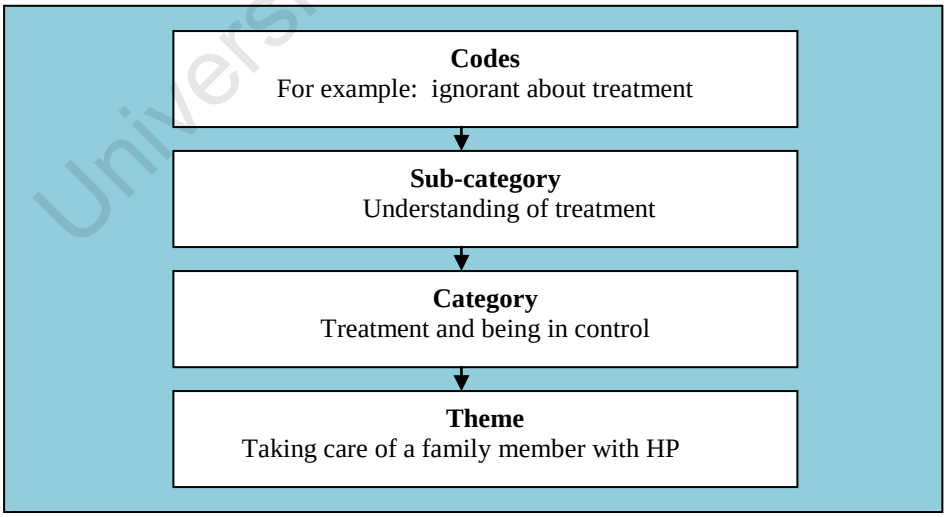


Figure 6: Flow diagram depicting the final stages involved in data analysis

During data coding, notes were made about how the coding process had been conducted and how decisions were reached.

3.8 Trustworthiness

3.8.1 Reflexivity

Reflexivity requires the researcher to acknowledge that it is hardly possible to remain 'outside of' one's subject matter while conducting the research (Nightingale and Cromby, 1999). It requires the examination of the self as instrument and compels the researcher to take into account how his/her personality and presence influence the study (Ely et al., 1991; Ryan, 2005), and to recognize that one's subjectivity is intimately involved in scientific research (Ratner, 2002).

Willig (2001) makes a distinction between personal reflexivity and epistemological reflexivity. Personal reflexivity involves reflecting upon the ways in which the researcher has shaped the research (Willig, 2001; Ryan, 2005). In order to examine the self and to deal with issues pertaining to the researcher's motives, background and perspectives in this study, the researcher self-reflected and wrote a brief description of experiences with the phenomenon of interest and the researcher's own racist attitudes and background (Appendix ix). This helped to set aside her own personal experiences and to keep the focus on the participants (Malterud, 2001; McMillan and Schumacher 2001). Various studies have emphasized the importance of critical self-awareness towards developing multicultural competence (Carter, 1991; Richardson and Molinaro, 1996). Sue et al., (1992), and Ancis and Szymonski (2001) further emphasise that self-awareness includes admission of ones own racist attitudes and beliefs. Throughout the study and especially during analysis the researcher continued to guard against premature interpretations of the data by ongoing reflection and awareness of her own subjectivity (Patton, 2002).

'Epistemological reflexivity' encourages the researcher to critically evaluate how the research question could have defined and limited the results and whether the design of

the study and method of analysis has influenced the data and the findings (Willig, 2001). The main aim of this study was to explore the basic understanding of genetic inheritance amongst isiXhosa speaking caregivers of patients with haemophilia. The researcher hoped to learn how the process of inheritance is understood by the participants as well as their understanding of how and by whom traits are passed on from generation to generation.

This line of investigation looked at inheritance from a western and scientific point of view. Because the researcher was not familiar with amaXhosa culture, the questionnaire was discussed and developed in a focus group. The researcher explained the aim of the questions and received input from project supervisors, an isiXhosa lay person as well as from an isiXhosa social scientist on how to develop and formulate the questions in a culturally-sensitive way. The approach to the investigation did, however, not take possible limitations of the Xhosa language into account which did not allow the translation of modern scientific terms such as 'chromosomes' and 'genes'. The adopted line of questioning may also have intimidated some participants because they felt that they were being questioned, thereby affecting the richness or depth of the data. With a slightly different approach to the questions a different picture might have emerged. Instead of asking questions about inheritance, participants might have been encouraged to tell the story of their family and/or ancestors, highlighting the connection between everyone and how sickness might influence the relationships.

3.8.2 Reliability and validity

The researcher endeavoured to approach the research by being aware of, and bracketing preconceived ideas and biases. Further strategies to enhance design validity and confirmability of data included interim data analysis and corroboration to refine ideas, recording of descriptions of people and situations as well as the use of tape recorders and photographs. The reliability and validity of the translated transcripts was tested by the isiXhosa first language interpreter/interviewer to verify the original transcripts.

Validity in qualitative research rests on data collection and analysis techniques, the effect of an investigator on a study and the principles and consequences of sampling (Malterud, 2001; McMillan and Schumacher, 2001). Validity within the phenomenological research paradigm is not so much about exact replication of research findings, but more about the generation of a credible and insightful interpretative account of the research participants' views and their situations (Seymor and Clark, 1998). Although replicability is not an aim of phenomenological research, Marshall and Rossman (1999) emphasise the importance of clearly stating the design and methodology used, demonstrating how data were analysed, presenting the study in a scholarly context and keeping records available for reanalysis. This study used phenomenological methods as a tradition of enquiry and used rigorous data collection procedures involving tape-recorded, semi-structured interviews that were transcribed and analysed using qualitative content analysis.

Reliability and validity are conceived as trustworthiness, rigor and quality in qualitative research methodologies (Golafshani, 2003; Davies and Dodd, 2002). A researcher's background affects every aspect of the research, including decisions on what is to be investigated, how it is investigated, which method is the most adequate and which findings are considered most appropriate (Malterud, 2001).

3.8.3 Triangulation

Triangulation is a process or strategy whereby several information sources are used to corroborate key findings, to improve the validity and reliability and increase the chances of depth and accuracy of investigative finding (Russel and Gregory, 2003; Golafshani, 2003). Research strategies may include triangulation of data sources, methods, analysis and theory to reduce bias and increase credibility of investigative finding (Patton, 2002).

Forms of triangulation in this research included the use of a focus group to develop the interview schedule as well as the combination of interviews with observations. Observations were made by the researcher and included notes on

- distractions during the interviews
- whether and how the interviews were affected by others present
- characteristics of the participants
- observations regarding body language of interpreter as well as participants
- follow-up points of discussion with the interpreter

Process of constant analysis

Capture of data and selection of categories was corroborated by research supervisors. Thoughts, reflections, and conclusions were also regularly discussed with supervisors and the interpreter for critical evaluation. Different sections of the research were discussed with supervisors and experts in the field of Human Genetics, Anthropology and Linguistics. These discussions provided closely monitored feedback on the research process. Data were further verified through presentation and discussion of results during a research symposium as well as the studying of relevant literature.

3.9 Ethical considerations

Approval was obtained from the Departmental Research and the Faculty of Health Sciences Research Ethics Committee of the University of Cape Town before commencement of the study (Appendix i). Participants were approached by the researcher, in the presence of a haemophilia nurse and support group organisers at the monthly support group meetings. Information sheets and consent forms were handed to the participants at the meetings, delivered to their home a few days before the interview or faxed as per the participant's request. Participants were informed at recruitment and again before the interview, that tape recordings would be used. Signed consent was obtained from participants who had willingly agreed to an audio-taped interview. Photographs were taken only with explicit permission of the participants.

Information sheets that introduced the study and explained the purpose of the research as well as separate consent forms were formulated in easily comprehensible language

(Appendix ii and iv) and translated into isiXhosa (Appendix iii and v) To verify the translation, Information sheets were back-translated by an independent isiXhosa speaker. Participants were further assured that any personal information obtained during the interviews would be treated as highly confidential and that no names would be mentioned in any resulting scientific communication. Audio recordings would not be allowed out of the hands of the researcher carrying out the study unless explicit permission had previously been obtained from the participants.

Transcripts contained, amongst others, the demographic data of the participants and were kept in a safe place in the Division of Human Genetics. In general, data were kept confidential by encoding the identification of the individuals and only the researcher had access to the codes. The audiotapes will be destroyed after the study has been completed.

3.10 Limitations of the study

Because the researcher is not familiar with isiXhosa or the cultural life of the study group, it was necessary to employ an interpreter to conduct the interviews. This reduced control of the interview from the researcher's perspective, making it impossible to control the direction of the interview or to direct adequate probes. It also put extra pressure on the interpreter who had to quickly become familiar with the subject matter, the aims of the interview and the interview schedule.

A further limitation was the counselling experience of the investigator and the interpreter. Because participants did discuss sensitive and sometimes distressing experiences in their life, the interpreter and researcher endeavoured to remain sensitive to their emotional state throughout the interview. If, however, any issues needed to be dealt with in more detail, it was necessary to refer the participant for genetic counselling by an experienced counsellor.

The findings and discussion are presented in Chapter Four.

Chapter 4: Findings and Discussion

The order in which data are presented in this chapter roughly corresponds to the order of questions in the interview schedule. Participants' socio-demographic and biographical data from the structured part of the interview schedule are presented in Table 6. The cultural group in the current study was defined by first-language Xhosa speaking caregivers of patients with haemophilia living in the Cape Metropole. Most of the participants (8) had migrated to Cape Town from the Eastern Cape, two were born in the greater Cape Town area but still have family in the Eastern Cape. All participants reside in townships and have been living in the Cape Metropole for at least 4 years. Two families live in informal dwellings, or shacks (Photographs 6 and 7), and seven families live in formal brick houses (Photographs 4 and 5). All participants were female family members of the patients. There were six mothers between the ages of 30 and 50, two aunts, aged between 30 and 40 and two grandmothers, aged between 60 and 70.

Table 6: Socio demographic and biographical data of the participants

Code	Age	Relationship to patient	Prev. Res.	Pres.Res.	Educt. Grade	Years in CT	Urban settlements	
							House	Shack
C1	40-50	mother	EC	PH	3	9		X
C2	n.d.	mother	EC	PH	12	4	X	
C3	30-40	aunt	KA	KA	11	__	X	
C4	40-50	mother	EC	KA	12	11	X	
C5	n.d.	mother	n.d.	KA	n.d.	n.d.		
C6	30-40	aunt	EC	KA	12	15	X	
C7	60-70	grandmother	KA	KA	8	__	X	
C8	60-70	grandmother	EC	GU	none	27	X	
C9	40-50	mother	EC	KA	6	20	X	
C10	30-40	mother	EC	KA	10	19		X

EC: Eastern Cape; KA: Khayelitsha; GU: Guguletu; PH: Philippi; CT: Cape Town; Prev.:previous; Pres.: present; Res.: residence; Educt.: education; n.d.: no data

The researcher only became aware that demographic details for C5 were incomplete once the interviews had been transcribed. C5 was interviewed in the tea room of the day hospital and no information concerning her dwelling was recorded. Several attempts

were subsequently made by the interpreter to contact C5 telephonically, but without success.

Photograph 4: Self-owned brick house in Guguletu, a township 15km from Cape Town

Photograph 5: Inside a council-built brick house in Khayelitsha

Photograph 6: Front yard of an informal dwelling in Khayelitsha

Photograph 7: Inside an informal dwelling in the Brown's Farm squatter camp: Interpreter with patient and caregiver.

Data obtained from the open-ended questions are presented under headings representing themes that emerged after thorough content analysis. Data were initially grouped into 'content areas' that were related to the interview questions. Responses to those questions represented 'meaning units' that were condensed and then represented as 'codes'. Codes were grouped and sorted, from the bottom up, into sub-categories, categories until ultimately five themes emerged (Fig 7).

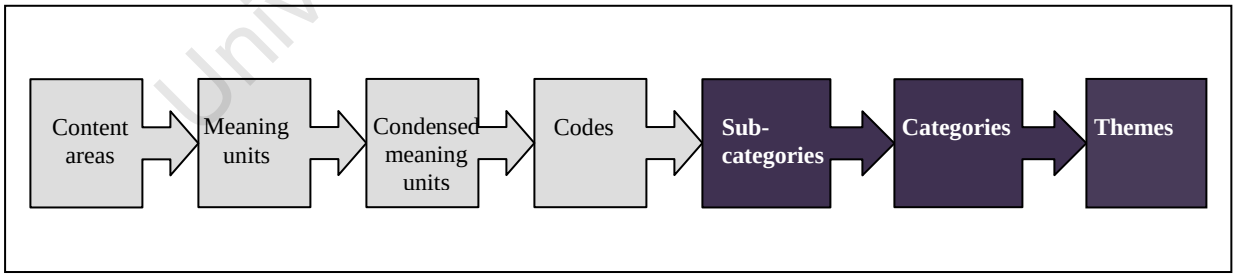


Figure 7: Flow diagram depicting the process of developing sub-categories, categories and themes.

Direct quotations by the participants are used throughout this chapter to illustrate themes and issues. An overview of sub-categories, categories and themes is presented in Table 7.

Table 7: Overview of sub-categories, categories and themes

	Categories	Themes
• Communication with community • Community support	Interaction with community	**1. Interaction with community and extended family**
• Communication with extended family	Interaction with extended family	
• Background knowledge of HP • Participants' experiences associated with HP symptoms • Participants' Experiences associated with HP care	Participants' experiences associated with HP	**2. Taking care of a family member with haemophilia**
• Understanding of treatment • Being in control	Treatment and being in control	
• Understanding of pathogenesis of HP	Perceived cause of HP	
• Inherited from whom? • Inheritance of illness	General inheritance	**3. Understanding of inheritance**
• Haemophilia • Family history of haemophilia	Inheritance of HP	
• Meaning of bleeding	Bleeding	**4. General and cultural beliefs**
• Rituals • Traditional healers	Traditional Medicine	
• What are genes and how are they passed on?	Understanding or knowledge of genes	**5. Genes**

4.1 Interaction with community and extended family

Questions were asked to elicit how much or how easily participants communicated with their community and their extended family about haemophilia. How much does the community and family know about haemophilia, this bleeding disorder that is affecting the child in their care? It was also queried whether caring for a child with this condition has influenced their status in their community, and whether the affected children were freely accepted and allowed to play with other children. The interpreter further inquired about what the community's and extended family's thoughts were about the condition and what they thought had caused it.

4.1.1 Interaction with community

A summary of the category 'interaction with community', subcategories and codes under the theme 'interaction with community and extended family' is given in Table 8.

