People whose siblings have had a premature heart attack: their views and experiences in the era of human genomics

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Abstract

Coronary heart disease (CHD) is a major health problem, and a key area of health policy. Family history of CHD is known to be an important risk factor, and having a sibling affected by CHD increases an individual's risk significantly. Genetic technology may offer new mechanisms for prevention and treatment. However, It is unclear how individuals at high risk of CHD perceive their personal risk and view the possibility of genetic susceptibility testing in the future.

A mixed methodology approach was adopted employing both qualitative and quantitative methods to investigate the views of unaffected siblings of people who had experienced a heart attack under the age of fifty years. For the qualitative phase 20 semi-structured interviews were undertaken. All interviews were audio taped, transcribed verbatim and analysed using the constant comparison method. Participants appeared to have a strong sense of detachment from their siblings' heart attack, and some were very resistant to the prospect of genetic susceptibility testing. Additionally, some participants felt that their concerns were trivialised by health care professionals when they sought reassurance.

For the quantitative phase a questionnaire was developed and posted to a group of unaffected siblings (n=59) and a comparison group where there was no family history of CHD (n=148). Unaffected siblings were significantly more likely than the comparison group to view their risks of developing CHD as high, and to fear CHD. Broadly similar beliefs about genetic technology and the possibility of genetic susceptibility testing were observed in both groups. Only 30% of unaffected siblings sought medical help or reassurance following their siblings' heart attack.

This study has highlighted some important issues about how individuals interpret family history of CHD, and how advances in genetic technologies are viewed. Of particular concern are the experiences of people seeking reassurance from health professionals, which has raised questions about how the cardiac risks of this group of people can be assessed more systematically.

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Chapter 1 Introduction

1.1 Background

1.1.1 Overview

The study reported here investigated the risk perceptions, health beliefs and views about genetic research, and the possibility of future genetic susceptibility testing, amongst a group of people who are probably at high risk of developing heart disease. Coronary heart disease (CHD) is a major health problem in the Western world, and a positive family history of CHD, particularly myocardial infarction (MI) (heart attack), is arguably one of the most important risk factors for an individual (Marian, 1998, Kardia et al., 2003). Previous research has investigated lay perceptions of the causes of heart disease (Davison et al., 1991) and how a family history of CHD is interpreted (Hunt et al., 2001). However, the majority of these studies have focused on the general population. Other studies have investigated beliefs about how individuals interpret risk for familial hypercholesterolaemia (Senior et al., 2002), which is known to be a single gene disorder.

The precise nature of the genetic components of CHD is not yet fully understood (Samani & Singh, 2001), but it is anticipated that advances in molecular technology may elucidate the genetic involvement, and offer the possibility of novel mechanisms for the treatment and prevention of CHD in the future (Collins, 1999). A high risk of illness may not necessarily motivate an individual to modify their behaviour (Marteau & Lerman, 2001). However, it remains unclear how individuals, who may be at substantial risk of developing heart disease by virtue of their family history, assess their individual risk and how they view these advances in genetic technology. People currently unaffected by CHD who have a sibling who has experienced a premature heart attack, are a group of people who have been little studied, and whose views and experiences merit further investigation.

This chapter will discuss the epidemiology and aetiology of CHD (section 1.1), and provide an overview of research on *lay* epidemiology and causal attributions of heart

disease (section 1.2). An overview of genetic developments including views about predictive testing will also be provided (section 1.3) before summarising this material (section 1.4). The current study employs a mixed methodology approach, with both qualitative and quantitative methods. Chapter 2 introduces the sample source, chapters 3 and 4 describe the qualitative phase (methods and results) and chapters 5 and 6 describe the quantitative phase (methods and results). Chapter 7 presents a discussion of the results.

1.1.2 The epidemiology of coronary heart disease

Coronary heart disease (CHD) is one of the leading causes of death worldwide: it accounted for over six million deaths globally in 1990 (Murray & Lopez, 1997). In the United Kingdom, CHD is the commonest cause of death, accounting for over 125,000 deaths in 2000 (Petersen, Peto & Rayner, 2004), 41,000 of which were before the age of 75 years (Department of Health, 2000). CHD affects both sexes resulting in the deaths of one in four men and one in six women (Petersen, Peto & Rayner, 2004).

CHD is also a significant cause of morbidity in the United Kingdom (UK) with approximately 300,000 individuals experiencing a heart attack each year, and 1.5 million people living with angina (Petersen, Peto & Rayner, 2004). The average incidence of heart attack per year across the UK for individuals aged 30-69 is in the region of 600 per 100,000 for men, and 200 per 100, 000 for women (Lampe et al., 2000). Data from the General Household Survey indicates that 10% of adults report a long-standing cardiovascular condition (Office for National Statistics, 2000), and in 1998 CHD accounted for 19% of all reported longstanding illness in the UK (Petersen, Peto & Rayner, 2004).

An important feature of CHD in the UK is its unequal distribution amongst the population. Death rates are significantly higher in manual occupational classes than in professional classes (Townsend & Davidson, 1992; Whitehead, 1992; Acheson et al., 1999). For example, mortality rates from CHD amongst males in manual unskilled occupations are

58% higher than males in professional or managerial occupations, and more than double for females (Office for National Statistics, 1997). Even within particular occupational groups or settings, there has been found to be an occupational gradient in death rates from CHD. The Whitehall study, for example, found that CHD was strongly associated with lower grades of civil servants, even after accounting for smoking levels (Marmot et al., 1984). Ethnic differences in mortality have also been observed. People living in the UK whose ethnic origin is South Asian (from India, Bangladesh, Pakistan and Sri Lanka) have mortality rates 46% greater in men and 51% greater in women than the national average (Wild & Mckeigue, 1997).

1.1.3 Economic burden of CHD

In addition to the high mortality and morbidity levels associated with CHD, there are some important economic costs to the UK economy. It has been estimated that CHD represents a total burden of over £10 billion a year (British Heart Foundation, 1998). The cost of direct care in 1996 for CHD was £1,630 million, which makes CHD the single most costly disease to the National Health Service (NHS) for which an economic evaluation has been undertaken (British Heart Foundation, 1998). The remaining cost to the economy due to CHD is a combination of the number of days lost to industry due to death or illness and the subsequent informal care provided by people of working age.

1.1.4 The aetiology of CHD

CHD has a complicated and multi-factorial aetiology (Tunstall-Pedoe et al., 1997). The largest prospective epidemiological cohort study of CHD is the Framingham Heart Study (Dawber et al., 1951), which was followed by the Framingham offspring study (Kannel et al., 1979). The Framingham Study demonstrated that major independent risk factors for CHD are cigarette smoking, elevated blood pressure, cholesterol levels (elevated levels of low density lipoprotein; and low levels of high density lipoprotein), diabetes mellitus and increasing age (Grundy et al., 1999). This research also suggests that the residual risk attributable to increasing age, without the involvement of any other risk factors, indicates

the presence of currently unexplained influences that may collectively be as great as the risk associated with smoking, and may be attributable to genetic factors. Furthermore, it has been estimated that as much as 50% of the risk of developing CHD remains unexplained (Nora, 1983; Braunwald, 1997), much of which may be accounted for by family history.

That heart disease runs in families is not a new revelation; nearly a hundred years ago, for example, William Osler highlighted that angina (then a relatively rare condition) frequently occurred in members of the same family:

> "The disease may occur in three generations, ...a father and four children, ... father and son, ...brothers, brother and sister" (Osler, 1910, page 879)

Family history (or a familial aggregation) of CHD is an important predictor for premature disease, particularly if CHD occurs in the affected relative before the age of 50 years (Eaton et al., 1996). Some conventional risk factors for CHD are known to have an element of genetic predisposition, particularly for the following risk factors, lipids (Dammerman, 1995), blood pressure (Doris, 2002), and blood clotting mechanisms (Weiss et al., 1996). However, the overall effect of family history of CHD could be explained by shared social and environmental factors, especially considering that CHD has such a complex aetiology, and that many risk factors have some degree of genetic mediation. A study of the Danish adoption register estimated that the death of a biological parent before the age of 50 years, resulted in a relative risk for the individual of 5 (for cardiovascular and cerebrovascular causes), whereas, the death of an adopted parent before the same age, equated to a relative risk of 3 for the same causes (Sorenson et al., 1988). Similarly a study of Swedish twins estimated that if one twin died prematurely of CHD, the relative risk amongst identical twins was 8 and 15 (for males and females respectively), compared to a relative risk of 4 and 3 for non-identical twins (males and females respectively) (Marenberg et al., 1994). These twin and adoption studies provide

compelling evidence that family history of CHD is important, but do not explain how significant family history of CHD is in the general population.