Table 8: Summary of the category 'interaction with community', subcategories and codes under the theme 'interaction with community and extended family'.

Theme	Category	Subcategory	Codes
Interaction with community and extended family	**Interaction with community**	**Communication with community**	• no desire to disclose • no desire to communicate • avoid contact with neighbours • community is not informed, no stigmatisation • only communicates to select few • communicates with neighbours to increase awareness of possible complications • community is informed
		Community support	• no support • sees no need to contact neighbours • help with first aid and lifting to hospital • only from one neighbour • has support but cannot rely on responsible care

4.1.1.1 Communication with community

Most participants displayed reluctance to communicate with members of their community.

> *C8: "I don't discuss my issues with my neighbours as you are aware that in the cities each person focuses on his or her business and not other people's problems. You cannot be telling your problems to everybody that you meet."*

> C7: "*I don't know people here. No one knows. Take for instance, telling my neighbours about Haemophilia. No, I can't do that, I am very sorry that persons will only know about it when her/his children is diagnosed with Haemophilia. I don't talk about my problems to other people. I don't even have a friend anyway, I just go out when I want to but don't visit any neighbours."*

> *C3: "I don't normally mix with my neighbours. They (friend's parents) don't know about his condition, so they just play"*

> *C2: "They (neighbourhood) do not know....."*

> *C1: "No my neighbours are not aware except for my family members. There's few of them that know about this. The parents of these children (friends) are not aware about the situation."*

One caregiver reported that she had informed the community, and that her explanations had been accepted. According to this caregiver there was no one in her community that had alternative beliefs regarding the cause of the symptoms.

> *C6: No, they (community) know and they don't think its something else.*

All participants lived in either formal or informal houses in very close proximity to their neighbours and privacy was often compromised. In spite of that, many felt that it was

best to keep to themselves in the urban environment, and decided not to inform community members about their children's condition. C7, for instance, had very little contact with her neighbours and did not want to involve them in her private affairs. Generally the affected boys played normally with other children and it was up to them to safeguard against knocks and bruises.

By and large there was very little knowledge of haemophilia in the general population (personal communication between researcher and support group coordinators) and people with haemophilia did not appear to be stigmatised. This is in contrast with a recent study undertaken in the UK where all participants reported that they encountered some degree of stigmatisation about their condition and that the general population still held a number of misconceptions, including that people with bleeding disorders must be HIV-positive and that they are homosexual (Barlow et al., 2007). This discord with the western world is possibly due to the fact that rural communities and segregated townships had very limited exposure to world news during the apartheid era in the 70's and 80's. Media was mainly published in English and Afrikaans and was primarily pitched at white readers (Kolbe, 2005). Reports of patients with haemophilia that were infected with HIV through tainted blood during the first stages of the HIV pandemic may thus not have reached this particular community.

4.1.1.2 Community support

Because of the nature of haemophilia, many bleeding episodes occur spontaneously and emergencies are unpredictable.

C2:'When he gets sick I must forget about work and rush to wherever he is'

This caused considerable disruptions to the routine daily lives of the patients and their families. The importance of communication is highlighted by one participant who has

involved some community members and subsequently is receiving assistance during crisis situations.

> *C2:'Our neighbour used to help us as she has done first aid training. So I usually call on her and also wake her up when there is a problem. I also usually call my other neighbour to rush him to hospital, even when we are not around.'*

Most participants reported reluctance to communicate their challenges around haemophilia-care to their neighbourhood community, particularly in the urban environment. Some participants spoke to a few select neighbours and in so doing, secured some support during emergencies. But, generally the community was not informed about the implications of haemophilia and remained unaware of the dangers and unpredictable nature of the condition. Many participants (8 out of 10) were rural migrants and, as discussed earlier, had to adapt to and fit into urban suburbs that had developed according to foreign, western norms (Mitchell, 1996). They maintained strong ties with their rural families and continued to define the rural place as their home and the home of their ancestors. The participants' reluctance to communicate serious issues with the urban community echoes previous research which suggested that rural migrants tend to view urban areas as strange and hostile environments in which they view themselves as temporary visitors. Within this environment they are inclined to trust and have dealings with people from their rural home areas and otherwise choose to keep to themselves (Magubane, 1973; Gugler, 2002). Thus members of a community, that is based on kinship and participation of all members, appear to find themselves isolated and without their normal channels of communication, preferring instead to keep their personal affairs private.

4.1.2 Interaction with extended family

A summary of the category 'interaction with extended family', subcategories and codes under the theme 'interaction with community and extended family' is presented in Table 9.

Table 9: Summary of the category 'interaction with extended family', subcategories and codes under the theme 'interaction with community and extended family'

Theme	Category	Subcategory	Codes
Interaction with community and extended family	**Interaction with extended family**	**Communication with extended family**	• the whole family knows (It became clear at a later date that even her son did not know); • family is informed; • paternal family is informed because they reside in vicinity; • communication with family improved after diagnosis • family take care of patient when he stays there; • family is informed but does not fully understand implications; • no communication; • before diagnosis kept symptoms from family; worried about stigmatisation in rural areas

4.1.2.1 Communication with extended family

Generally the extended family, many of whom still live in rural areas, was informed, especially if they needed to take care of patients in the absence of the caregiver. Some participants had migrated to Khayelitsha from the Eastern Cape out of necessity to be close to medical centres that are equipped to deal with their affected children**.** Health care in the Eastern Cape, as discussed in Chapter 2, is inadequate and not specialised enough to care for families with haemophilia. Participants did however maintain strong ties with their families in the rural areas and visited on a regular basis, especially during school holidays.

C4: 'Yes, his (grand)*mother knows from his dad's side because she is residing within our vicinity'*

C2: 'They know, because when we are not in the Eastern Cape and the child is there, my parents are the ones who have to look for help when he gets sick. They usually struggle because the clinics in the Eastern Cape are not able to help.'

C9*: 'They know about P5's problem, but not as deep as I do because the other time I went to the Eastern Cape when my brother's child died, I left P5 with my sister.*

When I came back from the Eastern Cape in the early morning hours, I went straight to work. When I came back from work I went straight to my sister's place in Site C, where I found out that P5 was sick, and my sister had been given money to take him to Hospital but she did not. When I took him to Red Cross the Doctors treated him with great urgency as they said he had nine minutes to live. I am the only one who knows about the seriousness of this illness'

Before diagnosis, this participant was reluctant to speak to anyone about her son's bruises, swellings and bleeding. She worried that her family would suspect that she was overreacting.

*C5: No, I didn't tell them about his problem of having skin growths (*swellings*). I only talked about it after I came from the hospital (*after diagnosis*). Before I couldn't explain this or tell anyone about it. It's only now that I am open about his condition but before I was sceptical to talk about it. As people who come from rural areas it is difficult to talk about certain issues and start telling everybody that so and so is having such a condition. It's rare that people will take note on a child that is bleeding - it's not something that they will take serious.*

C9's account describes her struggle of bearing the responsibility for her son's wellbeing on her own. It also illustrates that family members cannot always be relied upon as they may not realise how serious the condition is and how urgently any incident has to be attended to. C5 chose to initially carry the burden of her child's condition herself rather than discuss it with her family, especially while she had no explanation for the bruising and frequent nosebleeds. Her extended family reside in the rural areas of the Eastern Cape where traditional beliefs and practices are still followed and the approach to illness is symbolic and intuitive. Nosebleeds are considered to be a minor issue and her rural community may not have taken her concerns seriously. C9 similarly made a comment regarding nosebleeds and their triviality:

C9: **…..** *according to us, nose bleeding is some sign (spontaneous) from our ancestors. Maybe they have thought of you and are now reaching out to you - Okay I hear you.*

4.2 Taking care of a family member with haemophilia

Participants were asked whether they had any previous general knowledge of haemophilia, how and when they first became aware of the condition and what they experienced while caring for the children. Participants were further asked as to what they thought had caused the condition and whether they had any alternative explanations. These questions were posed to ascertain the effectiveness of communication between medical staff and caretakers and to elicit any alternative beliefs and explanations.

4.2.1 Participants' experiences associated with haemophilia

A summary of the category 'participants' experiences associated with HP', subcategories and codes under the theme 'Taking care of a family member with haemophilia' is presented in Table 10.

Table 10: Summary of the category 'participants' experiences associated with HP', subcategories and codes under the theme 'Taking care of a family member with haemophilia'

Theme	Category	Subcategory	Codes
Taking care of a family member with HP	**Participants' experiences associated with HP**	**Background knowledge of HP**	• None • Heard about it in school
		Experiences associated with HP symptoms	• links HP to injury • concern/confusion due to unawareness • no support in the Eastern Cape • treatment/self medication has improved situation • unaware of condition because of lack of communication • frequent swelling and nose bleeds • sleepless nights because of swellings • distress because bleeding episodes are unpredictable
		Experiences associated with HP care	• does not fully acknowledge HP • protective to try and prevent incident • strain of being on stand-by • anxiety about next incident • financial worries • feelings of guilt • HP controls participant's life and finances • Limitations regarding over the counter medicines

4.2.1.1 Background knowledge of haemophilia

Nine participants had no background knowledge of haemophilia

C9: No. Even the name haemophilia, I don't hear it when they say it, even when it is explained I do not understand it.

C8: I didn't know anything about this condition. I've never heard of it.

C1: No, I've never heard

C3: No, I never heard about it.

C5: None

C4: None

C1: None

C7: Never, we only heard from a doctor when he diagnosed him. We never heard about it before. Was this illness there before? When did you discover Haemophilia at the hospital?

C10: *No, I never heard about it.*

One participant had some knowledge and remembers having been taught about haemophilia at school.

C5: I first learnt about it at school, where we were told that it happens when the blood does not clot. I never thought I would have a (it would happen to my child) child with this disease, I just knew the name and that was all.

There appeared to be virtually no background knowledge of haemophilia amongst the participants. The interpreter asked what they knew about the disease or whether they had ever heard about it before, and 9 out of 10 participants responded with a straightforward 'no'. They had never heard of haemophilia or its symptoms until they were told about it by the doctors. C2 remembered having been taught about haemophilia in school and understands that it is caused by blood not being able to clot. C9 said that the name haemophilia means nothing to her and that explanations given by medical professionals did not help her understand the condition.

Because people naturally have a fear of the unknown it is important that explanations given are clear and adapted to the target audience to increase understanding and to help coming to terms with the condition. As mentioned previously, the dominant languages in most major hospitals in the Cape Town region are English or Afrikaans and isiXhosa speakers face the difficulty of communicating in a foreign language. Complex concepts

such as the causes of haemophilia, how blood clotting factors work together to form clots and inheritance patterns are difficult to understand. Interpreters are often not available and if they are, they are frequently chosen indiscriminately from amongst whoever is bilingual and available at the time (Levin 2005; Penn, 2007). Because there are often no isiXhosa words for genetic terms, interpreters without the knowledge of some genetics or at least a background in health care would find it difficult to interpret or paraphrase these complex issues. Since participants are not always able to converse in their home language and if they lack proficiency in English or Afrikaans they are probably also limited in asking appropriate questions, which could leave many issues unanswered.

The lack of background knowledge of haemophilia within this cohort is unlike what was found in the West, where there is at least some awareness, either because haemophilia has affected the Royal Families of Europe for the last few centuries or because of the tragic consequences of tainted blood transfusions which spread HIV amongst people with haemophilia during the 1980's (Barlow et al., 2007).

4.2.1.2 Participants' experiences associated with haemophilia symptoms

There was no known family history amongst any of the participants and none of the children were diagnosed at birth. Participants were thus initially unaware of the condition and frequent nose bleeds and bruising caused fear and confusion for most.

> *C5: I didn't take notice of anything before the time he went to Red Cross Children's Hospital. I could see that he liked to bleed, to have bruises and swell a lot. Sometimes I would think that he has been beaten or he bumped himself. He would lie to me and say he bumped into something and I would believe him because he is a child and children like playing and he would have skin growths (swellings).*

C1: *...when he was 5 years old he started having this condition but his problem was with nose bleeding…..It used to trouble me before I got help from the Red Cross. I am not worried* (now) *as I am getting help from the hospital.*

C9: *I just noticed that this problem doesn't go away these marks just develop. I noticed this on that leg when I used to take him to the Red Cross. I would notice that more marks have developed elsewhere whilst he still had this operation.*

C8: He used to bleed and he was bleeding all the time. He bleeds through the nose only. He used to bleed profusely on the pillow in such a way that he used to have blood clots.

C6: *he would get swollen joints and we didn't know what was happening.....* we *were so concerned and also scared of why he is not getting better when he gets swollen.*

C1: *What used to worry me was that I thought he wasn't going to get help and also I thought he was going to die.*

Especially in the rural areas of the Eastern Cape there appears to be almost no awareness of haemophilia amongst health care workers. Participants reported that clinical staff in those areas had a poor understanding of the symptoms, treatment required and that patients with haemophilia did not receive adequate care.

C6: We originally come from Eastern Cape. He was not getting help there and we decided that he must come here in Cape Town

C2:… they (extended family) usually struggle because the clinics in the Eastern Cape are not able to help.

C1: I've noticed when he played and sometimes if he fell off or he trip off and he would swell up. When I took him to the clinic, nurses at the rural clinic could not see what he was suffering from (didn't know). *My brother invited me to come to Cape Town to the Red Cross children's Hospital. At Red Cross I was told about my son's illness and what he was suffering from.*

One participant, the maternal grandmother of two cousins, who have both been diagnosed with haemophilia, was still largely unaware of haemophilia and of the fact that it is a genetic disorder. She linked the cause of the condition of one grandchild to a blow to the head and that of the other to a tooth extraction.

C7: *I thought it is because of the head injuries. He was beaten with a golf stick (Question: is it the same as P1's [cousin's] condition?) No, because P2 was beaten up and P1 was bleeding a lot resulting from the tooth that he took out. No, I first notice this with P2 and you don't want to lie, I was accusing the doctor*

Participants experienced fear and uncertainty when confronted with symptoms of haemophilia. Bruising and swelling, also of the joints, are common in children causing concern and distress amongst the caregivers, especially before diagnosis. The unexplained symptoms continued to reappear and caregivers were concerned because swellings and bruising took a long time to subside. Prolonged and frequent nose bleeds caused fear and feelings of uncertainty were compounded by the fact that very small children and babies cannot explain their symptoms. One caretaker suspected that her toddler had been beaten or had been fighting and was lying about it. C7, on the other hand, had never been aware of symptoms and was suspicious of the diagnosis. She repeatedly asked, during the interview, when and how the doctors had discovered this disorder and questioned whether they (the doctors) were not, perhaps, the cause of it.

All participants who originally came from rural areas reported lack of proper medical care for patients with haemophilia. Depending on the region in South Africa, haemophilia care can range from very good in urban centres to inadequate in rural areas.

Participants experienced that clinical staff in those areas were mostly ignorant of the condition and its symptoms, were unable to identify persons with haemophilia and were generally unaware of the urgency with which patients with haemophilia had to be treated during bleeding incidents. Generally rural medical personnel were not trained in reconstituting factor or diagnosing bleeds and often gave erroneous information or advice, which could be dangerous to the child's health (personal communication with haemophilia support group co-ordinator). It thus remained up to caregivers or patients themselves to decide whether the doctor had adequate experience with haemophilia and it was up to them to make ad hoc decisions in crisis situations. Studies conducted in Brazil and New Zealand similarly concluded limited clinical awareness and misdiagnosis continue to be a major problem in developing countries (Park and York, 2008; Fontes et al., 2003). The inadequate medical care makes it all the more important for caregivers to be well informed and to fully understand the condition in order to feel empowered to take action if necessary.

4.2.1.3 Experiences associated with haemophilia care

Haemophilia is a life-long chronic disease which imposes a heavy burden on families. Many participants suffered the stress of being on stand-by and having to rush to hospital at a moment's notice.

> *C8: There used to be a big concern because sometimes I had to rush P6 to the doctors after work at around 17H00. At times I am exhausted but I have to take him the Red Cross Hospital because of this bleeding and lump problem that used to happen. He hasn't had the bleeding problem for a long time now since the beginning of the year but he still get lumps especially if he bumps in to anything*

> *C2: Whenever there is a call from school I usually have to leave work immediately to see to the problem. When he gets sick I must forget about work and rush to wherever he is*

C1: *When he gets swollen I rush him to Hospital for I was given a direct order to rush him to the hospital anytime he complicates or encounters health problems.*

Participants were often very protective of the patients in order to avoid incidents and unnecessary bleeds.

C6 … we were always watching him. Take for instance, if we saw him limping, we would tell him not to kick the ball or run around again. And we understood him very well, in fact we were looking after him.

C3… every time he bleeds he becomes very bad, we fear for him to bleed and are always cautious of him injuring himself.

C10*: I fear because as he grows older, he is going to play rough with others as he is a boy, this can affect him and endanger him.*

Haemophilia is a chronic disease for which there is no cure, and children and their families have a life-long responsibility for self-management, to promote health and to prevent complications. Particularly the physical vulnerability of the children caused uncertainty and caregivers tended to react by being very protective and watchful. As discussed in chapter 2 children often do not like to be seen as being different from their peers and will strive to be part of the group. They will want to participate in the same activities other children their age do which may conflict with the caregiver's desire to keep them from harm (Canclini et al., 2003; Goldstein and Kenet, 2002). Caregivers thus have the additional difficult task of balancing the urge to be over-protective in order to avoid physical harm, with allowing the child to explore the world and promote a normal physical and psychological development.

Occurrence and nature of symptoms are unpredictable. Any type of trauma can very quickly develop into haematomas or haemarthrosis and factor concentrates have to be administered as soon as possible to stop the haemorrhages. The urgency with which any

trauma had to be treated, forced caregivers to fit unexpected and often frequent visits to clinics into their schedule. This severely disrupted daily life and was especially difficult if caregivers had to repeatedly and unexpectedly rush away from their place of employment.

One sad incident during this study highlights the, often tragic, consequences of what would otherwise have been a non-fatal accident. On his way to the haemophilia support group in Khayelitsha with his mother, a six year old affected boy was hit by a car and died because of an intra-cranial bleed.

Many participants felt overwhelmed and were burdened by constant worry, feelings of guilt and financial worries.

> *C1: What used to worry me was that I thought he wasn't going to get help and also I thought he was going to die. I used to be troubled before I got help from Red Cross Hospital. I am not worried because I am getting help from the hospital*

> *C8: I am concerned what if he dies. I don't sleep worrying about this as his mother is irresponsible. She leaves him with neighbours and she goes to her own places. That's my main worry.*

This participant was troubled because limited funds are restricting her from properly caring for her son and she felt that it also prevents her from finding proper help and possibly even a cure.

> *C9: The problem with P5 is that his sickness just comes up, it stays off for a bit and when it comes back, it overwhelms him, he becomes very sick and I struggle because I have no means….. It did, it has.* (Crying). *This situation has changed my life.* (Sobbing). *It changed it a great deal because I cannot do anything* (with a shaking voice). *Even when I crave meat from the vendor stands, when I buy it and get home to find my child sick I feel very guilty because I feel as if I should have known. When*

I find him sick I regret delaying at the vendor stand and buying the meat, and I would not know what I will come to when I get at home…… It's his sickness and him not getting a grant for it. I don't have support in my house and if P5 had a grant at least, I would be able to use the money to find help and a cure for him, I would also be able to take him to hospital. I am struggling a lot now because I do not see my income because it sees to P5 and taking him to hospital.

One participant, a school teacher explained how she suffered under the emotional burden of having an affected child. She also related how difficult it was for her to make plans for the future and pointed out that the need for support and medical care had stopped her from furthering her career.

C2: *I do not have fears, the problem is the pain that one goes through, it is not easy. When he gets sick I must forget about work and rush to wherever he is…… I am not able to apply for higher or better positions, for fear of being sent away or transferred elsewhere. I have to stay close to my child and be here when he gets sick. So it has affected my life very much.*

Caregivers were deeply affected by the children's condition and felt restricted and overwhelmed. They were constantly faced with the possibility of another incident and many were frightened because they feared that the child might die as a consequence of a serious bleed. In addition to constantly feeling anxious caregivers also incurred considerable indirect financial costs. Most participants came from low socioeconomic backgrounds and the indirect costs involved in caring for a child with haemophilia had a significant impact on their lives. Some pointed out that the unpredictable nature of this bleeding disorder may affect their employment and employment options. Their lives were transformed by the unpredictability and the ensuing loss of control. They had to be available 24 hours a day, in case of emergencies, and needed to be in close proximity of centres or clinics that are experienced in treating patients with haemophilia. For most participants in this study, treatment consisted of hospital-based, on-demand therapy. The relevant hospitals however are situated near the city, while participants live in

townships on the outskirts of Cape Town. Thus transport costs were always an issue for patients and caregivers, and even if they were able to self-medicate, patients needed to regularly travel to hospitals to collect factor concentrates. Similar findings were discussed in chapter 2 where participants reported on the negative impact on working lives and financial circumstances of caregivers of patients with haemophilia in the UK (Barlow et al., 2007).

4.2.2 Treatment and being in control

A summary of the category 'treatment and being in control', subcategories and codes under the theme 'Taking care of a family member with haemophilia' is given in Table 11.

Table 11: Summary of the category 'treatment and being in control', subcategories and codes under the theme 'Taking care of a family member with haemophilia'

Theme	Category	Subcategory	Codes
Taking care of a family member with HP	**Treatment and being in control**	**Understanding of treatment**	• ignorant about treatment, • only doctor knows, • not familiar with terms; • just follows doctor's instructions; • participant administered drip • patient administered drip
		Locus of control	• With doctors • With hospital • With God • No control due to lack of understanding • With participant/patient

4.2.2.1 Understanding of treatment

Half of the participants were ignorant about treatment for haemophilia, were not familiar with the terms used in the hospitals and generally just followed doctors' instructions. Only one participant had previously heard about haemophilia during her school education.

Participants were asked what they thought about the kind of treatment their boys should receive. The aim was not only to elicit the understanding of treatment, but also whether there were any alternative beliefs regarding treatment.

C7: I don't know his treatment and I can't really comment on his treatment as I do know nothing about it and am also not familiar with the terms since I am not a nurse nor a doctor so I can't say this is not right for him and that is good for him.

C4: The doctors told me what kind of a child he is and how I should treat him and how he should play with other children.

C3: I am not sure, because when he went to Red Cross he got something, I just cannot think what it was. ……..No, he is not on any medication, but he got something when he went because of the accident (the fall). *He is not taking anything, when he goes to Red Cross he gets whatever relevant medication at the time, otherwise he is not sickly.*

C8: I can't tell you. I don't know. They inject him.

Three older patients, in their teens, and one participant had been taught to administer the drips themselves and were thus able to infuse blood clotting factor as and when needed for an injury or a spontaneous bleed.

C2: *They also taught me how to treat him so that I don't have to go there all the time for the treatment*

C6: When he feels that he played a lot or hurt his wrist he usually takes himself his medication

One participant had a reasonably good understanding of the treatment because her son is able to self-infuse, but felt disempowered as she is not able to make decisions regarding

over-the-counter medication. This puts added strain on her, her time and her finances as any other ailments, not related to haemophilia, may require additional trips to the hospital.

> *C9: He needs to be put on a drip for him to be all right, you cannot get medication from the shops even if he has asthma or anything else, I cannot buy him medication from the shops. I usually take him to hospital.*

It is important that people with haemophilia are not given aspirin or medications that contain aspirin. Medicines that contain aspirin or NSAIDs (non-steroidal anti-inflammatory drugs such as ibuprofen or naproxen sodium), can affect blood platelets and lead to increased bleeding (World Federation of Hemophilia, 2009). Other common household remedies, such as Alka-Seltzer, also contain aspirin, making it necessary to read labels very carefully before administering any medication to a person with haemophilia. Medical professionals had probably advised C9 to check with the hospital every time medication is required. This was presumably a safety precaution but resulted in added restrictions for her.

Overall the understanding of treatment was poor, especially considering that patients had to visit clinics frequently and drips had to be administered on a regular basis. Caregivers stated that they leave it up to the doctors to decide and did not express a desire to learn more about the treatment or treatment options. No alternative beliefs regarding the nature of the treatment were elicited through this line of questioning. However, alternative beliefs regarding the cure of the condition are discussed under the heading 'General and Cultural Beliefs'.

4.2.2.2 Being in control

Locus of Control

Locus of control is usually defined as a generalized expectancy about where control lies, namely within oneself, another, chance, or God. At least half of the participants did not

fully understand the treatment and were unable to administer factor replacement at home. They depended on doctors and clinics for guidance and direction, and the locus of control thus remained with the health professionals. Participants relied on doctors or hospital staff when it came to their children's health and did not appear to perceive themselves as being the person in control. The following quotes are typical of the responses given by the participants.

C7*: Its only the doctor who can comment on that* (treatment)

C8: I can't tell you. I don't know. They inject him.

All participants in this study resided in townships and came from lower socio-economic backgrounds. There was either a reluctance to question the healthcare hierarchy or, alternatively, language issues hindered effective communication. As mentioned previously, apartheid had a great impact on people's lives and especially the older members of this community still have to relearn or get used to the fact that they have the right to question authority. Ignorance and lack of treatment can unfortunately prove fatal and the chairman of the South African Haemophilia Foundation in his report of November 2007 stressed that it is important for caregivers and patients to take initiative and accept responsibility for their own health in order to prevent permanent damage or unnecessary deaths (Rayner, 2007).

Lipman et al. (1994) in an American study described the life-view of people of lower social status and reports a sense of powerlessness and meaninglessness, resulting in a feeling of isolation from knowledge of health and a reluctance to have any part in the middle classes' need for preventive measures. Good management of haemophilia would, however, require full knowledge of the complications associated with the condition and the preventive behaviours to avoid complications. Ideally caregivers and patients would receive extensive training in home treatment to manage complications (Nazarro, 2006). Alternatively, support groups could serve as effective platforms for caregivers and patients to learn more about the condition and how to deal with it, to get encouragement,

to voice their concerns, and to share management experiences. The South African Haemophilia Foundation, together with some volunteers, have taken the first step in this direction and facilitated the formation of several support groups in the Cape Town area, including one at the treatment centre of Khayelitsha Day Hospital. This support group, the Khaya Reach Out Centre for Kids (Khaya ROCK) was formed with the help of a young person with haemophilia who wanted to help reach out to other young affected boys who lacked knowledge about their illness, did not know how to source information and were overwhelmed by language barriers. Unfortunately, attendance is often poor and volunteers become despondent and lose interest. Attendance of the support group meetings, which are scheduled once a month on a Saturday morning, may be affected by contents or timing of the meetings or by transport costs to the venue (Khayelitsha is very spread out, covering an area of approximately 47km^2).

The reluctance to become empowered and to take control of the situation is different to western cultures where it is commonplace for patients and their families to find independent information on their medical condition either from books or on the internet. Patients feel empowered to challenge their physicians and question treatment and treatment options. This is especially important with chronic disorders like haemophilia, and it has been shown that people who are permitted to, or are able to become partners in their own care, can expect much better overall outcomes (Paper, 2002).

Then again it is also relevant to remember that different cultural beliefs and practices give rise to different expectations. As mentioned previously, up to 80% of Black South Africans use traditional healers who are expected to diagnose without having to question the patient. Traditional healers appeal to their ancestors for help in diagnosing problems and prescribing remedies (Nattrass, 2006). Similarly, the traditional healer's diagnosis would be accepted without questioning. In addition, historically, people have been taught to respect and not question the doctor, and thus the reluctance to query and seek information may not just stem from a sense of disempowerment, but also from educational and cultural issues that discourage them from taking a more active role in decision-making about their care.

4.2.3 Perceived cause of haemophilia

A summary of the category 'perceived cause of HP', subcategory and codes under the theme 'Taking care of a family member with haemophilia' is given in Table 12.

Table 12: Summary of the category 'perceived cause of HP', subcategory and codes under the theme 'Taking care of a family member with haemophilia'.

Theme	Category	Subcategory	Codes
Taking care of a family member with HP	**Perceived cause of haemophilia**	**Understanding of the pathogenesis of haemophilia**	• HP is a result of injury • HP is a result of medical treatment • cause not communicated by medical staff • unsure about the cause • would like to know the cause • presumes its some kind of cancer • believes only God knows • has not thought about it

4.2.3.1 Understanding of the pathogenesis of haemophilia

Most participants have very little understanding of the genetic cause of haemophilia and that, at the moment, there is no cure for it.

One mother, who has tertiary education, presumes that it is some kind of cancer.

> *C2: I don't know (*what caused it*). In Grade 12 they did not get into details about it, we were just told of it as a type of cancer and nothing more.*

Participants are generally not well informed and the cause of haemophilia is possibly not effectively communicated by medical staff.