There is an extensive literature about family history of CHD. In a recent review of the evidence on family-centred approaches to understanding CHD, Kardia et al. highlighted some important considerations about this topic; definition of family history, how family history explains a significant fraction of CHD prevalence, and how family history predicts future CHD (Kardia et al., 2003). The definition of what constitutes a family history is problematic. Studies have been inconsistent about whether this includes just first-degree relatives (parents and siblings), second or even third degree relatives. The age limit that defines a strong family history has also been inconsistent, with some studies using 55 years, and others 60 years as a cut-off level. Case-control studies highlight that people who have had a coronary event (for example, a heart attack) are significantly more likely to have a first-degree relative with CHD than a control group (for example, Roncaglioini et al.; 1992, Bertuzzi et al., 2003). Kardia et al, however, point out that these studies do not indicate the prevalence of family history of CHD. They (Kardia et al., 2003), cite a study of Utah families, which found that in a general population 14% of families had a family history of CHD, but this accounted for 72% of premature CHD (in men aged under 55 years, and women aged under 65 years), and accounted for 48% of all CHD (Williams et al., 2001). This is really quite astounding, and as the authors suggest:

> "a relatively small subgroup of families in a population may carry the vast majority of the population's burden of disease. These are the families and individuals who need to be identified and strongly encouraged to engage in primary prevention measures as well as early detection of disease" (Kardia et al., 2003 page 144)

Much of the mechanisms involved with family history of CHD may be explained in the future by genetic research. A review of the literature about genetic aspects of acute coronary syndromes, highlight that, depending on which study is examined, CHD in a first-degree relative increases an individual's relative risk by 2-7 fold, compared with individuals where there is no family history of CHD. Furthermore, in this review of the

literature, a maternal history of CHD under the age of 50 years increases the relative risk by up to 10-fold (Samani & Singh, 2001). Genetic predisposition to CHD is rapidly emerging to account for much of the unknown risk, particularly in relation to a heart attack (Marian, 1998). Molecular research, studying pairs of affected siblings, has identified loci on several chromosomes, which are strongly associated with CHD (Broeckel et al., 2002, Halgadottir et al., 2004, Wang et al., 2004).

A brief overview of developments in genetic research is discussed later, but there is currently some scepticism as to whether these developments will be able to be utilised in clinical practice in the near future (Epstein, 2004), especially in a complex condition such as CHD, where a simple family history would provide so much information already (Kardia et al., 2003). Family history of CHD, particularly in a sibling under the age of 50 years, is therefore an extremely important indicator of an individual's risk of developing the disease and also a possible mechanism for identifying a relatively small number of people at significant risk of developing the disease.

1.1.5 Identifying people at high risk of developing CHD

Clearly, CHD is a major health problem in the UK, and a key part of health policy. Over recent years, the government has established a number of measures to tackle the problem of CHD, particularly the National Service Framework (NSF) for CHD. These policies were first mooted in the white paper "Saving Lives – Our Healthier Nation" (Department of Health, 1999), which laid down a plan to address the main causes of death in the UK, including CHD. This put forward the target of reducing death rates from CHD and stroke amongst the under 75-year age group by two fifths before the year 2010. The NSF for CHD contains a detailed strategy for improving the prevention and treatment of CHD through the implementation of a number of "standards". One of its key provisions is to prevent CHD in high-risk individuals. Standard 4 of the NSF states, that:

> "*General practitioners and primary health care teams should* identify all people at significant risk of cardiovascular disease but who have not developed symptoms and offer them appropriate advice and treatment to reduce their risks" (Department of Health, 2000 page 4)

This aspect of the NSF probably will have a significant impact on primary care services. It has been estimated that in the average primary care practice of 10,000 patients, about 900 pieces of data will need to be recorded (for example blood pressure and body mass index) and that over 2000 disease control measures (for example, advice on diet or smoking cessation) will be required. This could place a considerable burden on primary care if it were to be implemented (Hippisley-Cox & Pringle, 2001). The Healthcare Commission suggests that progress has been made towards achieving the national standards of the NSF particularly in establishing CHD registers, improved recording of risk factors and higher rates of advice and treatment given (Healthcare Commission, 2005). However, there is no mention about the importance of family history in identifying people at risk of developing CHD. This represents a paradox, for while there is a clear commitment to primary prevention of CHD, particularly for groups of individuals who are readily identifiable, the report seemingly overlooks one of the most readily accessible tools for assessing an individual's risk: family history. Indeed, most risk assessments tools, including the one utilised for the NSF are based on the CHD risk calculator from the Framingham study. This tool utilises many of the known risk factors for CHD, for example gender, smoking, hypertension and high cholesterol, but has some important omissions (Jones et al., 2001), particularly the importance of family history, (Grundy, 1999, Grundy et al., 2001).

Family history could be utilised as a tool for public health measures and preventative medicine. Although genetic technologies offer much potential, especially genetic susceptibility testing and biomarkers for environmental exposures, until these technologies are readily available family history is likely to be an important component of identifying people at risk of a variety of complex, multi-factorial conditions such as CHD (Yoon et al., 2002). Studies have previously shown that offspring of women affected with

premature CHD often have more than one modifiable risk factor (Allen & Blumenthal, 1998). Similarly, asymptomatic siblings of people who have experienced a premature heart attack also frequently have modifiable risk factors (Yanek, et al., 1997, Hengstenberg et al., 2001). Furthermore, it has been suggested that siblings of people who have had a heart attack are overlooked in primary prevention strategies (Hengstenberg et al., 2001). In the Hengstenberg study, many asymptomatic siblings of heart attack patients also had at least one other modifiable risk factor, and those who were being treated were often treated sub-optimally (Hengstenberg et al., 2001).

Some interventional studies have utilised family units as the interventional group for a trial, but many of these have been based around couples rather than siblings or offspring. Kardia makes the point that the reason for this is because spouses are known to share many risk factors, including smoking, body mass index, high blood pressure and high cholesterol (Kardia et al., 2003). Additionally, it is thought that lifestyle interventions based around couples rather than individuals are more likely to succeed because of mutual support (Pyke et al., 1997). Some of these studies have demonstrated impressive results, notably a study in Norway that achieved significant reductions amongst men of total cholesterol and body mass index (Knutsen et al., 1991), and the British Family Heart Study. The British Family Heart Study was a national trial involving some 12,000 couples across the UK. The intervention group received nurse-led lifestyle intervention and achieved a 12% reduction in CHD risk status after 1 year (Wood et al., 1994), although the reduction in smoking was very modest at 4%.

Interventional studies with siblings of heart attack patients have been conducted, most notably in the USA, where a mediated study of cholesterol management found that specially trained nurses were effective in treating high cholesterol in siblings, even though many siblings did not achieve the target reduction in cholesterol (Becker et al., 1998). More recently, a study among African Americans that was designed to eliminate barriers to individuals' access to health care, found participants were twice as likely to reduce their

cholesterol and achieved an overall lower CHD risk if major barriers were reduced (Becker et al., 2005).

In summary, family history is a major risk factor for heart disease and an important predictor of future events. It is likely that some of the excessive familial risk will be explained by genetics in the future, and there is currently considerable research being undertaken in this field which may offer the possibility of genetic susceptibility testing at some stage in the future (Epstein, 2004). The precise mechanism of the familial effect on CHD risk is unclear, although it is universally accepted to increase an individual's risk of developing the disease. Family history has been proposed as a route for identifying individuals who are probably at significant risk of developing CHD, which is in-line with the NSF, although family history is not included in the discussion of the NSF about identifying people at risk. Given that family history of CHD is so important, and that primary care staff are required to identify individuals at high risk of developing the condition, it seems surprising that family history is not utilised more systematically as a means of identifying people. However, it is also necessary to understand how people who may be at high risk of developing CHD by virtue of their family history perceive their personal risk, since this may influence how acceptable services are to them, or their health-related behaviour.

1.2 Lay Epidemiology of CHD

This section includes the lay epidemiology of CHD and causal attributions, or explanations that people give for events and illnesses. These are from two different theoretical perspectives; lay epidemiological research has tended to be conducted from a sociological perspective and causal attributions have tended to be investigated from a psychological or health promotion perspective. Lay epidemiology is based on the study of populations and explores cultural beliefs about causes of illness, examines social patterns and sometimes involves qualitative methods. Research on causal attributions, on the other hand, is usually based on the study of individuals, and is mainly based on quantitative research aimed at quantifying the beliefs of individuals.