> *C8: I honestly don't know. This is something that I cannot answer, for it's only God knows as I have alluded before, nobody would know the reason why this happens only God knows especially about this particular condition.*

C3**:** *I can't say, I don't know. I want to know what caused this disease, where it comes from.*

C9**:** *Yhu! I do not know. This thing with P5 is rare, I never even thought of what it is or what was wrong with him.*

C6: (Grade 12) …no we were not told (the cause of haemophilia)

C7 relates haemophilia to a head injury caused by a fellow pupil and believes that this injury is the cause of the bleeding disorder.

C7: I thought it is because of the head injuries. He was beaten with a golf stick.

Similarly, as with the understanding of treatment, despite frequent contact with medical professionals, participants seemed to have no or, at most, only basic knowledge of the cause or pathogenesis of haemophilia. The line of questioning did not elicit whether participants had their own theories or ideas about the **cause** of the disease. Three participants had grade 12 education but did not feel compelled to research and to find their own independent information about the condition. All seemed to be satisfied to accept and follow whatever instruction or information is given by the medical professionals. Unfortunately, lack of knowledge hinders planning for the future, communication with possible carrier daughters and decision making. Again, it is reasonable to infer that language difficulties made it difficult for participants to fully comprehend verbal information or to read and understand available publications. At a later stage of the interview, however, it appeared that some participants had alternative beliefs regarding the primary cause of the condition, which is discussed in more detail under the heading 'General and Cultural Beliefs'.

4.3 Understanding of inheritance

To ascertain what participants understood by inheritance, open questions were asked about their thoughts on why some people get ill and others do not, about what they

understand if a condition is referred to as being inherited and from whom a condition would be inherited. They were also asked whether they were aware of any illnesses in their family that were passed down and to share their thoughts about inheritance (descent) and about the location of genes in the body.

4.3.1 General Inheritance

A summary of the category 'general inheritance', subcategory and codes under the theme 'inheritance' is given in Table 13.

Table 13: Summary of the category 'General inheritance', subcategory and codes under the theme 'Understanding of inheritance'.

Theme	Category	Subcategory	Codes
Understanding of inheritance	**General inheritance**	**Inheritance from whom?**	• inheritance related to resemblance to parents • passed down from the forefathers • questions inheritance - from where? • inherit from someone who already has the condition • inheritance through mother's blood • passed on by mother who carried him • passed on through breast feeding • questions possibility of inheriting from a deceased relative inheritance of stronger genes • Maybe it's a transmission of blood • suspects that it comes from father's side as he is the one performing during intercourse • compares it to ingestion of alcohol during pregnancy • does not know how it is transferred.
		Inheritance of illness	• inheritance of cancer from a family member • family history of allergy to red meat • mother - by transferring it through the blood • transmitted through somebody else's blood • inheritance through staying together in a house • same as inheritance of TB from a family member • does not know • gets confused • not aware that a condition can be inherited

4.3.1.1 Inheritance from whom?

Several participants acknowledged that some conditions run in families.

> C2: *It (inheritance) means you got something from somebody else. Maybe when someone has some disease, it is said that they inherited it from another family member.*
>
> C6: *I can say it's an inheritance from the fore fathers*
>
> C9: *People do take after others; I think sometimes you take after someone whom you have never even known*
>
> C10: *When you talk of inheritance, I think of someone who has taken after some old family members who have long passed away, or close relatives, for an example from mine or his father's side*….(How do you mean?) *If for instance there was someone who had this problem, but someone I never even knew.*
> ……. *I am going to say again that I think it's a case of someone having a problem that another member of the family had before, even if it is not exactly the same problem, I am not sure if I am answering you appropriately*

One participant appeared confused about the question of inheritance and was unsure from whom her child would have inherited haemophilia. At the same time she made a connection to her own mother who died during childbirth. It was not clear whether she believed that genetic ties were broken because her mother died while giving birth to her.

> C5**:** (is condition inherited?) *I don't know, maybe, I can say but I don't know what else to say. Where did he get this inheritance from? My life story is that my biological mother passed away when she gave birth to me. You know in the rural areas women used to bare* (bear) *children at home* (not in hospitals) *I don't know why.*

When speaking about inheritance, participants referred to physical, behavioural and moral characteristics possessed by a parent and passed on to a child. One participant seemed to have the view that, given that there are two children, one will inherit from the father and one will inherit from the mother. Unfortunately, no further probing questions were asked by the interpreter to explore this concept in more detail.

> C4: *I can say for example as I have two children one of them will inherit from me. Let's say the other child is naughty and the other is sweet (well behaving). The other one might be naughty and his dad my not have such behaviour maybe I might be the one who is naughty and not their father. I can pick up my bad behaviour from the child and I can easily say that this child's behaviour resembles my character when I was a child. That's how I know inheritance.*

> C9: *No I don't know about that. I only know that when someone is good (has good character), people would say she takes after her mother who was also good. Others may be become the opposite and they also would be said to have taken after someone*

Participants explained inheritance as character traits, behaviour or resemblances that have been passed on by parents, relatives or long dead ancestors. It was, however, not clear how they understood these traits to be passed on. It is possible that a more experienced interpreter and more appropriate probing questions would have elucidated this. The following exchange between C10 and the interpreter highlights some of the drawbacks associated with having to rely on an interpreter-interviewer: "…*I think of someone who has taken after some old family members who have long passed away"*. The interpreter then probed by asking "*What do you mean*?" to which C10 responded by repeating what she said before, just putting it into other words: "*If for instance there was someone who had this problem, but someone I never even knew"*. The interpreter then left it at that and did not continue to probe into the participant's thoughts regarding the mechanism by which things are passed on from generation to generation. It also appears that C10 felt as if she were being tested and was under pressure to give an appropriate answer: "…*I am not sure if I am answering you appropriately"*. The interviews did not

reveal whether or not participants, while they accept that certain traits or conditions are passed on from generation to generation, have any ideas about how they are passed on. Personal observations by the researcher and transcripts showed that participants were reluctant to elaborate when asked about inheritance and may have felt apprehensive because they felt unable to answer certain questions.

Participants also referred to stronger genes (or dominant parental sides of the family) and attribute physical resemblance or behaviour to the parent with the stronger genes.

> C4: *Let's say his daddy's genes are no as strong as mine. My strong genes will dominate the father's and the child will inherit my genes.*

> C5: *You do understand that it is common with us African (Black) people to hear people saying a child looks just like his father and the mother has lost the battle. What I meant is that the most powerful and strongest and dominating side is my mother's in this sense and my dad's side is weak.*

C4 and C5 actually referred to genes when asked about inheritance. There is no equivalent Xhosa word for genes and interpreter and caregivers uses the word '*ii-genes'* which is borrowed from English.

> *C4: I'm saying that as we had an encounter with the father of my child my genes will regenerate to my child during my pregnancy period and when I give birth to him, he will be born with my genes in him and not his dad's as I have the dominant genes.*

The interpreter referred to this and continued probing: "You talked of genes that are inferior and dominant to others. Do you think that the child will not take the weak genes and is bound to take yours just because they are dominant or strong?"

> C4: *Yes, I do*

C5 had similar ideas about strong and weak genes.

> C5**:** (Question**:** So I want to know if the child can not have the weaker genes as well?*) No I don't think so, he will take those strong ones, maybe since they are strong they will quickly be passed to the child as they are strong.*

Unfortunately the interpreter did not continue probing as to how she thought the genes would be passed on.

There were several instances where reference was made to the mother that gave birth to the child when referring to inheritance.

> C4: *… he has all the rights to have my inheritance because I am his mother and I've given birth to him. He can too inherit his father…….. Resemblance? He looks just like me as a mother who bore him. He can look exactly like his dad or me. It's something of that nature.*

> *C3: …something that was passed on to you by the person who gave birth to you. ……maybe whoever gives birth to you, transfers it to you through the blood, and then it only comes out at a later stage.*

While there were various ideas about how a child could inherit from the mother during pregnancy, the questioning/probing during the interviews did not elicit an explanation on how it would inherit from the father. One participant and her husband maintained that the child draws everything from the mother and believed that it is connected to the shared blood between mother and child. C8 believed that the child can only inherit from the mother, because she carries it in her womb.

> *C8:* (Question: Can the child inherit from the father?) *No, he doesn't inherit from the father because he is kept inside his mother's womb.*

> ..... *It's like he is sharing the same blood with his mother, he eats from her. He draws everything from his mother. It is like a drinking mother (*that transfers alcohol to the child*) there is something happening to the child when she drinks - such things are bound to happen. That's how I understand it that all things come from the blood.*
>
> *C8 (husband):* (How is the condition transferred to the child?)*To be infected (contraction)? Yes, it is your blood especially if you were not treated for this and as you breastfeed he grows and it manifests itself in his body.*

When asked about how they thought a condition could be inherited, several participants made reference to 'the mother that gave birth to the child'. There was a common notion that conditions are transferred through the mother's blood to the child. Several participants believed that while the child lives and grows inside the mother's womb and is nourished by her, certain conditions can be passed on from her to the child. C8 made the comparison to a drinking mother, who would transfer alcohol to the child. Thus inheritance could be seen as an act of transferring a condition or problem to a child through the mother's blood or even through what she eats.

> *C10:* (What do other people think is the cause?) *They just say that maybe I ate something wrong, so when I got pregnant I carried it with the pregnancy.*

From a traditional healing perspective pregnancies are seen as a very delicate time when woman may have lots of problems (Abrahams, 2002). Women and their babies are susceptible to environmental influences and are particularly vulnerable to 'evil' or 'spells' during that time. The perception of heightened susceptibility during pregnancy helps to explain the notion that this is the root cause of many problems that the child may develop.

Another participant believed that the child will inherit only from the father, because he is the one that 'performs' during intercourse and that 'genetic material' is passed on by means of a hormone.

C1: *I would say it comes from the father's side. I think that it is the father that performs when he sleeps with the mother ..... maybe there is a hormone that gets secreted when you mate.*

4.3.1.2 Inheritance of illness

This participant recognised that there is a family history of cancer. She also strongly believed that one must continue to fight the illness regardless of where it comes from.

C7: My brother reminded me that our father also had Oesophagus Cancer so the child had inherited it from him. So that is when I heard about the inheritance of illnesses. I said nonsense; I was also diagnosed with Cancer in 1986 but look at me now I am still alive and rocking. But it was unfortunate for the child; he passed away three months back.

There appeared to be an idea that conditions are inherited because people are sharing the same house, and comparisons were made between the inheritance of cancer and contracting TB.

C1: …*maybe you stay together in a house, for instance, maybe the person is your relative, it's a must that you will be affected because you stay together in one house ….. it simply means that you inherit something from the person who has already got that and you are bound to have it too…… Once I inherit the thing that the person had, I'll definitely have the same thing.*

C8: *Inheritance is for example if I had cancer it is likely that one member of my family will also inherit it. I know that as a fact. The same with TB if I had one member of my family will to have it. It is likely that at home there is someone who has TB definitely one person is also infected that how I know things.*

It is possible that there is some confusion between inheritance and infection. Further open questions, follow-up interviews or discussion groups would be necessary to elucidate whether there are uncertainties or misunderstandings in this regard.

C5 explains inheritance by referring to her father and all her siblings who are allergic to red meat.

> C5: *What I can say that I know of, is that this is the hereditary resemblance. You know back home in my family, my dad is allergic to red meat but sometimes he would eat it sometimes and nothing would happen to him (health wise). All my siblings inherited dad's allergy. I am the luckiest one. I've never encountered such problem. I am not allergic (to red meat) but my siblings are……I am saying my dad is allergic to red meat and most of my siblings inherited this. Most of my siblings refrain from eating red meat. I do eat red meat and I am not allergic to it.* (Interpreter: Do you resemble your dad?) *I do resemble my dad but I haven't taken after him. I didn't inherit his illness of allergy. Definitely, I took after my mom.*

Although C7 seemed to be familiar with the fact that certain conditions (for example cancer) run in families, the principle of genetic disorders did not seem to be understood. She questioned the possibility of inheriting from a deceased relative, especially if the child was born after that person had died.

> *C7: my father had passed away a long time ago with throat cancer, where is the connection, and the child was born in 1992? The child [my brother's] also passed away from the same illness that my father had.*

Two participants were either not familiar with the term inheritance in connection with illnesses or may not have understood the question.

C4 **(**When asked about inheritance of an illness): *No. I haven't heard of that one. I don't want to lie to you.*

C9: Of sickness - *No I don't know, I get confused.*

Most participants seemed to believe that the susceptibility to certain types of illnesses runs in families. In the amaXhosa worldview illnesses like epilepsy, asthma, chronic bronchitis, some skin complaints and mental illness are regarded as natural illnesses. Natural illnesses just happen, are not due to any fault of the patient and are referred to as *ufuzo* (Ngubane, 1977). Medicines used to cure diseases of this class are believed to be effective in themselves and are thus not ritualised. The need for finding a cure or solution in the traditional sense does not apply to them. Rituals are only considered necessary for rare or more serious illnesses.

During the interviews the interpreter, amongst others, used the word *ufuzo* when asking about heredity. The word *ufuzo* is understood to refer to resemblance as well as inherited physical and behavioural traits, and is not normally used in the context of other, but 'natural illnesses' (personal communications with N. Zitho, interpreter and with M. Ralarala during a workshop). It is possible that this caused some confusion and that the participants may have misunderstood the questions. After discussing this with the interpreter it was concluded that there may be some danger in using *ufuzo* in the wrong context and that in future it would be better not to use it at all and to replace it with simpler terms like "How did you get it?", "Where did you get it?" or "Who did you get it from?". It was discussed previously that, even in the western world, public understanding of Mendelian genetics is very limited and Mendelian explanations of inheritance are often poorly accepted and understood. Richards' (1998) study in the UK showed that in lay understanding there is no concept of phenotype and genotype, or of a genetic material that is transferred across generations. Although in many lay discussions of inheritance there are notions of blood or blood lines, participants are usually not implying that in a literal sense blood is the hereditary substance. Lay knowledge, across

cultures, seems to develop through concepts of family relationship and is associated with familiar resemblance and similarity (Richards, 1998; Richards, 2000).

4.3.2 Inheritance of haemophilia

A summary of the category 'inheritance of HP', subcategories and codes under the theme 'inheritance' is given in Table 14.

Table 14: Summary of the category 'inheritance of HP', subcategories and codes under the theme 'Understanding of inheritance'.

Theme	Category	Subcategory	Codes
Understanding of inheritance	**Inheritance of HP**	**Haemophilia**	• nobody knows whether it can be inherited • questions family history as cousins have different mothers • confused about inheritance • aware that boys are affected • understood that inherited but believed it would pass • suspected that injection caused the problem(HP) • Outstanding cultural rituals could be the cause of the illness • Only God knows
		Family history of HP	• no known family history • .not aware of family history • does not believe that nephews have the same disorder • maternal grandfather's brother had the same condition

4.3.2.1 Haemophilia

Because haemophilia is inherited in an x-linked recessive manner, it is passed on by the mother to her male offspring. Girls may be carriers and some may show rare symptoms like heavy periods or excessive bleeding during the birthing process. To ascertain how well participants understood genetic inheritance of haemophilia and the implications for future generations, they were asked whether they thought that boys and girls can inherit (get) this disorder.

C6: *yes , they (girls) can have haemophilia, she would have inherited it from the forefathers.*

C9: Boys and girls? *I don't know if they would or not, I can't lie.*

C7 believes that nobody knows whether it can be inherited. She also questions the concept of family history because her grandchildren (cousins) have different mothers.

*C7: ... nobody knows (*whether haemophilia can be passed on*), they can only tell their children if they are diagnosed with it that they had the same illness as well. So tell me where does this factor come from?* (Probably referring to clotting factor VIII)
I also want to know as they are two different children with two different parents. Their mothers are sisters. I want to know how possible it is for one to have it. How can they both have the same illness because one had taken the tooth out and he had a problem with bleeding, he was bleeding a lot (The other boy was diagnosed after a blow to the head) … *I would understand if they have one biological mother but their mothers are sisters. I really like to know how this can happen. I am not well informed, so I don't know* (whether boys and girls inherit this condition).

C3 is C7's daughter-in-law. She had been caring for her two nephews after C7 passed away. Although she mentioned earlier that she believed that there is a family history, she was not at all clear about it and also questioned the fact that the boys have different mothers. She did however make a connection to C7, who is the grandmother.

C3: *I want to know what caused this disease, where it comes from. When you say it comes from mothers, what causes it, because P2 and P1 have different mothers and fathers, but their mothers both come from C7?*

C8 is also a grandmother caring for her affected grandchild. She has no idea of how and why the child could have inherited haemophilia.

C8: I don't know. I wouldn't know and I cannot answer to that. Only God knows.

C2 maintained that she understood that haemophilia was an inherited disorder, although she initially believed that it would pass. Her case is complicated by the fact that results of a genetics test imply that she is not a carrier.

C2: According to my knowledge it is a boy's problem, although they inherit it from their mothers. My knowledge was increased when I heard of it at Red Cross, where they drew blood from my genes, and I learnt that it is something that is apparently inherited. Even when they explained at the Red Cross, I never took it serious, I just told myself that it was going to pass, I did not think it was a life disease. I do not know, but they did some blood tests on me and they said they did not see in me. I really don't know because according to them, a child inherits it from the mother.

As mentioned in chapter 2 it is possible that the mother is a mosaic and carries the mutation only or partly in the germ cells, or she may even be a somatic mosaic. Routine mutation analysis is not able to identify all causative mutations and especially somatic mosaicism may remain undetected (Leuer et al., 2001). Somatic mosaicism should definitely be considered in this case, especially during risk assessment in genetic counselling. Because, as mentioned earlier, a mother has an 85-90% chance of being the carrier of her son's de novo mutation (Graw et al., 2005).

During the interview her husband interjected and explained that outstanding cultural rituals could very well be the cause of the illness. Judging by body language and facial expressions, C2 seemed to be in agreement with her husband.

C2 (husband): *I initially thought it was because of the injection from the KTC Hospital doctor that was the problem, or that caused the problem. As time went I also realized that there are outstanding cultural traditions and rituals that I needed, which could be causing this illness.*

(KTC is the name of a squatter camp near Guguletu. The KTC Day Hospital was formerly known as the Guguletu Maternity Obstetrics Unit)

It became apparent that no participant had a clear understanding of x-linked recessive inheritance. Only one participant expressed concern regarding a family history, but only raised the subject once the meeting was over and everybody was ready to leave. The interview had left her wondering, whether her nephew's inherited condition could have any implications for her or her own offspring. Although she was in regular contact with clinical staff, the implications of a genetically inherited disorder for the family had not been effectively communicated to her. Referral to genetic counselling was suggested to her together with an offer to contact her regarding an appointment. She was hesitant and instead elected to note down the telephone numbers and to contact the counselling service in her own time.

Because there was limited understanding of the genetic inheritance of haemophilia it is doubtful that information about genetic risk was shared with relatives. Genetic diseases are family diseases and open communication about haemophilia within the family is important. Especially communication with daughters about their actual and possible carrier status and to make them aware of and prepare them for potential future reproductive issues.

Personal experience in this cohort does not seem to have influenced the perception of familial risk, but rather drove the desire to find traditional solutions. This differs from research in the USA, Great Britain and New Zealand, which showed that the impact of witnessing a relative's illness, especially if there is a strong emotional bond between the individual and the relative, will influence the individual's perception of familial risk. Walter et al. (2004) highlighted the fact that the greater the lived experience the greater the perceived importance of family history.

4.3.2.2 Family history of haemophilia

There is a definite family history of haemophilia in one family and there appears to be a family history in another family.

First family

C3 and C7 belonged to the same family. C7 was the grandmother of two affected cousins and C3 was their aunt. Both participants were caretakers of the children.

C3 recognised that there is a family history because two of her nephews are affected.

> C3.: *I think so, because there is another nephew (P2, my other sister in law's child) who has the same problem. I think they were born with this.*

C7 did not believe that the nephews have the same disorder. She believed that P1's bleeding problems were caused by a head trauma and was not aware that the hospital had diagnosed P1's cousin with haemophilia three years earlier.

> C7: *I am the eldest at home and no one had haemophilia. No, I first noticed this from P1 and you don't want to lie, I was accusing the doctor. No, there's no one who bleeds a lot in my family. We discovered then (*P1's head trauma*), when the doctors tried to find out who has this illness in the family; and they found that P2* (cousin) *was diagnose with this also 3 years back.*

C7 is an obligate carrier because both her daughters had affected sons (Fig. 8). P1's mother has passed away and C7 tragically passed away recently following an accident. The one surviving daughter, P2's mother is, according to her family, mentally unstable and suffers from epilepsy. She is also an obligate carrier, but it is not known to the investigator whether she is aware of her status or whether she has ever been tested. C3, the new caregiver of the boys is C7's daughter-in-law and is thus not related.

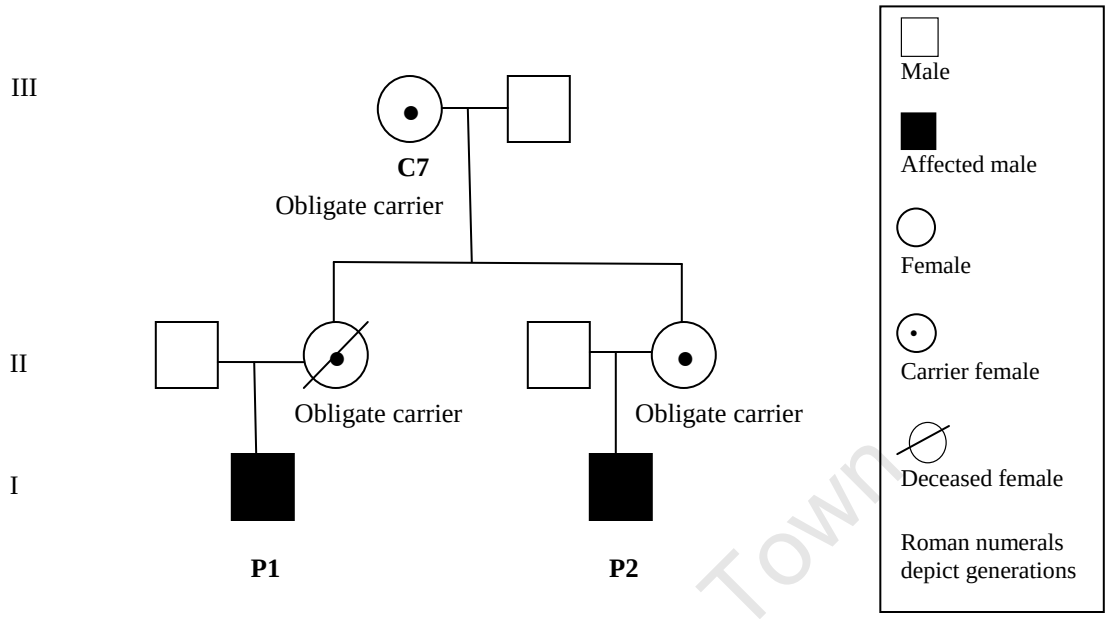


Figure 8: Pedigree Family 7

Second family

C1 believes that her nephew's (patient) maternal grandfather's brother had the same condition, although this has never been verified and no female members of the family have undergone carrier testing. It is possible that the great-grand mother was a carrier and that both, the patient's grandfather and his brother (X) had haemophilia (Fig. 9). The patient's grandfather may have presented with a mild clinical phenotype without showing any obvious symptoms. A correct diagnosis would have been unlikely in those days, particularly if the clinical phenotype was mild. Even today approximately 70% of patients with hemophilia are under-diagnosed and untreated, especially in developing countries (O'Mahony and Black, 2005). Barnes et al. (2007) have further highlighted the possibility of different clinical phenotypes amongst family members with the same mutation in a recent case study of non-identical triplets with haemophilia A.

If the patient's grandfather was indeed affected, it would mean that the patient's grandmother and mother are obligate carriers and that there are further implications for the participant's siblings and their offspring.

C1: *it was P4's grandfather's brother and our mother only told us when we asked. We didn't know and we were asking my mother, just to know if he inherited it from his forefathers.*

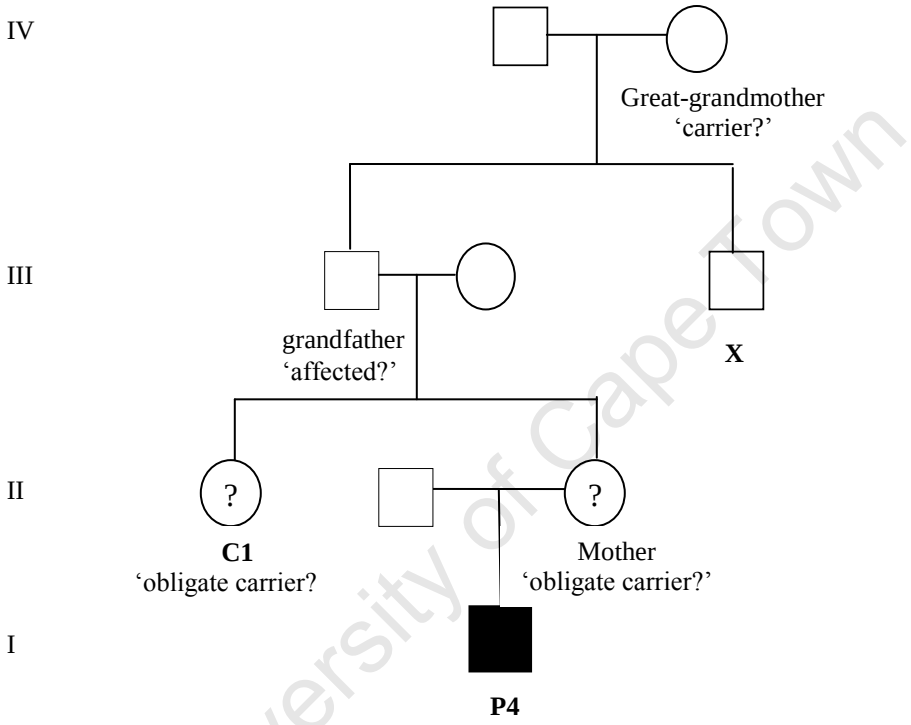


Figure 9: Pedigree Family 4

Third family

C10: *I just heard that P3's grandfather, from his father's side, used to have nose bleeds, but as a man who believed in herbs, he used to treat it with traditional medicines and he would come right.*

P3 could not have inherited haemophilia from his paternal grandfather because haemophilia is X-linked and only the Y-chromosome is passed on from father to son.

Even if P3's father was affected, he could only have passed the mutation on to his daughters, who would have presented as carriers (Fig. 10).

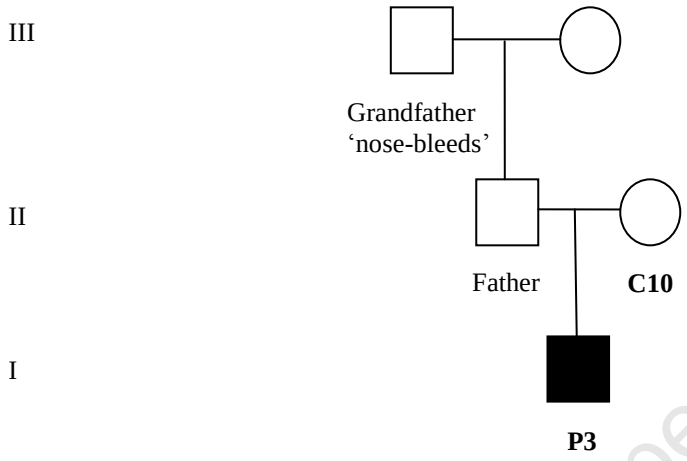


Figure 10: Pedigree family 10

4.4 General and cultural beliefs

Interview questions aimed to elicit how the participants understood this bleeding disorder and how they were making sense of the condition. They were asked what their fears or worries were and what other people thought of the condition.

4.4.1 Bleeding

A summary of the category 'bleeding', subcategory and codes under the theme 'general and cultural beliefs' is given in Table 15.

Table 15: Summary of the category 'Bleeding', subcategory and codes under the theme 'General and cultural beliefs'

Theme	Category	Subcategory	Codes
General and cultural beliefs	**Bleeding**	**Meaning of bleeding**	• Bleeding caused distress • caused headache and 'heartache' • not sure what to say • unable to explain bleeding • Possibly associated with outstanding Xhosa traditional rituals • Could be a (spontaneous) sign from the ancestors.

4.4.1.1 Meaning of bleeding

The unexplained bleeding and bruising caused distress and heartache to C4.

> C4: (what does this bleeding mean to you?) *At the time I had as serious problem and because of pain I was enduring I couldn't notice or take note of what was going on around me. I had a terrible headache. I had heartache. I lost touch of things.*

C3 was bewildered by the spontaneous bleeding and had no explanation for it.