1.2.1 The general population

The sociological literature regarding health beliefs and the lay interpretation of disease and illness starts with questions about how health is defined and how health beliefs are socially and culturally patterned (Calnan, 1987; Nettleton, 1995; Scambler, 2005). This is probably a useful platform from which to tackle the subject, because, as Scambler (1997) points out, individuals do not conceptualise health and illness in isolation. Nettleton (1995) goes further to state that:

> "...beliefs about health are rooted in wider socio-cultural contexts and that lifestyles are inseparable from the socio-economic structures in which individuals live out their lives." (Nettleton, 1995, page 37)

There has been a steady growth in the literature about the public perception (or lay epidemiology) of the causes of CHD. Some of the most influential work in this area was undertaken by Davison, against a background of a largely medically orientated and, often "victim-blaming", health promotion ethos of the time which placed little emphasis on family history. Davison's interviews with lay people suggested that they had a good understanding of the role that inheritance plays in the development of CHD, which was not matched by health promotion approaches at the time (Davison et al., 1989). Davison went on to describe the sophisticated frameworks lay people use to explain the causes of illness and the prediction of future illness, frameworks which were centred on ideas of coronary candidacy (Davison et al., 1991). Typically, the "coronary candidate" is identified as an overweight person who smokes, worries a great deal and as someone who is quite stressed. However, there is a very important addition to this framework of candidacy, that of the exceptions to the rule, which Davison describes as "violations of the candidacy system" (Davison et al., 1991, page 14). Examples in lay accounts of coronary candidacy violations include "the last person you'd expect" to have heart problems, or of "Uncle Norman's", the latter seemingly indestructible characters who are reported to have survived to "ripe old age" (Davison, 1989, page 46) despite an unhealthy lifestyle.

Lay ideas of fate and fatalism appear to play an important role in candidacy violations. Davison et al. (1992), make the point that much of the published health promotion research about the perceived causes of illness appears to assume a dichotomy between lifestyle and fate in lay understanding of CHD causation, beliefs about fate as a cause of heart problems on one end of the spectrum and unhealthy lifestyles on the other end. However, as Davison et al. (1992) argue, people are in reality more sophisticated than this simple dichotomy suggests, and are often aware of the behavioural health risks associated with an action and the physiological changes that accompany it, even if they, for example, continue to smoke. They continue by arguing, that although fatalism is often judged as being a flawed perspective, it perhaps should be viewed as an:

> "important element in a sophisticated and balanced belief system"
> (Davison et al., 1992, page 677).

This is developed further by Backett et al. who highlight that lay evaluation of health often involves a subtle balance and weighing up of ("trading-off") positive and negative aspects of health-related behaviour (Backett et al, 1994, (Hughner & Kleine, 2004).

1.2.2 *Individuals with a family history of CHD*

Clearly then, there are many subtleties to lay conceptualisations of personal risk in the general population, but what of the beliefs amongst "high-risk" groups? As previously mentioned, first-degree relatives (siblings and off-spring) of individuals with CHD (especially a heart attack) are at particularly high risk of developing the disease themselves, and are therefore an important potential "target" for the primary prevention strategy implicit in target 4 of the NSF for CHD (Department of Health, 2000). While there has been some research into the physical risk factors for CHD amongst the offspring of women with premature CHD (Allen et al., 1998), and the unaffected siblings of people with CHD (Yanek et al., 1998; Hengestenberg et al., 2001), unaffected siblings of people with CHD are often overlooked in primary prevention strategies even though they are an easily identifiable group (Hengestenberg et al 2001, Hunt et al., 2003).

Not surprisingly then, there has been relatively little research to examine the health beliefs and perceived risk of individuals whose siblings have experienced a heart attack. One exception to this conducted by Becker & Levine (1987) involved structured telephone interviews with 80 unaffected siblings of CHD patients following discharge from hospital. They selected patients who were consecutively admitted to hospital (under the age of 60 years) with a diagnosis of heart attack or unstable angina, or who were to undergo bypass surgery. The results suggested that the unaffected siblings of these patients did not perceive their risk to be elevated relative to the general population, and that their lifestyle remained unchanged following their siblings' event. While these results are interesting, having a sample with mixed diagnosis categories makes these findings difficult to interpret. A heart attack is the most likely clinical presentation of CHD to have a genetic component, but only 5% of the index cases in the Becker & Levine study had experienced a heart attack. Importantly their study involved structured telephone interviews so access to participants' perspectives in their own terms is likely to have been very limited. Moreover, this work is now nearly 20 years out of date and precedes many developments in genetic research, which have received considerable media coverage. Therefore, although this is an important study it is of limited value now because of the methodology used, the inclusion of angina and bypass surgery as diagnosis groups, and the fact that there have been such enormous advances in genetic technology in the intervening periods, which are likely to have influenced perceptions of familial risk factors.

There is some evidence to suggest that a family history of CHD may be underreported. Kee et al. (1993) interviewed a sub-set of the World Health Organisation MONICA cohort, enquiring of them details about first-degree relatives, and a group of individuals randomly selected from the general population. Family history of CHD was underestimated by about 30% in both GP and hospital records. Similarly Watt et al. (2000) found that only 23% of men and 34% of women, who had at least one parent who had died from CHD acknowledged that they had any family "weakness" attributable to heart disease. Moreover, they noted that individuals from manual occupations (who are more likely to

suffer from CHD) were less likely to perceive themselves at increased risk due to a family history of CHD – even when a parent had died prematurely from the disease.

Hunt et al., (2000) explored the relationship between the perception of a family history of CHD and health-related behaviour. They found that 40% of respondents reported a family history of CHD, and that people who did perceive themselves as having a family history of CHD were more likely to report the role of a "family illness or weakness" as being important in the development of a disease. An important finding, particularly from a primary prevention perspective, is that the people who identified themselves as being vulnerable to CHD, were often least likely to adhere to health promotion measures. Furthermore, they had the most fatalistic views and were significantly more likely to smoke. Fatalism is an important dimension in how CHD is viewed, and It has been suggested that the framework of the "coronary candidate" (described by Davison et al. 1991) is central to the lay beliefs about disease causality, especially in relation to fatalism (Hunt & Emslie, 2001). Hunt & Emslie go on to highlight the differences and similarities between lay and professional viewpoints of disease causality, particularly when there are family members affected by CHD. For example, lay people did not automatically perceive a family history of CHD even if there were multiple members of the family affected, but health professionals would aim to quantify a family history of CHD accurately (Hunt & Emslie, 2001). This, they conclude, could lead to misunderstandings between patients and professionals.

Lay beliefs about gender and heart disease have been an important area of research. Men and women readily identify inheritance as a risk factor for CHD, although men often require multiple members in the family to be affected before they acknowledge the familial nature of the problem (Hunt et al., 2000). A study by Hunt et al., (2001) examined lay concepts of what constitutes a family history of CHD and found that men had (or reported) little knowledge about family history, whereas women seemed to talk more freely about families and their experience of illness. Other research has suggested that women are

less likely to be seen at risk of CHD. Emslie et al. suggest two possible reasons for the apparent "invisibility" or lack of recognition of the CHD risks for women (Emslie et al., 2001b). Firstly, women typically experience CHD at a later stage of life, most commonly after the menopause, and therefore later than men. Consequently they are less likely than men to "fit" into the typical "coronary candidate" framework. Secondly, women are likely to have a longer illness associated with CHD and accounts of their death may be perceived as due to old age rather than CHD (Emslie et al., 2001b).

1.2.3 Summary of lay epidemiology

Understanding lay beliefs of disease causation is important if effective primary prevention strategies are to be implemented. Central to the lay epidemiology of CHD is the framework of "coronary candidate" (or the person most likely to be expected to have a heart attack), as well as violations of the candidate system (Davison et al., 1991). Fatalism is also often embraced as an explanation for CHD (Davison et al., 1992), as is some degree of "trading-off" risk factors (Backett et al., 1994). The perception of family history is interesting: many people appear unable to acknowledge family history of CHD (Watt et al., 2000), men appear less able to recognise, or acknowledge, a family history, frequently requiring multiple members of the family affected before acknowledging this (Hunt et al., 2001). Furthermore, CHD continues to be viewed as a predominantly male condition, probably because women do not "fit" into the coronary candidate framework (Emslie et al., 2001). The only study to investigate the beliefs of individuals likely to be at elevated risk of CHD (Becker & Levine, 1987) utilised telephone interviews, which do not allow for an in-depth exploration of issues. Furthermore, they interviewed the relatives of a mixed cohort of CHD patients, including people who had experienced a heart attack and those who had undergone revascularisation (for example with surgery). This will have included many with distinct forms of the condition and therefore possibly different genetic elements (Marian, 1998). Moreover the increased publicity regarding genetic technologies will have affected lay perceptions of genetic factors and the significance of family history of CHD. It would seem timely, therefore, to undertake another study with unaffected siblings of people who

have experienced a premature heart attack to explore their risk perceptions and health beliefs in relation to their (probably high) risk of developing CHD.