> *C3: I am not sure what to say. If he was a girl, I would say it had to do with periods, but because he is a boy I don't know what to think. Bleeding is not something that just happens, it happens when you have had an operation, or if you have been involved in some accident, it does not just happen. That is why I cannot explain it.*

Two participants' husbands associated bleeding with outstanding cultural rituals.

> *C2 (husband): I usually associate it with some cultural ritual that must be done, I am not sure.*

> C8 (husband): *You mean the bleeding without being beaten? I don't know as I don't have expertise not unless he needs traditional rituals maybe. I think he has taken this*

after her mother that's what we were told by the doctor and that's where the problem lies. It can be possible that he wants Xhosa traditional rituals.

C9 believed that spontaneous nose bleeds are a friendly message from the ancestors.

C9: I know only of nose bleeding, for us in Xhosa the nose bleeding is not usually a serious or a fatal thing. According to us, nose bleeding is some sign (spontaneous) from our ancestors. May be they have thought of you and are now reaching out to you - Okay I hear you.

The spontaneous bleeding caused distress and bewilderment. Participants had no explanation for the bleeding episodes, especially as they occurred without obvious trauma. Some participants thought that it was connected to the ancestors and that outstanding rituals should be performed. Nosebleeds on their own are on the other hand not considered to be serious and would not require spiritual intervention. On the contrary, according to C9, they are just a friendly message from the ancestors reminding the living that they are there and that they are thinking of them.

4.4.2 Traditional medicine

A summary of the category 'traditional medicine', subcategories and codes under the theme 'beliefs' is given in Table 16.

Table 16: Summary of the category 'traditional medicine', subcategories and codes under the theme 'General and cultural beliefs'

Theme	Category	Subcategory	Codes
General and cultural beliefs	**Traditional medicine**	**Rituals**	• rituals or traditional ceremonies have not been properly attended to • illness may be a sign from the ancestors • outstanding rituals for the child or for the ancestors must be the reason why the child has not been cured • traditional healers confirmed that all necessary rituals had been performed • believes that patient had improved after rituals were performed
		Traditional healers	• wants to look for guidance from traditional doctors • cannot visit sangoma due to lack of funds • spiritual healers conflicts with her church beliefs • does not believe in traditional healers

In order to gain insight into how participants make sense of the condition afflicting their child questions were asked about what they thought should be done regarding the treatment of their child. Participants were also prompted to share their thoughts on why they think some people have certain conditions and others do not. Many participants believed that there was a connection between the cause of the condition and traditional rituals and that this had to be explored in order to find solutions.

4.4.2.1 Rituals

In Xhosa society, traditional healers appeal to their ancestors for help in diagnosing problems and prescribing remedies and routinely check to see if illnesses are caused by their clients having violated cultural norms and traditions (Nattrass, 2006). One boy's father, understandably, suspected that his child was affected because rituals or traditional ceremonies had not been properly attended to and was planning to look for guidance from traditional healers.

> C2 (husband)*: Yes, a child would be sickly when young, and turn around to be the most helpful when old. Like my son, he is sickly now, may be when he grows old that*

will stop, the illness may be a sign from the ancestors. The meaning of it could be that the there is an outstanding ritual that has to be performed for the child, or for the ancestors. When a child is sickly you would not know what to do, and overlook the possibility that it is related to some traditional ceremony that is required. You only realize at a later stage when the child is not getting cured. The issue of our rituals is a delicate and difficult one, it is not easy to just get into it.

Another participant's husband also thought that the disorder was linked to some (missing) rituals, because he could not imagine what else could cause the symptoms.

C8 (husband): *We sometimes think of that (rituals) honestly because what could be the cause of the lumps?*

C4, whose own father suffered an African death, related that the child's father also suspected outstanding rituals or ceremonies to be the cause of their son's condition.

C4*: He* (child's father) *says he suspect that it is because of the rituals or traditional ceremony that they have not attend to. And from my side I cannot say much because my parents died. My dad suffered an African death (*witch craft*) as a result he died suffering from a leg problem.*

C9 recounted that rituals were performed by the child's paternal family and that that improved his condition to some extend.

C9: The reason he went to Cifimvaba was for his family (father's side) to fix him. There was some relief after that until he came back to Cape Town. He came back with some boxes and he was a bit better. (By fixing what do you mean?)
Slaughtering, they slaughtered for him and performed cultural rituals.
(He came back better?) *Yes he came back better.*

A different participant doubted that tradition is linked to haemophilia because her traditional healer had confirmed that all necessary rituals have been performed. She did however contemplate the possibility that some ritual may have been overlooked.

> C1: *I don't think my tradition has got link to the bleeding because when I went to find out about the tradition perspective to this problem, we were not told to do any ritual because we've done all necessary rituals. Not unless there is something that we are missing, that we are not aware of, we have done everything*

While some families might have accepted the diagnosis and treatment for haemophilia they continue to depend on traditional methods to find reasons for the cause and to ultimately find a cure.

Traditional rituals are still very much intertwined in the daily life of most participants. It is believed that ignoring the spirits of the ancestors, who protect the living community, can cause problems that can manifest in poor health or misfortune (Wessells and Monteiro, 2004). A newborn's entry into the local community, for example, is marked by a series of rituals aimed at introducing the new baby to the community and ancestors and to get ancestral protection against sickness (Jewkes and Wood, 1998)

> C10: *I sometimes think may be there are some traditional ceremonies that need to be performed, but we have done most of those, especially those serious ones that are likely to cause this kind of problem. However, we are afraid to do some, for an example the "cutting of the finger".*

Some clans perform an additional ritual (*ingqithi or 'the cutting of the finger')* to gain ancestral protection. The ritual of *ingqithi* entails cutting off the last joint of the fourth (for girls) or fifth (for boys) finger (Jewkes and Wood, 1998). C10 in her search for an explanation or cure questioned whether all rituals had been performed, but had shied away from rituals that involve cutting because they could cause excessive bleeding.

In the traditional worldview, relationships with family, relatives, environment, ancestors and ancestral spirits are all interconnected and solutions to problems are sought in terms of all factors (Wessells and Monteiro, 2004). While participants were generally willing to accept the treatment of symptoms by western medicine, they continued to persue alternative solutions by consulting their spiritual healers who would be able to mediate with their ancestors. This consultation of dual systems amongst the amaXhosa, as mentioned earlier, has similarly been demonstrated by Sifunda et al. (2007).

4.4.2.2 Traditional healers

It has been mentioned previously that a large majority of South Africans still use traditional healers as their first call for medical advice. These practitioners, who are often also respected opinion leaders, are generally caring people who treat their clients holistically and consider all aspects of their lives (Kvale, 1995; Pretorius, 1999). Participants were asked what kind of treatment their child should receive and whether they thought that there were possible alternatives to western medicine.

C1 was adamant that she does not believe in traditional healers.

> C1: *No, we don't believe in those things.*

C2's husband believed that consulting with a traditional healer will give him guidance and help him understand why his child is affected with this condition.

> C2 husband: *I have even mentioned to my wife that may be I need to go away to Botswana, or maybe Johannesburg, Gauteng, or may be Durban, to "consult"* (getting in touch with the traditional doctors), *because I need some guidance on what this is and what its meaning is.*

C4 contemplated visiting a sangoma but was not able to due to lack of funds.

> C4*: One lady suggested that I should try taking the child to a Sangoma or traditional doctor so that I can ascertain what the problem is. I told myself that I won't go there because to pay a visit there I have to pay something. I don't have money as I am unemployed.*

She also considered visiting a spiritual healer, but is hesitant because it conflicts with the teachings of her church that does not condone spiritual healers. Another reason is, again, lack of funds.

> C4: *One person (child's paternal grandmother) say I should take him to a Spiritual Healer so that I can get to the bottom of this problem. She attends a spiritual healer's church. Even so I get discouraged to go there because of my church beliefs in my church Wesley we do not have a Spiritual Healer. I tell myself that I won't go there because I remember when I was home (Eastern Cape) we had a similar problem my relative's child was sick and he was going to be taken to the healer but I didn't have money at that particular time because I've used it in travelling.*

The concept of traditional medicine and traditional healing is an important part of the participant's model of health (Buehrmann, 1984; Pretorius, 1999) and health issues seem to be viewed and discussed, not only from the perspective of western medicine, but also from that of traditional medicine. Some participants reported that they were prompted by community and family members to consult traditional or spiritual healers in order to find a diagnosis or a cure.

The concept of health, in the African traditional sense, requires the balance of all facets of a person: body, mind and spirit as well as that the person is in balance with the family, community, or the forces of the natural world. An illness, except common illnesses such as colds or 'natural illnesses', are often ascribed to ancestor activity or witchcraft (Buehrmann, 1984). Even if it were accepted that the condition was caused by, for example, a mutation in a gene, most participants still turned to the spiritual world to explain the primary cause. They would refer to traditional methods to uncover what

caused the mutation in the first place and to determine what had to be done for the ancestral spirits to remedy this. Other studies have similarly shown that models of disease causation held by individuals are often in contrast with scientific concepts and may incorporate notions of bad luck, chance and fate (Walter et al. 2004).

4.5 Genes

Questions were asked to try and determine whether participants had any knowledge or concepts of what genes are and where they would be found in the body.

4.5.1 Understanding or knowledge of genes

A summary of the category 'understanding or knowledge of genes', subcategory and codes under the theme 'genes' is given in Table 17.

Table 17: Summary of the category 'understanding or knowledge of genes', subcategory and codes under the theme 'genes'.

Theme	Category	Subcategory	Codes
Genes	**Understanding or knowledge of genes**	**What are genes and how are they passed on?**	• does not know what genes are; possibly in the blood • drew blood from genes • aware of existence of genes (cells?)

4.5.1.1 What are genes and how are they passed on?

The concept of genes was not understood. Questioning was difficult because there is no equivalent Xhosa word for genes and the interpreter initially used the word *i-genes* which is borrowed from English. During feed-back and discussion with the researcher after the interviews this issue was discussed and it was decided that the interpreter would try and paraphrase. The translated transcripts however revealed that on some occasions

the interpreter misrepresented the word ‘genes’ and used misleading expression by referring to ‘white blood cells’, ‘cells that defend your body’ and ‘blood cells’.

C9: (There’s sometimes talk of cells that defend our bodies from diseases, have you ever heard of them?) *I don’t know. I just know that they exist and don’t know much*

C10: (Where do you think the white blood cells are in your body?) *I think they are around the body. (P3’s father says he thinks they are in the veins or in the blood.)*

C1: (There are blood cells. Have you heard of them?) *I don’t know that thing. I just hear about the blood cells. I know about joints, the body parts.*

Generally participants did not know what or where genes are and after some prompting suggested that they might be in the blood. Many communities use blood as a metaphor for the medium through which conditions are inherited (Featherstone, 2005). People use expressions like ‘it’s in the blood’ and ‘blood’ links generations when people speak about blood lines and blood relatives.

C7.: *I don’t know and I don’t want to lie. The only thing that I know is when the child is born he/she could be like her mother or father. But I don’t know how. I should think in the blood.*

C3: *Are they not in the blood?*

C2: *I think they are in the blood.*

The questions referring to genes or cells again seemed to intimidate the participants who might have had the impression that they were being tested. They did not know what the interpreter was referring to and were unsure as to how to answer. The wording of the questions may have influenced the responses and it would be necessary to find more

suitable isiXhosa words or expressions to ask about or ultimately explain genetic inheritance.

The concepts of ‘risk’, ‘genes’, ‘DNA’ or ‘chromosomes’ are not easily understood even when there are no language barriers. Research in the western world has shown that, although there is mention of these terms in lay accounts, most do not specify how these may be related to inherited characters and there is generally no clear idea of what exactly is transmitted between generations (Richards and Ponder, 1996; Richards, 1998).

Recent research has shown that it is important for health care providers to know what participants believe and understand about genetics when genetic-related health care issues are discussed (Lanie et al., 2004). This will equip health care providers to build on their level of understanding, ascertain whether there are any misunderstandings and to communicate ideas about the nature of genetics and haemophilia. Patients and families will benefit by getting a more complete picture of the condition that is affecting them and by becoming empowered towards making more informed decisions about their health care as well as the genetic implications of their condition.

The major findings and implications of the study will be summarised in the conclusions and recommendations in Chapter 5.

Chapter 5: Conclusions and Recommendations

Conclusions

The aims of this research were to explore the level of understanding of basic genetic concepts among Xhosa-speaking mothers or caregivers of patients with haemophilia; to investigate the cultural background of the participants; to explore the participant's understanding of genetic inheritance and its implications for genetic counselling and to describe how caregivers of patients with haemophilia experience living with this condition.

Analysis of ten interviews suggested that participants, their families and their community had very little, if any, background knowledge of haemophilia. It also appeared that health care workers in rural areas of the Eastern Cape similarly had a poor understanding of haemophilia and its pathology, and that patients with haemophilia did not receive adequate medical care in those areas.

Living with haemophilia posed a heavy burden on caregivers and their families, especially as occurrence and nature of symptoms were unpredictable. Daily life was often severely disrupted and participants had to repeatedly and unexpectedly rush patients to hospital. Participants seemed reluctant to speak to their community about haemophilia and the challenges they faced. Several participants maintained that it was best to keep to oneself in the urban environment and only a few could rely on help from neighbours during crisis situations.

Some participants had migrated to Khayelitsha from the Eastern Cape to be close to medical centres, but continued to regularly visit and maintain close ties with their rural families. The extended family in those rural areas was generally informed, especially if they needed to take care of patients in the absence of the caregiver.

Caregivers were generally deeply affected by their children's condition and, in addition to constantly feeling anxious, also incurred considerable indirect financial costs. There was no known family history amongst any of the participants and none of the children were diagnosed at birth. Participants where thus ill prepared and experienced fear and uncertainty when confronted with typical symptoms like frequent nose bleeds and bruising. The unpredictable nature of the disorder also affected employment and employment options for some.

The interviews showed that participants, despite frequent contact with medical professionals, were often ignorant about treatment for haemophilia and were unfamiliar with terms used by professionals. They generally seemed to trust medical professionals in treating symptoms and tended to leave the locus of control with them. Similarly, participants seemed to have no or, at most, only basic knowledge of the cause or pathogenesis of haemophilia. Many mentioned that they wished to turn to traditional medicine to find answers to the cause of the condition.