1.2.4 Causal attributions for CHD

Causal attributions are mainly based on psychological and health promotion research, aimed at quantifying the beliefs of individuals about the explanations that people give for events and illnesses. A recent systematic review of research on causal attributions for heart disease found that chronic stress and fate were very common explanations for heart disease, particularly among CHD patients, and that lifestyle factors (for example being overweight or having high blood pressure), were more likely to be rated as risk factors among non-patient groups (French et al., 2001). This is an important point because previous research has shown that CHD patients are more likely to modify their lifestyle if they can attribute a cause for their condition (De Valle & Norman, 1992). This phenomenon has also been found amongst spouses of people following a heart attack (Weinman et al., 2000). Family history (or heredity) as a causal attribution for CHD was identified in 78% of the 54 datasets examined in the systematic review, but was ranked highest in only one study (French et al., 2001). Very little research has investigated beliefs about causal attributions in the context of family history of CHD. For example, scrutinising the titles of references in the French et al. review reveals that siblings are mentioned in only one previous study – the Becker & Levine study of 1987. However, another study attempting to establish if participants were aware of interactions among multiple risk factors for CHD, presented vignettes of hypothetical patients including risk factor information and family history (French et al, 2000). Causal attributions, therefore, are from a different theoretical tradition to lay epidemiology and provide a quantification of an individual's beliefs about explanations for illness. Whether family history is viewed as a causal attribution for CHD among people at high risk because of their family history, should therefore be investigated, particularly considering the previously mentioned shortfalls of the Becker & Levine study.

1.3 Genetic Research

1.3.1 The Nature of Genetic Information

The discovery of the now familiar double-helix structure of deoxyribonucleic acid (Watson & Crick, 1953) was a pivotal moment in the development of human biological sciences. However it has taken the last fifty years, with relatively recent advances in molecular technologies, to more fully understand the significance of this discovery. The Human Genome Project has initiated a rapidly expanding body of literature debating the consequences of advances in molecular biology. Central to much of this debate are questions such as: what is genetic information, why is it important, and how should it be used? In the UK, a Government advisory body on the social and ethical issues involved with human genetics, the Human Genetics Commission, defines genetic information very broadly:

> *"...personal genetic information is any information about the genetic make-up of an identifiable person, whether it comes from DNA testing or from any other source (including the details of a person's family history)..."*
> (Human Genetics Commission, 2002, page 5)

This broad definition of what can be considered as personal genetic information allows the inclusion of sensitive aspects such as particular gene mutations that may increase an individual's susceptibility to a certain disease, and also non-sensitive aspects that are already in the public domain, for example, an individual's physical appearance. Secondly, the Human Genetics Commission set out a principle for the use of human genetic material, which they call "genetic solidarity and altruism":

> *"We all share the same basic human genome, although there are individual variations which distinguish us from other people. Most of our genetic characteristics will be present in others. This sharing of our genetic constitution not only gives rise to opportunities to help others but it also highlights our common interest in the fruits of medically-based genetic research"* (Human Genetics Commission, 2002, page 38)

These two definitions provide starting points from which to assess the potential consequences of human genetic research, and a code for the use of genetic information

has been proposed, which includes respect, privacy, confidentiality and fairness (Human Genetics Commission, 2002).

1.3.2 Genetic Epidemiology and CHD

Genetic epidemiology is a branch of bioscience that studies the inherited causes of diseases in populations (Morton & Chung, 1978) and genomic medicine offers the possibility of predictive medicine in the future (Khoury et al., 2003). Developments in molecular technology offer the possibility of novel mechanisms for the prevention and treatment of many diseases identifying multi-factorial conditions such as CHD (Collins, 1999). Genetic epidemiology of CHD is a rapidly advancing body of work and one of the key study methodologies have been investigations of affected sibling pairs (Iliadou & Snieder, 2004). Siblings share 50% of their genetic inheritance, hence the importance of studying pairs of siblings who are both affected by a particular disease (Keavney, 2001). Linkage analysis studies of affected sibling pairs have identified areas on several chromosomes that are associated with CHD (Broeckel et al., 2002, Halgadottir et al., 2004, Wang et al., 2004). Ultimately, these developments may lead to the possibility of some sort of genetic susceptibility testing.

The White Paper *Our Inheritance, Our Future* (Department of Health, 2003) also highlighted that primary care teams will be at the forefront of health promotion and prevention and will therefore be expected to enable the maximum utilisation of genetic technologies. Others have argued that primary care staff are simply not trained sufficiently to undertake this role (Epstein, 2004), and although this may become a role for primary care staff in time (Emery et al., 1999), there are major educational needs for primary care staff that will need to be addressed before primary health services can take on this responsibility (Watson et al, 1999, Bankhead et al., 2001). It has been suggested that there is insufficient capacity in current clinical genetic services and these have struggled with the demands placed on them (Donnai & Elles, 2001, Wilson et al, 2005).

1.3.3 Views about Predictive Testing

Predictive genetic testing is set to become an important aspect of medicine and health care in coming years. Some medical scientists have argued that genetic research will revolutionise medicine (Bell, 1998; Braunwald, 1999; Collins, 1999; Todd, 1999; Mathew, 2001). Bell argues that a "new taxonomy" of disease based on genetics will redefine many diseases (Bell, 1998). Van Ommen et al., (1999) make the point that this will enable the development of more tests, particularly diagnostic tests, than was possible before the era of the Human Genome Project. Others have voiced more sceptical views. Holtzman & Marteau (2000) for example have highlighted that the molecular basis of conditions such as sickle cell anaemia has been known for over 40 years, but no definitive treatment has as yet been developed. Epstein argues that risk assessment should combine genetic and non-genetic information for effective disease prevention in the future (Epstein, 2004).

Predictive genetic testing offers some degree of risk assessment, although there are some important uncertainties as to whether and when a condition will develop, and the value of the test is very much dependent upon the disease. Evans et al. (2001) have suggested that the usefulness of predictive genetic testing depends upon a range of factors, including the power of the predictive test, the availability of effective treatment for the condition, and the levels of mortality and morbidity associated with the disease. However, these considerations have also been applied to standard (non-genetic) screening procedures, that the test must be acceptable, demonstrate a marker before symptoms are present and have appropriate treatment available (Altman, 1991).

However, what remains unclear at this time is how individuals will cope with and react to such information, and what factors may influence an individual's conceptualisation of their risk status. Predicting an individual's likelihood of developing a disease using biological markers, for example cholesterol, has a long history, but predicting illness using DNA does not (Marteau & Croyle, 1998). A systematic review of the literature on the psychological impact of predicting an individual's risks for illness found that a positive test

result was generally associated with depression, anxiety and psychological distress. Anxiety and depression were significantly more likely after a positive test, but only in the short term (Shaw et al., 1999). However, of the 54 studies included in the review, 10 specifically involved genetic screening (for Huntington's disease), the remainder, including the 21 for cardiovascular disease (CVD), involved some sort of screening or testing for particular risk phenotypes, for example, checking blood pressure or cholesterol, or for infection - HIV testing. Since the latter conditions are not necessarily associated with particular genotypes this review is therefore of limited use within the context of understanding people's view of genetic susceptibility testing. However, it has recently been reported that there were no long-term psychological reactions to information about increased risk of developing CHD in a trial of 2,000 men in Denmark (Christensen et al., 2004).

Marteau & Senior (1997) have suggested that it is important to investigate lay perceptions of the role of genetics in disease causation as this may have important implications for individuals' feelings of control over their health, which may in turn have adverse consequences on health behaviour if there is little that can be done to alter risk status. Furthermore, Marteau and Senior indicate that it may be difficult for patients to conceptualise how a genetic test could indicate susceptibility to a particular condition. Moreover, individuals are known to hold defensive biases in relation to their health, frequently believing that they are healthier than they are, and may demonstrate a biased appraisal of threat by minimising the importance of a positive test result (Croyle et al., 1997).

Studies of the psychosocial aspects of single gene conditions, as well as chromosomal disorders, have generated some important insights. For example, there are differences between professional and lay understanding of carrier risk status in families where there is a male affected by Duchenne Muscular Dystrophy (Parsons & Atkinson, 1992). Pre-test anxiety for Huntingdon's disease has been reported, particularly for individuals

approaching the perceived age of onset of the condition (Decruyenaere et al., 1999). Similarly, a false negative result for Down's Syndrome screening has been shown to increase stress of parents and have an adverse effect on parental adjustment (Hall et al., 2000).

Of the more common conditions for which a genetic component has been identified, cancer seems to have generated the largest body of literature. Breast cancer particularly, has been the focus of much previous research about decision making in the context of genetic testing (Jacobsen et al., 1997; Watson et al., 1998; Brain et al., 2000; Meiser et al., 2000; Meiser & Halliday, 2002). Meiser et al. (2000), found that the majority of women wanted to have information that would enable them to take steps to avoid risk, and that most women felt the benefits of testing outweighed any potential risks.