Genetic bleeding disorders have historically been recognized by cultures that practice ritual circumcision. Responses to the interview questions did, however, not reveal whether any such condition was likewise recognized amongst the participants, although the amaXhosa have practiced ritual circumcision for hundreds of years**.**

Participants generally referred to inheritance in connection with physical, behavioural and moral characteristics that are passed on by parents, relatives or long-dead ancestors. It further became apparent that participants had no clear understanding of Mendelian genetics and that previous explanations of inheritance given by medical professionals had either not been understood or been accepted. These preliminary findings could have important implications for health care providers and genetic counsellors as they suggest that counselling interactions may not have adequately informed participants regarding genetic implications of haemophilia.

It is essential that health care providers and counsellors are aware of the issues faced by isiXhosa speakers living with haemophilia and that these issues are focussed on and discussed with caregivers and their families in either isiXhosa or, if that is not possible, in the presence of an interpreter. Support groups could be used as platforms to educate and encourage members at a level and in a language that is appropriate for that particular community. The attendance at the existing support group meetings was unfortunately poor.

Recommendations

The following objectives guided the process of inquiry

Specific objectives of this study were to explore the possibility of:

- developing strategies to improve the ability of isiXhosa-speaking families affected with genetic disorders to make informed decisions;
- improving the general genetic management of haemophilia families in South Africa;
- developing strategies for health care workers to explain genetic terminology to the Xhosa-speaking population of South Africa; and
- developing strategies for interpreters to effectively communicate genetic concepts to Xhosa-speaking individuals in South Africa.

The following recommendations in terms of future research can be made:

1. This research showed that there was little awareness about genetic diseases among the participants which indicates that there is a need for *socio-culturally tailored, language-specific* education for families with haemophilia. A recent report on haemophilia-care in South Africa highlighted the need for genetic counselling (Mahlangu, 2009). To promote better care, genetic counsellors might be motivated to interact with the support group members at the monthly meetings. Genetic counsellors could thus play an important part in treatment centres or during support group meetings, to educate not only patients, but also to

inform mothers and sisters of patients with haemophilia about the genetics of the disorder and the hereditary risks involved should they decide to have children. Experienced interpreters, with broad life skills and good interpersonal relationships should be available if education or counselling in isiXhosa is not possible. This would give patients and families a more comprehensive picture of the condition that is affecting them, the genetic implications of their condition as well as to help them make more informed decisions about their health care. In order to develop guidelines to explain genetic concepts to participants and isiXhosa speakers in general it would be advisable to consult a range of people in different roles or positions. This may be achieved through focus group discussions with isiXhosa speaking health professionals, members of the haemophilia support group, genetic counsellors and possibly traditional healer. These discussions could help explore ways of paraphrasing or explaining genetic concepts such as genes, chromosomes or mutations in isiXhosa and also examine the options of exploring a possible confusion between inheritance and infection.

2. In order to effectively counsel families with haemophilia and to integrate new information with prior understanding, genetic counsellors or medical professionals need to have a good understanding of the genetic knowledge base of participants. Beliefs about the cause of a condition are often culturally determined and in order to address any misunderstandings, health care providers should be aware of the cultural base that shapes the thoughts and actions of their clients (Cohen et al., 1998). Respect for cultural traditions will also help build a foundation of trust between counsellors, health care professionals and patients and their families (Turner et al., 2004). Future research should thus identify concerns that are culturally-based and develop strategies for addressing them in a meaningful way.

3. Follow-up interviews with a more experienced interviewer who has a background in science or genetics and is able to paraphrase genetic concepts may give deeper insight into how genetic inheritance is understood.

Counsellors need tools with which they can probe the different worldviews of their clients. In order to sensitize counsellors to African worldviews and to promote awareness of the difference between the world of science and the world of spiritual Africa, additional interviews with traditional healers should be conducted. Culture determines language patterns, behavioural patterns and patterns by which individuals interact with others. Culture also affects the means by which knowledge is transferred and future interviews should aim to determine how knowledge, including inter-generational knowledge, is imparted within Xhosa culture. These interviews should also aim to clarify how the 'mechanism' of passing on traits or conditions from generation to generation is understood and what terms should be used when referring to inheritance.

4. Counsellors have no linguistic reference system to guide them on how to develop suitable narratives to impart scientific information. Focus group discussions with isiXhosa speaking haemophilia nurses, interpreters, members of the haemophilia support group, genetic counsellors and traditional healers should help establish when and why certain isiXhosa words are used and to help understand what narratives can be built with these words. Members of the focus group could then investigate whether narrative lines can be built using these words to impart scientific information.

5. Support group education is important to back up information given by medical professionals and counsellors. A survey of families with haemophilia in Khayelitsha could elucidate the reasons for lack of attendance of the Khaya ROCK support group and help determine strategies to improve participation. It would be important to motivate more volunteers as well as to promote support for these meetings from the health care sector and counselling professions.

6. Tsang et al. (2003) have emphasized that it is important for researchers to investigate actual clinical processes, in this case counselling sessions, to determine which processes are associated with positive and negative counselling

outcomes. In order to test the effectiveness of cross-cultural genetic counselling it is recommended that further research examines interactions between genetic counsellors and isiXhosa haemophilia families. This may elucidate if and how client and counsellor differences based on ethnicity and language are negotiated during these interactions.

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Appendices

Appendix i: Ethics approval

UNIVERSITY OF CAPE TOWN

Health Sciences Faculty
Research Ethics Committee
Room E52-24 Groote Schuur Hospital Old Main Building
Observatory 7925
Telephone [illegible] • Facsimile [illegible]
e-mail: [illegible]

17 October 2006

REC REF: 383/2006

Dr J Greenberg
Clinical Laboratory Sciences

Dear Dr Greenberg

PROJECT TITLE: AN INVESTIGATION INTO THE LEVEL OF UNDERSTANDING OF BASIC GENETICS OF MOTHERS OR CAREGIVERS OF XHOSA HAEMOPHILIA PATIENTS

Thank you for submitting your study to the Research Ethics Committee for review.

I have pleasure in informing you that the Ethics committee has **formally approved** [illegible] mentioned study.

Could you please add the following to the information sheet:

- Your contact details should mothers have any queries before or after the interviews.
- Contact details for Prof Marc Blockman, chairperson of the Research Ethics Committee, should participants have any questions about their rights as research participant.

Please note that the ongoing ethical conduct of the study remains the responsibility of the principal investigator.

This serves to confirm that the University of Cape Town Research Ethics Committee complies to the Ethics Standards for Clinical Research with a new drug in patients, based on the Medical Research Council (MRC-SA), Food and Drug Administration (FDA-USA), International Convention on Harmonisation Good Clinical Practice (ICH GCP) and Declaration of Helsinki guidelines.

The Research Ethics Committee granting this approval is in compliance with the ICH Harmonised Tripartite Guidelines E6. Note for Guidance on Good Clinical Practice (CPMP/ICH/135/95) and FDA Code Federal Regulation Part 50, 56 and 312.

Please quote the REC. REF in all your correspondence.

lemjeds

Appendix ii: Information sheet - English

An investigation into the level of understanding of basic genetics of mothers or caregivers of Xhosa Patients with haemophilia

Investigator: Gabriele Solomon, MSc student, Division of Human Genetics, Department of Clinical Laboratory Medicine, University of Cape Town. Tel 021-4066425

This information sheet is part of an informed consent process for a research study. It will give you information that will help you decide whether you wish to volunteer for this research and will help you understand what the study is about and what will happen during the course of the study. If you have questions at any time during this research study, you should feel free to ask them and should expect to be given answers that you completely understand.

Why is this study being done?

This study is being done to find out how you as first-language Xhosa speaking parents or caregivers of Patients with haemophilia understand or explain inheritance. We would also like to know how you understand genes and what your thoughts are on what effects these genes may have. We are collecting this information to help parents and counsellors/nurses/doctors/interpreters understand each other better.

What sort of questions are we going to ask?

We would like to ask you some information about yourself such as your age, to what standard you went to school, what other languages you speak, your place of residence and your place of birth. We will then ask you how you understand the inheritance of haemophilia, what this condition means to you and how it affects your and your child's

life. We would also like to know how you understand genes, inheritance and genetic conditions in general.

How long will the interviews last and how will they be structured

The interview will last about one hour. There will be an English-speaking researcher asking the questions and an interpreter who will translate everything for you. Depending on your decision, the interview will take place at your home or at the Khaya Rock support group centre in Khayelitsha. We would like to record the conversation so that we can easily remember everything you say.

In a few weeks time we may contact you again to meet with the other people we have interviewed to discuss haemophilia, inheritance and genes in a group. We will pay you R60.00 for travelling expenses when you come back for this group session.

Your participation is entirely voluntary and you may withdraw at any time during the interview or during the study.

Confidentiality

We will make sure that none of your personal details will be made public. The information will only be used for this study and if anything you said will be used, it will be on an anonymous basis (i.e. your name will never be mentioned).

Contact details

Should you have any queries before or after the interviews please contact the principal investigator: Assoc. Prof. J. Greenberg, Department of Clinical Laboratory Sciences, Division of Human Genetics, UCT. Tel: 021-4066299.

Should you have any questions about your rights as a research participant please contact Prof Marc Blockman, chairperson of the Research Ethics Committee, UCT. Tel: 021-4066338

Appendix iii: Information sheet -Xhosa

ULWAZI NGOMTHATHI-NXAXHEBA KUNYE NEFOMU YEMVUMELWANO EYAZISIWEYO YOPHANDO OLUQUKA IZIFUNDO ZEMFUZO

ISIHLOKO SEPROJEKTHI YOPHANDO: Ingxaki yokopha

UMPHANDI OYINTLOKO: Gabriele Solomon

IDILESI: Division of Human Genetics, Department of Clinical Laboratory Medicine, University of Cape Town.

INOMBOLO YOQHAGAMSHELWANO: 021-4066425

Singathanda ukukumema ukuba athathe inxaxheba kwisifundo sophando esiquka uhlalutyo lwe-DNA (lwemfuzo). Nceda thatha ixesha lokufunda ulwazi oluvezwe apha oluzakuthi luchaze iinkcukacha zale projekthi. Nceda buza nayiphina imibuzo emalunga nayiphina indawo ongayiqondiyo ngokupheleleyo kubasebenzi besi sifundo okanye kugqirha. Kubaluleke kakhulu ukuba waniliseke ngokupheleleyo yinto yokuba ucacelwe kakuhle ukuba yintoni ebangwa sesi sifundo kwaye ungabandakanyeka njani. Kwakhona, ukuthatha kwakho inxaxheba kungentando yakho ngokupheleleyo kwaye ukhululekile ukuba ungarhoxa ekuthatheni inxaxheba. Ukuba uthi hayi, oku akusayi kuchaphazela ukungavumi kwakho nangayiphina indlela. Ukwakhululekile ukuba ungarhoxa kwesi sifundo naninina, nkqu nokokuba ubuvumile ukuthatha inxaxheba ekuqaleni.

KUTHENI KUSENZIWA OLU PHANDO NJE?

Olu phando lwenzelwe ukuqonda kuthi bantu bantsundu ukuba sinolwazi na ukuba ufuzo lwenzeka njani. Sifuna nokuqonda ngolwazi lwakho ngokunxulumene nofuzo lwezigulo nokuba zikuchaphazela njani kwakunye neziphumo ezo zochaphazeleko. Senza olu phando khonukuze sifumane ulwazi oluphangaleleyo sikwazi ukunceda abazali/onontlalontle/ogqirha/abongikazi/netoliki sibenokwazana ngcono.

YIMIBUZO ENJANI ESIZAKUKUBUZA YONA?

Sicela ukufumana inkcukacha ngawe ezifana neminyaka yakho, ufunda kweliphi okanye waphela kwiliphi ibanga esikolweni, zeziphi ezinye ilwimi ozithethayo, indawo ohlala kuyo nozalelwe kuyo. Sizokuphinda sikubuze ukuba unolwazi olungakanani malunga nokosuleleka kwezifo ngenxa yemfuzo. Unolwazi olungakanani lwesisifo sokopha (haemophilia) ngenxa yeyokunikezelana kwencindi yomzimba.

IZAKUTHABATHA ITHUBA ELINGAKANANI OLUDLIWANO-NDLEBE?

Luzakuthabatha iiyure ezimbini. Abantu abazakubakho koludliwanondlebe ngumphandi othetha ulwimi lwasemzini Isingesi nomntu ozakube etolika ngesiXhosa. Kuyakuxhomekeka kwisigqibo sakho esizakuthatyathwa nguwe ukuba ufuna uphando olu lwenziwe apho uhlala khona okanye e Khaya Rock support centre e Khayelitsha. Sicela ukulushicilela udliwanondlebe ukwenzela ukuba sibenako ukukukhumbula yonke inkcazelo ozakusinika yona.

Emveni kweveki ezimbalwa sizakuphinda sidibane nawe kunye nabanye abantu ebesithabathe udliwanondlebe nabo sizokuxoxa siliqembu ngendaba yokopha ngenxa yokwasuleleka ngokunikezelwa kwencindi yomzimba. Sizakunihlawula amashumi amathandathu eerandi (R60.00) xa nithe nabuya ukuza kudibana nabanye abantu. Le mali yeyendleko zokukhwela.

Ukuthabatha inxaxheba akunyanzeliswa ungarhoxa nangawuphi na umzuzu.

IZAKUGCINWA NJANI IMFIHLELO YAKHO?

Siyakuthembisa ukuba amagama akho akasokuze apapashwe. Iinkcukacha osinike zona zakusetyenziselwa esi sifunda kuphela.

UQHAGAMISHELWANO

Xa unemibuzo ngoluphando ungatsalela inqununu u Assoc. Prof. J. Greenberg, Department of Clinical Laboratory Sciences, Division of Human Genetics, UCT. Tel: 021-4066299

Xa unemibuzo malunga namalungelo akho njengo mthathi nxaxheba nceda utsalele u Prof. Marc Blockman, chairperson of the Research Ethics Committee, UCT. Tel: 021-4066338.