A systematic review of evidence on the psychological consequences of genetic predictive testing, found no evidence to suggest genetic testing increased distress or anxiety in the pre-test period, and indeed found decreased distress in the post-test period (Broadstock et al., 2000). This does, however, seem, to contradict the study by Decruyenaere et al, where pre-test anxiety was reported. It should be noted that none of the studies included in the systematic review examined testing for cardiovascular conditions, indeed the majority had investigated predictive genetic testing for Huntington's disease, a single gene disorder. Few adverse psychological outcomes were found, but the authors acknowledge that this may be due to self-selection of individuals seeking the test (Broadstock et al. (2000). However, as no studies of predictive testing in CHD were included in this review, this too is of limited value.

One important aspect of CHD that has a monogenic component, and for which there is a genetic test available, is familial hypercholesterolaemia. Familial hypercholesterolaemia is a risk factor for CHD (Grundy et al., 1999) and a common single gene disorder affecting one in 500 of the population (Bhatnagar et al., 2000), and is clearly of interest from a

psychological perspective. Senior et al. (1999) interviewed the parents of children who had received a positive genetic screening test. They found that the parents who perceived the test as simply detecting raised cholesterol seemed to feel greater levels of control than those who perceived the test as indicating a genetic component to the condition. The latter group of parents felt that the condition was out of their control and therefore more threatening. A qualitative study of people with familial hypercholesterolaemia, who were receiving cholesterol-lowering therapies but had no diagnosis of CHD, found that participants perceived themselves to be at risk of CHD, and actively sought causal attribution for their condition (Senior et al., 2002).

Of particular interest in the context of the current study, several authors have written about the effect of genetic testing on families. Marteau & Croyle (1998) for example have pointed out that the amount of social support people have affects their ability to cope with the results of genetic tests, so adverse psychological outcomes may not be inevitable. Another important dimension to the family with a genetic disorder is that of disclosure of information. Hallowell (1999) for example found that among women with breast cancer, there is a strong feeling of responsibility to kin to establish the magnitude of their risk and how it may affect other members of their family.

1.4 Summary

The current study addresses an important health topic (CHD), which is a key area of health policy as identified in the NSF. CHD accounts for over 125,000 deaths per year (Petersen, Peto and Rayner, 2004) and represents an economic burden of over £10 billion per year (British Heart Foundation, 1988). Family history is known to be an independent risk factor for CHD (Bertuzzi et al., 2003) and having a sibling affected increases an individual's risk significantly (Samani & Singh, 2001). Rapid advances in molecular technology may offer the possibility of some sort of genetic susceptibility testing for common multi-factorial conditions such as CHD (Collins, 1999), although improved risk assessment is most likely to be also associated with non-genetic aspects (Epstein, 2004).

How individuals at high risk of CHD interpret their risk, and their views about genetic technologies and the prospect of genetic susceptibility testing, has only been partly investigated so far. There is a considerable body of literature about lay epidemiology (Davison et al., 1991, Becker & Levine, 1987, Hunt et al., 2000, Emslie et al., 2001), and about causal attributions for heart disease (French et al., 2001, French et al., 2005). The potential negative psychological consequences of predictive genetic testing have been highlighted, including depression (Salkovskis & Rimes, 1997, Shaw et al., 1999), although it has been acknowledged that changing behaviour is difficult and that providing genetic information alone may not increase an individual's motivation to modify their lifestyle (Marteau & Lerman, 2001).

Primary care teams are required to identify people at substantial risk of developing CHD, but who are as yet undiagnosed (Department of Health, 2000). A review of progress towards achieving the standards of the NSF makes no mention of family history (Healthcare Commission, 2005), though making use of family history as a mechanism for identifying a small group of people who are at very high risk of developing CHD has been proposed by several researchers (Yoon et al., 2001, Hunt et al., 2003, McCusker et al., 2004). In general, however, siblings of people who have experienced a heart attack have been neglected in terms of primary prevention (Hengstenberg et al., 2001), which represents a missed opportunity for primary prevention. Given that CHD is common, identifying people at significant risk of developing CHD is important, and genetic developments offer much potential for future prevention and management of complex diseases such as CHD (Epstein, 2004), therefore it seems an opportune time to undertake the current study.

This study will therefore investigate the health beliefs, experiences, risk perceptions and attitudes to genetic technologies amongst a group of people whose siblings have experienced a heart attack at a young age. While there has been some work investigating

the lay beliefs about the causation of CHD, this has predominantly focused on the general population and the beliefs and experiences of siblings have been under investigated (Davison et al., 1991; Hunt & Emslie, 2001). By using a previous case-control study (described in chapter 2) as a sample source, the study allows access to a group of siblings of people who have experienced a heart attack at a young age (under 50 years), and a group of people where there is no family history of CHD (the controls of the previous study). The overall aims of the study therefore are to characterise the risk perceptions and health beliefs and views about genetic susceptibility testing of a group of individuals who are probably at high risk of developing CHD by virtue of their family history, and to quantify these findings in a later postal questionnaire. The unaffected siblings of the participant cases from the previous study were approached for the preliminary qualitative phase and all participants of the previous study were approached for the quantitative questionnaire phase.

Integrating qualitative and quantitative methods is becoming increasingly popular and demonstrates a move away from the rigid epistemological paradigms of mono-method research to an understanding that the two methodological approaches can complement each other. There are however some important philosophical assumptions inherent in both methodologies that require consideration. Quantitative research is traditionally viewed as being Positivistic, or based in scientific realism, while qualitative research is traditionally viewed as being based in idealism (Smith & Heshusius, 1986), although Guba and Lincoln use the term Constructivist (Guba & Lincoln, 1989). Quantitative and qualitative research has therefore been viewed being diametrically opposed as they are from the different philosophical assumptions of Positivism and Constructivism, which has resulted in certain tensions existing, and a debate as to which research method is superior (Murphy et al., 1998). Increasingly, this dichotomy between philosophical approaches is being challenged with an argument that integrating qualitative and quantitative methods is both useful and desirable, particularly with one method being utilised as a precursor to the other (Bryman, 1988, Bryman, 2005). This argument for a sequential mixed method study

design has been further developed to emphasise that by integrating qualitative and quantitative methods, this can help to overcome some of the limitations of mono-method research (Kelle, 2005). Kelle demonstrates this point with the example of a study design of qualitative interviews, to access and identify a previously unknown phenomenon and to develop the variables for a quantitative study, followed by a quantitative study to test some of these observations in a larger study population (Kelle, 2005). This was the format followed for the current study.

Chapter 2 Sample source

2.1 PRAMIS; a brief overview

This brief chapter provides an overview of the sample source for the current study.

The sample source for this study, were participants in a previous study undertaken at Glenfield Hospital, Leicester – the PRAMIS study. This is the Platelet Reactivity and Myocardial Infarction Study, a case – control study investigating genetic regulation (platelet polymorphisms) of premature heart attacks (myocardial infarction) (Singh et al., 2001). PRAMIS involved approximately 200 participants in each group. The cases were recruited from coronary care units in the three acute hospitals in Leicester, and the control group was recruited via two GP practices in the area. The criteria for inclusion as a case in the PRAMIS study was having an acute heart attack under the age of 50 years, now being in a stable state, and at least six months following the heart attack, and currently being under the age of 55 years. The control group were identified by their GP practice and were recruited to have a similar age and sex distribution as the cases. Furthermore, the control group had no family history of heart disease in two generations, which was ascertained during the interview with the participant.

For the qualitative phase of this study, a sample of the unaffected siblings of PRAMIS participants (cases) were recruited, but in the quantitative phase, both unaffected siblings of the cases and controls from PRAMIS were recruited, with the controls becoming the comparison group in this study. Using the PRAMIS cohort to study the risk perceptions and health beliefs of individuals unaffected by heart disease limited the current study just to white participants. This is therefore an important limitation to sampling for the current study. All participants in PRAMIS were white and had parents who were both born within Europe. This was to reduce genetic heterogeneity, as resources for PRAMIS were not sufficient to recruit the size of sample necessary to detect inter-ethnic variation. However, the PRAMIS cohort was the most accessible sample population available for this study.

2.2 Characteristics of PRAMIS participants

Table 1 details the recruitment, response rates and some characteristics of the PRAMIS sample (Quoted by kind permission of Dr. Ravi Singh).

Table 2.1 Characteristics of PRAMIS participants

	Cases		Controls	
Questionnaires sent	496		500	
Questionnaires returned	393		350	
Recruited	224		200	
Total included in PRAMIS analysis *	205		200	
Mean age	46.8 (±6.1)**		47.3 (±6.0)	
Mean event age	42.3 (±5.7)		N/A	
Range of event age	23- 50 years		N/A	
Gender	Male 175 *(85.4%)*	Female 30 *(14.6%)*	Male 174 *(87%)*	Female 26 *(13%)*
Non-smoker	At MI 36 *(17.6%)* At recruitment 36 *(17.6%)*		100 *(50%)*	
Ex-smoker	At MI 33 *(16.1)* At recruitment 127 *(61.9)*		64 *(32%)*	
Smoker	At MI 136 *(66.3%)* At recruitment 42 *(20.5%)*		36 *(18%)*	

* Of the 224 cases recruited, 12 were excluded because they their cardiac history could not be subsequently validated from the medical records, and a further 7 were removed from the analysis as they were unwilling to return for one of the biological measurements.

** Years (plus or minus 6.1 years)

2.3 Summary of PRAMIS results

The main findings of PRAMIS were that despite being on anti-platelet medication (aspirin) and anti-angina medication, individuals that had experienced a heart attack still exhibit a wide variation in platelet function, similar to the controls, who were not on any medication. The main parameters that influence this variation are platelet volume, age, glycoprotein IIbIIIa receptor density polymorphism, gender and the novel glycoprotein Ia/IIa receptor polymorphism, which influences the expression of this receptor, but not the risk of heart attack (Singh et al., 2001).

Chapter 3 The methods for a qualitative study of unaffected siblings of people who have experienced a heart attack

3.1 Introduction

The aim of this study was twofold. Firstly, to characterise the health beliefs and risk perceptions of individuals currently unaffected by heart disease, whose siblings had experienced a heart attack at a young age, and secondly, to examine beliefs regarding the potential of modern genetic technology, and how this may affect their future health and healthcare. The methods used in the qualitative phase are described here, while the quantitative phase, which involved a comparison group where there is no family history of heart disease, is described in chapter 5.

3.2 Overview of qualitative research

Qualitative research is concerned with issues that are not easily reducible to numbers (Murphy et al., 1998) and has now become well established within health services research (Murphy et al. 1998, Malterud, 2001 a). Once viewed as the antithesis of quantitative research, particularly randomised controlled trials (Mays & Pope, 1996), there is now considerable evidence that qualitative methods have an important contribution to make to health services research and that findings of such studies can make important contributions to understanding patients' perspectives and therefore to the design of services that address their needs (Murphy et al. 1998; Malterud, 2001). Therefore qualitative research is seen as having an important complementary role to quantitative research (Malterud, 2001), though qualitative research is also frequently used in its own right to address particular questions that are not suitable for quantitative approaches. In health services research, qualitative research is particularly useful as a means of investigating issues about which little is already known, as a pre-curser to other research methods, or to help interpret the findings of quantitative work (Murphy et al., 1998). Therefore, the qualitative phase of this study was very important as a building block for the later quantitative work.

3.2.1 Grounded theory

There are several distinctive theoretical and methodological backgrounds to qualitative research, including ethnography, phenomenology and grounded theory. The qualitative phase of this study was undertaken within a grounded theory framework. Grounded theory is a collection of approaches to qualitative research first described by Glasser and Strauss (Glasser & Strauss, 1967, Strauss & Corbin, 1998) from their research on the institutional care of the terminally ill. The overarching aim of this approach is to generate theories and abstract concepts that are "grounded" in the emerging data (Pidgeon, 1998). In its purest form grounded theory is undertaken with very little or no prior knowledge about the subject before data collection is commenced (Strauss & Corbin, 1998; Pidgeon, 1998), though in practice this position of naivety can be difficult to achieve.

Charmaz (2000) has pointed to some important criticisms of grounded theory, particularly its' many permutations, which have been a source of a good deal of confusion. This proliferation of methods of uncertain origin has rendered grounded theory as a largely meaningless description of methodology because it is unclear which version of the methodology a researcher has used.

3.2.2 Constant comparison method

A more precise methodology, which still adheres to some of the principles of grounded theory, is offered by the constant comparison method (Pidgeon, 1998; Pidgeon & Henwood, 1998). This is a set of methods for analysing qualitative data following an iterative process where analysis starts with open coding, working systematically through the body of the data, gradually generating more codes and subsequently moving to higher-level and more abstract themes to describe and represent the data (Pidgeon, 1998). As with much qualitative research, analysis commences at a very early stage, so that preliminary findings can guide an interview prompt list, and proceeds beyond the completion of data collection. Chamberlain (1999) has argued that this involves:

> "...systematic comparisons for similarities and differences at all levels; between data codes within and between cases, between incidents, between contexts, and between categories as they are developed" (Chamberlain, 1999, page 197)

The constant comparison method is therefore a structured and systematic approach to analysing qualitative data that has proved to be a practical means of implementing and applying some of the principles for grounded theory (Pidgeon & Henwood, 1998). The constant comparison method informed the analysis of the qualitative data for this phase of the study.

3.2.3 Rigour in qualitative research

Ensuring academic rigour and quality within research is fundamentally important. Recent years have seen an expansion in the attention given to ensuring the quality of qualitative research and the publication of various guidelines for ensuring rigour has gathered pace (Mays & Pope, 1995; Seale & Silverman, 1997; Chapple & Rogers, 1998; Barbour, 2001; Malterud, 2001 b). Indeed, a framework for assessing the quality of qualitative research has recently been published by the Government Chief Social Researcher's Office (Spencer et al. 2004). This is likely to become influential in the assessment of qualitative research. Some of the key features of these guidelines include:

- The defensibility of the approach used. This includes the clarity of research questions, methods that are "fit for purpose" and a detailed sampling profile.
- The rigour of conduct, including careful recording of data, and approaches to analysis that allow in-depth interrogation of data.
- The relationship of the researcher to the researched, including reflexivity (an awareness of the impact the researcher can have on the study.
- The credibility of claims, demonstrating clear links between the data and conclusions.
- The broader impact and contribution of the study.

The authors of, and commentators on, these guidelines argue that having a systematic approach to research design, data collection, interpretation and communication will improve rigour (Mays & Pope, 1995). But this is true of all research and a systematic approach does not in itself guarantee the rigour of research (Barbour, 2001). For example, there is no single correct formula for conducting qualitative research and much depends upon on how well the methods "fit" with the research question and the context in which the research is being undertaken. However, the guidelines do provide important measures that, if appropriately applied, may help to enhance the rigour and quality of qualitative research (Mays & Pope, 2000) or as criteria for assessing qualitative research (Murphy et al., 1998). The measures utilised in this study to enhance rigour included the checking of transcribing with tape recording of interviews, the checking of transcript coding assignment by supervisors experienced in qualitative research and the writing of memos.

3.2.4 Sampling considerations in qualitative research

Sampling is a key aspect of much health research and there are important differences in sampling between qualitative and quantitative research. These differences are best explained in relation to the different aims of qualitative and quantitative research. Quantitative research aims to produce generalisable results and therefore utilises probability-based techniques such as random sampling. Qualitative research on the other hand, aims to generate theories and findings that can be applied or transferred to particular contexts rather than make any statistical inferences, so random sampling in a qualitative project is usually inappropriate (Thompson, 1999). For example, a qualitative project may aim to describe the in-depth range of views of patients in a particular health setting. By contrast, a quantitative project might aim to estimate the numbers of patients who hold particular views. In qualitative research, therefore participants are selected because they have certain characteristics:

> "The purpose of [sampling in qualitative research] is not to establish a random or representative sample drawn from a population but rather to identify specific groups of people who either possess characteristics or live in circumstances

> relevant to the social phenomenon being studied. Informants are identified because they will enable exploration of a particular aspect of behaviour relevant to the research" (Mays & Pope, 1996)

Murphy et al. (1998) highlight that there are four broad categories of sampling techniques utilised in qualitative research: probability, opportunistic, non-random sampling for representativeness and theoretical sampling. This is not however an exhaustive list and there are some very practical constraints and limitations to sampling in qualitative research. In this study, the sample source (PRAMIS participants) was one such constraint. How this influenced the sampling will be discussed later.

3.3 Protocol and ethical considerations

A study protocol was developed during Autumn 2000 (appendix 1) in collaboration with supervisors from the University of Leicester *Department of Epidemiology and Public Health,* and *Division of Cardiology.* This was submitted with an application to the Leicestershire Research Ethics Committee in December 2000. Ethics project reference number 6685 (Leicestershire Health Authority reference number 6154) was granted approval in January 2001 (letter of approval, appendix 2). The Research and Development of University Hospitals of Leicester also approved trust indemnity for the study (appendix 3).

3.4 Sample source

The sample source for this study, were participants in a previous study undertaken at Glenfield Hospital – the PRAMIS study. This is the Platelet Reactivity and Myocardial Infarction Study, a case–control study investigating genetic regulation (platelet polymorphisms) of premature heart attacks (myocardial infarction) (Singh et al., 2001). The cases in this study had experienced a heart attack before the age of 50 years and were under the age of 55 years at the time of recruitment to PRAMIS. In the introductory chapter, evidence was presented that clearly demonstrates that family history of heart disease is an highly important risk factor, particularly for those people whose relatives

experience a heart attack under the age of 50 years. The unaffected siblings of PRAMIS cases are therefore an ideal group as the sample source to investigate the health beliefs and risk perceptions of people who are currently unaffected with heart disease, but who are probably at a high risk of developing CHD themselves.