Appendix iv: Consent form - Xhosa

IPHEPHA LESIVUMELWANO NGOPHANDO LWEZIFUNDO

Ingcaciso ngamagama asetyenziswayo xa kuphandwa ingxaki Zokopha/Haemophilia ezinxulumene nemfuzo nokuqonda umehluko walamagama ngabantu abathetha isiXhosa

Gabriele Solomon, MSc student, Division of Human Genetics, Department of Clinical Laboratory Medicine, University of Cape Town

(021 4066425)

Utyikiko lonobumba

1. Ndilufundile okanye ndalufunda olu lwazi kunye nefomu yemvumelwano kwaye ibhalwe ngolwimi endiliciko nendikhululekileyo kulo. _____

2. Ndibenalo ithuba lokubuza imibuzo kwaye yonke imibuzo yam iphendulwe ngokwanelisayo. ____

3. Ndiyakuqonda ukuba ukuthatha inxaxheba kolu phando kube kukuzithandela kwam kwaye ndingarhoxa nangawuphi umzuzu. _____

4. Ndiyavuma ukuthatha inxaxheba kolu phando. _____

5. Ndiyavuma ukwenza udliwano-ndlebe oluzakushicilelwa _____

6. Ndiyavuma ukuba ushicilelo ngetape lungasetyenziswa nakwezinye izifundo kodwa angapapashwa amagama am.

Ngokutyikitya kwam le fomu ayithethi ukuba ndibophelelekile kuphando lwesisifundo, ndingarhoxa nangawuphi na umzuzu.

Igama lomthathi nxaxheba womthathi-nxaxheba		Umhla	Umtyikityo
Igama lomphandi		Umhla	Umtyikityo womphandi

Appendix v: Consent form - English

An investigation into the level of understanding of basic genetics of mothers or caregivers of Xhosa Haemophilia patients

Gabriele Solomon, MSc student, Division of Human Genetics, Department of Clinical Laboratory Medicine, University of Cape Town (Tel: 021 4066425)

		Please Initial
1.	I confirm that I have read and understand the information sheet for the above study and that I have had the opportunity to ask questions.	_____
2.	I confirm that my questiones have been answered in a way that I could understand.	_____
3.	I understand that my participation is voluntary and that I am free to withdraw at any time, without giving a reason.	_____
4.	I agree to take part in the above study.	_____
5.	I agree to the interview consultation being audio-recorded	_____
6.	I agree to the use of quotes from the interview in scientific publications, as long as my name is not mentioned	_____

Signing this consent form does not mean that you have to take part in this study; you may still withdraw at any time.

_____	_____	_____
Name of Participant	Date	Signature
_____	_____	_____
Name of Researcher	Date	Signature

Appendix vi: Interview schedule

An investigation into the level of understanding of basic genetics of mothers or caregivers of Xhosa haemophilia patients

Interview Schedule

We are doing this study to find out what haemophilia means to you and how you as a first-language Xhosa speaker understand inheritance and genes. We would like to know what you actually think, and your ideas and knowledge of this are of great interest and importance to us.

First we need to ask you a few questions about yourself

What is your relationship to the child? (are you related? Are you a biological relative?)
What is the child's name?
If not related: childs family name/father's clan/mother's clan

Name: What name do you use?
Are you married or single?
What is your family name?
Is that your husband's name?
What is your clan name (paternal)?
(maternal)?

If single: What is your relationship to the child's father
Is the child still in the father's clan

Age

Gender

Place of birth

Place of residence

How long resident in Cape Town

Are you migrating? (going home for holidays? Taking HP child to Transkei/Ciskei?)

Level of formal schooling

Now we would like to talk about your child's illness and where it comes from

1. **How** did you become aware that your child has a (this) problem?

2. When did you first become aware of this problem?
   Prompt: Where you aware of anything before the doctors told you?

3. Do **you** think the child was born with it? (was it there at birth?)

4. What did you know about this condition before the child was diagnosed?
   Have you heard about it before?

5. What do you think has caused the *bleeding* (problem) (take lead from how mother refers to condition)
   Prompt: Have you got any other explanations/thoughts?
   What did you notice?

6.

- How would you explain (eg to someone else, **a friend**) what is wrong with your child?
- Is there another explanation? (cultural explanation?)

7. What kind of treatment should our child receive? What do you think? (besides western medicine?)

8. What do you fear most (**what worries you most**) about your child's 'problem'?

9. Do you think boys and girls can have this problem?

10. What does blood (this bleeding) mean to you?
    Prompt: in your culture?

11. When you think about inheritance (descent) 'ufusile' – what do you think about?
    (inheritance can refer to: behaviour, characteristics, disease, bewitchment, traditional healing – inheritance belongs to each individual)

12. Is there a known illness in your family that has been passed down?

13. Do you know of other family members (cousins, uncles, brothers, fathers) that had this disease?
    Do you know of family members of previous generations that had this disease?

14. a) Does everyone in your clan know about your sons condition?
    b) Do other people know about your son's condition?

15. a) What do the members of your clan/husbands clan think about the problem and what do they think caused it?
    b) What do other people think about the problem and what do they think caused it?

16. How does your son's problem affect you in your community?
    Does your son play with other children?
    Do other mothers allow their children to play with your son?
    Has this condition changed your life?

Inheritance

17. Why do you think that it is that some people get ill and others don't?

18. Where do you think genes may be located in someone's body?

19. Can you tell me what you mean (understand) if you say that the condition is inherited? (Lanie et al., 2004)

20. Who do you think a (the) condition is inherited from?
    Prompt: mothers side
    Fathers side?
    Why ?

Appendix vii Interviewing guidelines

Interviewing guidelines for the MSc (Med) project:

An investigation into the level of understanding of basic genetics of mothers or caregivers of Xhosa patients with haemophilia

Introduction

The interview is one of the major sources of data collection in qualitative research. The interview itself is a form of dialogue between interviewees (person being interviewed) and interviewers (and, or interpreter). The record of that interview then becomes representative of the dialogue and is used by the researcher to analyse and interpret the data (University of Sheffield, http://www.computing.dcu)

The nature of the relationship between the interviewer and the interviewee is complex and it is important to be sensitive to challenges that are likely to arise. The interview can be seen as a hierarchical relationship, 'ruled' by the interviewer (interpreter) who directs the dialogue and usually has several hidden agendas. The interviewee in turn may, in an attempt to regain control, not answer or deflect questions. It is also possible that interviewees are not able or willing to articulate particular concerns (Creswell 2007). They may be reluctant to expose their past or to go on record about difficult aspects of their lives or they may be unwilling to admit ignorance.

Interviewing guidelines

There are no recipes for successful interviewing but Patton (1987) has recommended some useful guidelines. These include to ask truly open-ended questions, to avoid leading questions, to listen attentively and to let the person know he or she is being heard and to be sensitive to how the persons is affected by and responds to the different questions. While the interviewer must maintain control of the interview it is important to

always treat the person being interviewed with respect and to remember that it is a privilege and a responsibility for the interviewer/researcher to peer into another person's experience. Enjoy interviewing.

Role of the interpreter

Pitchforth, E. and van Teijlingen (2005) refer to a passive and an active role for the interpreter. In the passive model the researcher will ask a question (e.g. in English) and the interpreter will interpret the question to the interviewee and the response to the researcher. This is very time consuming and the researchers found that this method does not allow the conversation between participants to flow. The dialogue was constantly interrupted by interpretations and the interviews tended to become disjointed. In the active model the interpreter carries out most of the interview. Although this model hands over more responsibility to the interpreter it allows him or her to build a better relationship with the interviewees and allows the interview to flow better. A disadvantage of this model is reduced control from the researcher's perspective. It also requires the interpreter to quickly become familiar with the aims of the interview and interview schedule.

In this study the active model has been employed because it allows the interpreter to build a good rapport with the interviewees and the interview can progress smoothly.

The use of probes

One of the key techniques in good interviewing is the use of probes (Patton, 2002). The responses to open-ended questions can vary to a great extend between interviewees. Because the outcome of such an interview is unpredictable, interviewers will usually use probes to get more information or to subtly redirect the responses. Generally probes are used to enable the interviewer to get more information about the feelings, opinions, beliefs and perceptions of the interviewees. Patton (2002) maintains that one of the key techniques in good interviewing is the use of probes and identifies three types of interview probes:

1) **Detail-oriented probes**

In our conversations with colleagues or friends we ask each other questions to get more detail. We most easily ask these follow up questions when we are genuinely curious.

Examples of such questions include:

Who was with you?

What was it like being there?

Where did you go then?

When did this happen in your life?

How are you trying to deal with the situation?

2) **Elaboration probes**

This probe is designed to encourage the other person to tell us more. We may remain silent but attentive, gently nod our head as the person talks and softly voice uh-huh every so often. We may also encourage the other person to simply continue talking.

Examples:

Tell me more about that.

Can you give me an example of what you are talking about?

I think I understand what you mean.

Talk more about that, will you?

I'd like you talk more about that.

3) **Clarification probes**

The interviewer may be unsure what the interviewee means and will gently ask for clarification. It is important to communicate that the interviewer has problems in understanding and that this is not the fault of the interviewee.

Examples:

I'm not sure I understand what you mean by that.

Can you help me understand what that means?

I'm having trouble understanding the problem you've described.

Can you talk a little more about that?

I want to make sure I understand what you mean.

Would you describe it for me again?

I'm sorry. I don't quite get it. Tell me again would you?

Role of the researcher

During the current study the role of the researcher, because of language issues, is mostly passive and mainly that of an observer. The researcher will however always be present and as the interpreter becomes aware of key points he or she will summarise the interview to the researcher to allow for additional questions to be asked**.**

References

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Pitchforth E. and van Teijlingen E. 2005 International public health research involving interpreters: a case study from Bangladesh. *BMC Public Health* 5: 71 (http://creativecommons.org/licenses/by/2.0).

Electronic references

University of Sheffield, The qualitative paradigm. (http://www.computing.dcu.ie/~hruskin/RM2.htm)

Appendix viii: Transcription guidelines

Transcription guidelines for the MSc (Med) project:

An investigation into the level of understanding of basic genetics of mothers or caregivers of Xhosa patients with haemophilia

Introduction

In order to gather biographical information from mothers or caregivers of patients with haemophilia, semi-structured interviews are used in this project. These interviews aim to explore the subjects' beliefs about inheritance as well as their feelings, personal experiences and reasons for actions related to the inherited disorder haemophilia.

The transcribed verbal communication generated by the interviews needs to be thoroughly analysed. To achieve this, the data has to be carefully described and summarised by the researcher. The transcribed interviews thus serve as documentation for the researcher and it further allows someone else to challenge or confirm the interpretation of the data. (Maxwell, 1992). If the interview is conducted with an interpreter in a language that is foreign to the researcher, it is important to keep a verbatim (transcribe word for word in the language spoken) record of the interview. This record will include terms used in the respective mother tongue and can then be reviewed and translated by an interpreter who can help understand any unfamiliar terms and concepts that were used during the interviews (Carrese, 2000).

Descriptive accuracy is a primary aspect of the validity of qualitative research. It is thus an important concern for qualitative researchers to present accurate facts, without attempting to interpret the things they saw and heard (Maxwell, 1992). The transcripts must enable the researcher to give detailed descriptions and must also allow him or her to include in-depth, direct quotations from the interviews to highlight the informants' experiences, attitudes, beliefs and thoughts (Genzuk, 2003).

Transcription Guidelines

1. **Do not change anything.** Accurately represent each speaker's words, conversational quality, and speech patterns.
2. **Transcribe** conversations in the respective language – Xhosa, when Xhosa is spoken, English when English is spoken.
3. **Spelling.** Use correct (not phonetic) spelling of words, even if they have not been pronounced quite that way (but do not try to improve on the sentence structure and grammar).
4. **Be complete.** Be careful to transcribe all the words and 'transcribable' sounds (including sounds like "*ah, hmm…, well…..er…… what I mean*" as they are indicative of emotions expressed. Use parentheses [ ] with discretion to note audible expressions of emotion such as *[laughs]* when one speaker does, *[laughter]* when both do, or *[pounds fist on table]*, or *[tape turned off and on again]*, and to describe what is happening *[reading from newspaper]*.

Some transcription conventions:

.	Pause of less than a second
(1)	Pause or gap in seconds
/Slash Marks/	Overlapping speech
[*description*]	Non-verbal behavior and other editorial comments
italics	Sounds, e.g. *ah* …etc
hhh	Audible breath, in or out
CAPS	Relatively loud speech

Adapted from: Transcription Guidelines (http://www.iei.uiuc.edu)

It is generally understood that transcription takes 4-6 times the length of the interview, i.e. if the interview is one hour long, transcription will take 4-6 hours.

References

Carrese, J. (2000) Bridging cultural differences in medical practice: The case of discussing negative information with Navajo patients. *J Gen Intern Med* 15:92-96

Maxwell, J.A. (1992) Understanding and validity in qualitative research. *Harvard Educational Reviews* Fall 62, 3:279-300

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Genzuk M (2003) A Synthesis of Ethnographic Research. Occasional Papers Series. Center for Multilingual, Multicultural Research, Rossier School of Education, University of Southern California. Los Angeles. (http://www-rcf.usc.edu/~genzuk/Ethnographic_Research.pdf)

Transcription guidelines (http://www.iei.uiuc.edu/TESOL/transguide.html)

Appendix ix: Self-reflection

The researcher is white and was born and brought up in Germany. When coming to South Africa 25 years ago, at the age of 29, the researcher had to fit into a foreign country, improve her knowledge of English and learn to speak Afrikaans. The researcher is thus aware of the difficulties of being an 'outsider' and of trying to converse in a language other than the mother tongue.

The researcher has a background in human genetics and is familiar with genetic disorders and their inheritance patterns. Independent of that the researcher had been familiar with haemophilia as it was often been discussed during history lessons at school in Germany. A famous pedigree of haemophilia started with Queen Victoria of England who was a carrier of the genetic disorder. She passed haemophilia on to her son Leopold, and several of her daughters were carriers. Her daughters in turn passed haemophilia on to the royal families of Spain, Germany and Russia. Awareness in Germany was also raised through popular stories of Rasputin who claimed to be able to help the Russian empress's son, Tsarevich Alexis, who had haemophilia. The Russian empress, Alexandra Feodorovna was Queen Victoria's granddaughter and one of the carriers of the mutated haemophilia gene.

Various studies have emphasized the importance of critical self-awareness towards developing multicultural competence and have highlighted that self-awareness includes admission of ones own racist attitudes and beliefs (Carter, 1991; Richardson and Molinaro, 1996; Sue et al., 1992; Ancis and Szymonski 2001). While there are people living without means in Germany, they are looked after by a very well developed social network. Conditions, e.g. squatter camps, under which the poor of South Africa live, are virtually non-existent. Having been raised in Germany the researcher was, because of that countries history, well aware of the effects of racial segregation and racial injustices. Until coming to South Africa 25 years ago, at the age of 29 however, the researcher had had little personal experience of racial inequalities and only gradually learned to

understand the undertones of problems that exist in this country. The privilege associated with whiteness was recognised and the researcher endeavoured to continuously examine and query these privileges.