3.5 Recruitment process for this study

PRAMIS participants were approached by a senior research fellow on the PRAMIS study, to inform them of this study, which was due to start, and would be recruiting the siblings of participants from the PRAMIS cohort. He then briefly detailed what participation in this study would involve (one interview with a sibling of the PRAMIS participant who was unaffected by heart disease[1], and a later postal questionnaire to a larger group). If PRAMIS cases expressed an interest in this study, the research fellow requested individuals to discuss it with their siblings. Some time after this, he re-contacted the PRAMIS cases by telephone, to ask if they had had a chance to discuss the study with their siblings, and requested permission for the researcher to contact their siblings directly. Therefore, PRAMIS cases provided their siblings' contact details to the research fellow on the PRAMIS. This occurred after they had spoken to their sibling about the study and hence had the specific verbal permission of the unaffected sibling of the PRAMIS case to do so.

Once verbal permission to contact siblings had been obtained, potential participants were posted a letter of introduction by the researcher (appendix 4) and an information sheet about the study (appendix 5), both of which were printed on University Hospitals of Leicester headed paper. A reply slip (appendix 6) and a stamped, addressed return envelope were also included. When unaffected siblings returned a completed reply-slip with explicit permission for them to be contacted directly by telephone, the researcher then independently contacted them to discuss the nature of the study and to answer any

[1] For the purpose of this study, an "unaffected sibling" was a sibling of a PRAMIS participant who had never had a diagnosis of having suffered a heart attack or any other heart problems.

questions or concerns that they had regarding this study. The following figure (3.1) illustrates the recruitment process:

Figure 3.1 Recruitment process for participants (unaffected siblings) in this study

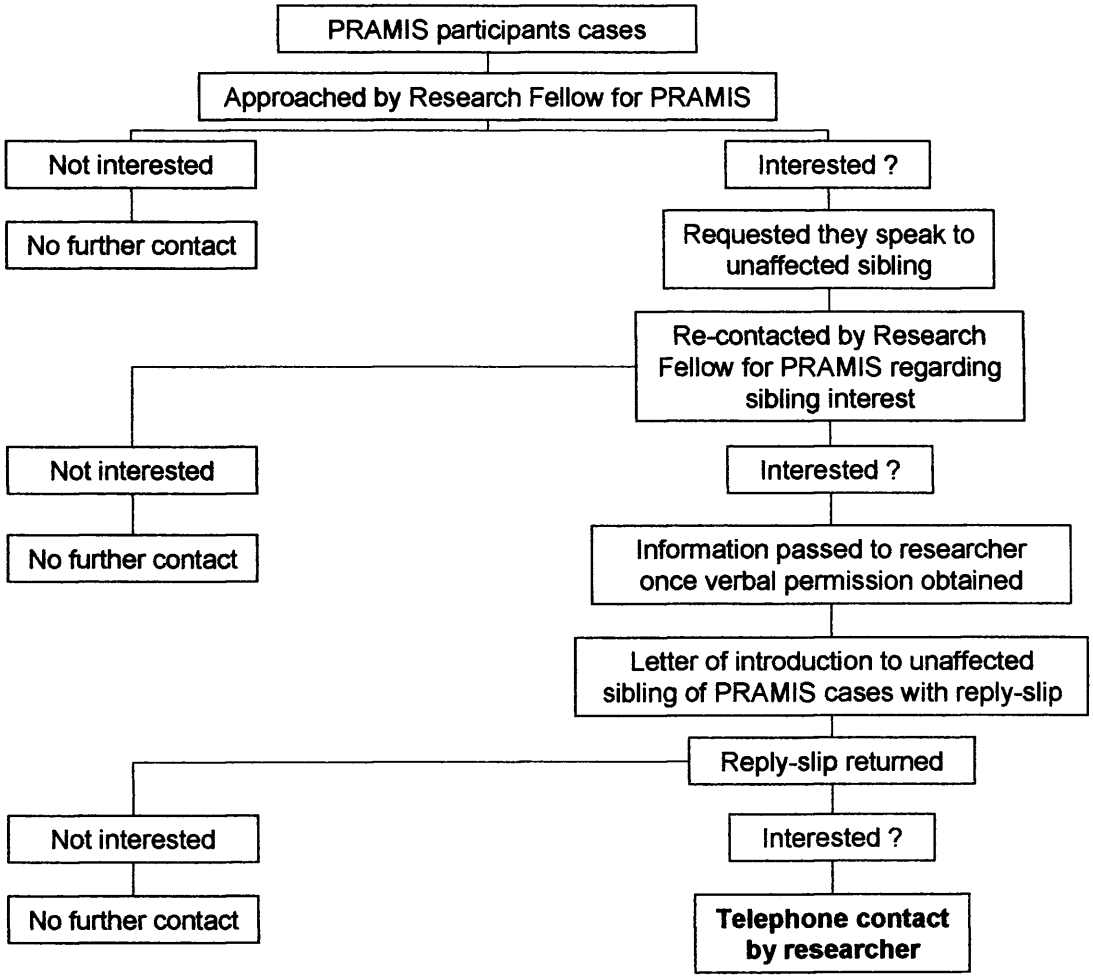


Figure 3.1 shows a diagrammatic representation of the recruitment process of the unaffected siblings of the PRAMIS cases to this study.

3.6 Influences on sampling for this study

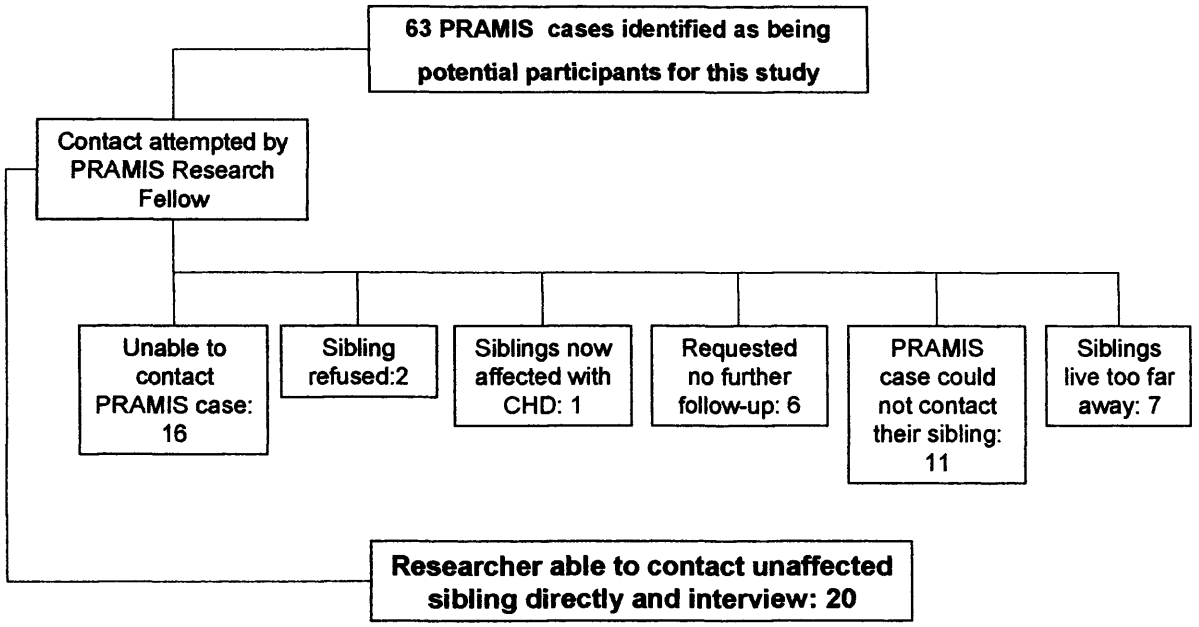
Certain features of the design of the PRAMIS study restricted the degree of control that could be exercised over sampling for the current study. For example, the nature of the PRAMIS database and the limited resources available for undertaking the interviews necessary for the qualitative phase of this study.

Participants in PRAMIS were recruited from the Coronary Care Unit of Glenfield Hospital (Leicester), a tertiary centre for cardiology, and therefore accessed by patients from all across the East Midlands, including Leicestershire, Nottinghamshire, Derbyshire, Lincolnshire and Northamptonshire. To participate in PRAMIS patients were required to visit Glenfield Hospital on at least one occasion. However, participants in the current study were the unaffected siblings of PRAMIS cases and had to be interviewed in their own home. Because of the limited funding available for the current study, only those PRAMIS cases whose siblings lived close to Leicester were selected for inclusion in this study. (The Clinical Research Fellow for PRAMIS who had developed the PRAMIS database included a detailed family structure for each PRAMIS participant, and was therefore able to facilitate the identification of potential participants within reasonable travelling distance).

After a pool of potential participants had been identified, limited quota sampling was used as far as possible in an attempt to ensure that a demographically diverse sample was obtained. As ethnic minority patients were excluded from PRAMIS, no individuals from ethnic minorities were available for participation in this study. The characteristics of age and gender were recorded in the PRAMIS database for the siblings of PRAMIS participants, therefore these characteristics could be used for sampling, and previous literature has suggested that these factors are important considerations regarding health beliefs. The PRAMIS database did not have information on the socio-economic status of siblings, therefore it was not possible to sample on this characteristic.

Sixty-three PRAMIS participants were identified as being potentially suitable for inclusion in this study and were approached by the research fellow on PRAMIS, as detailed in the section on recruitment. Sixteen PRAMIS cases could not be contacted at this stage and therefore no further attempt was made to include their siblings. Six of the PRAMIS cases requested no further follow-up, either because there was illness in the family, (amongst siblings or parents), or because they were not in regular contact with their sibling. Two of the unaffected siblings refused to participate in the study, and one sibling of a PRAMIS case who had previously been unaffected with heart disease had subsequently also experienced a heart attack since his/her brother/sister had participated in PRAMIS. Eleven PRAMIS cases could not contact their sibling, and seven PRAMIS cases had siblings who lived too far away (for example, Pembrokeshire or Worcestershire). Finally, 20 unaffected siblings of the PRAMIS cases were recruited as participants for this study. Figure 3.2 details this sampling process and illustrates some of the problems encountered by attempting to retrospectively approach unaffected siblings of individuals of participants from a previous study.

Figure 3.2 Sampling process



The sample size within qualitative research is difficult to estimate in advance as it is closely bound up with the aim of achieving theoretical saturation, which in turn is influenced by variability in the data. (For the purposes of the ethics application, the sample size was estimated to be 20 participants.) Theoretical saturation refers to the time when no new concepts are found in the data, that is the sub-categories and themes have expanded to a state where they are *saturated* and no new information or concepts are found (Strauss & Corbin, 1998). Interviewing can continue beyond this point to test the themes that have been developed and to confirm that theoretical saturation had been achieved. Further interviews can also help with testing the analysis and enables interesting ideas to be explored with subsequent participants, therefore developing the interview prompt guide and ensuring it is sensitive and appropriate.

3.7 Data collection

Each participant was contacted by telephone, by the researcher, to arrange a suitable time for an interview visit. All interviews took place between June 2001 and January 2002 in the participants' own home. Each interview visit began by the researcher explaining the full procedure of the study and the steps taken to guarantee confidentiality. This discussion enabled clarification of participant involvement and an assurance that they (the participant) could stop the interview at any time they wished, or decline to answer any questions that they felt uncomfortable answering. Permission was sought to audiotape the interviews, and participants signed a consent form (appendix 7). Demographic information about the participants were collected using a pre-interview participant characteristics form (appendix 8). This recorded details about the participant notably, their age, sex, marital status, occupation and smoking history. Semi-structured interviews were conducted with participants utilising the funnel approach, where general issues are discussed first before moving onto more specific issues (Britten, 1995). A prompt guide was developed to guide the interview (see below)

3.7.1 Prompt guide

A written prompt guide was utilised to ensure that similar topics were discussed in all interviews, but also allowed the flexibility to explore any new issues that arose in a particular interview, or which were of particular concern to the participant. The initial prompt guide was developed before the first interview, and was revised three further times up to the fifteenth interview in order to incorporate important topics that emerged from the earlier interviews (appendix 9). This is an important part of qualitative research, whereby significant ideas from early interviews can be tested for corroboration in subsequent interviews. The prompt guide was kept visible during all interviews, without being referred to obviously.

3.7.2 Data recording

All interviews were audio taped with the participants' permission using a desktop cassette recorder with an in-built microphone. All tapes were identified with the participant identification (ID) and the date of the interview. Participants were all offered a copy of the recording, as required by the ethics committee, but all declined. GPs of all participants were informed in writing of their patients' participation in the qualitative phase of the study (appendix 10).

3.7.3 Field journal

A field journal was kept to ensure that notes about contextual details and immediate reflections were recorded. This proved to be a particularly beneficial exercise and allowed the immediate thoughts to be documented before formal transcription of the interviews took place. This was also referred to during analysis and subsequent writing of results, and provided prompts for gathering thoughts and reflection on the interviews.

3.7.4 Transcribing

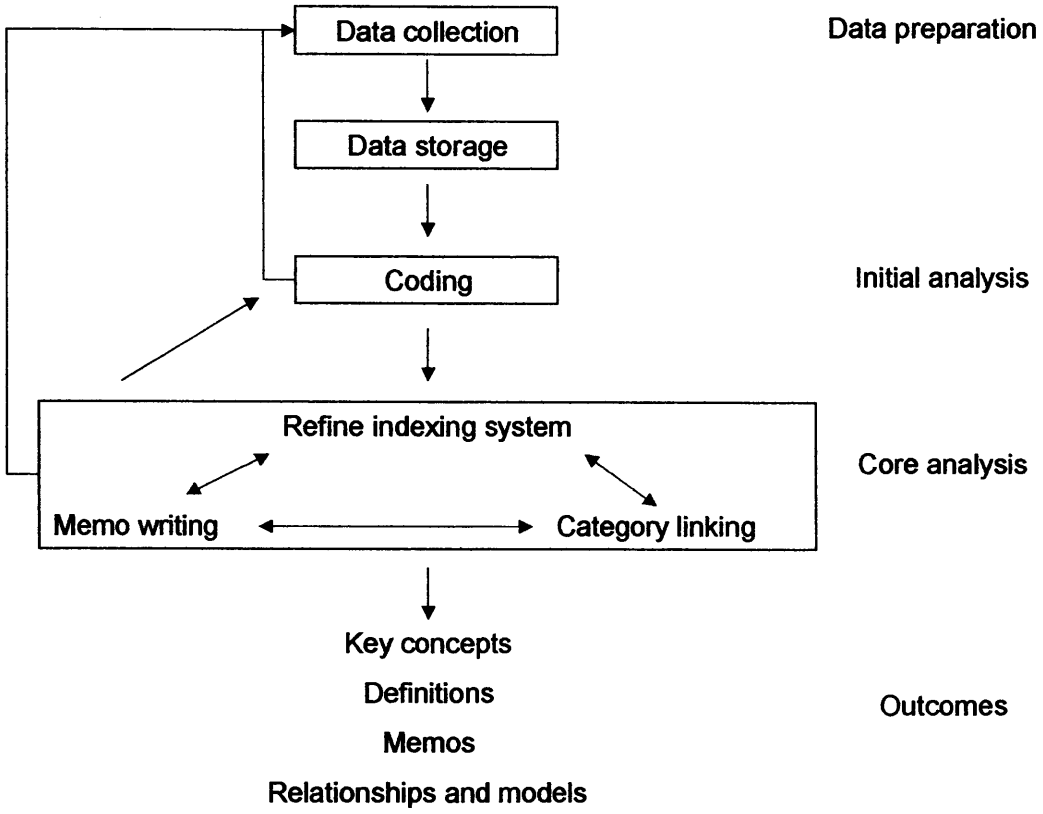
All audio-recordings of the interview were transcribed verbatim. The researcher transcribed early interviews, but some later interviews were transcribed by a professional transcribing service after a small grant had been obtained from a local charity. However, to ensure that these externally produced transcriptions provided an accurate record of the interviews, the researcher subsequently checked them all against the audio recordings.

3.8 Data analysis

Analysis was undertaken using the constant comparison method (Pidgeon & Henwood, 1998), with the analysis of the transcripts commencing at an early stage during the fieldwork and continuing throughout the course of the study. Analysis followed an iterative process with continual cycling between and within transcripts, and the codes and coding framework (Maykut and Moorhouse, 1994). Figure 3.3, (adopted from Pidgeon and

Henwood, 1998) illustrates the steps and procedures involved in using the constant comparison method.

Figure 3.3 Steps involved in data analysis



Initial analysis aimed to code, line by line, each relevant unit of meaning within each transcript. After the first four interviews had been transcribed, open line-by-line coding of transcripts was undertaken in a team approach with the researcher and experienced supervisors working together to generate the open codes. This involved systematic and very close examination of each transcript, with each new idea or unit of meaning identified being assigned an open code, usually using a word or phrase that was close to the participants' own words. From the few first interviews, forty-three separate codes were identified. By the time seven transcripts were coded (interviews A5, A3, A4, A2, A1, A9 and A7), this had developed into seventy-seven separate codes (appendix 11). As units of meaning were allocated open codes within the transcripts, comparison with coding in

previous transcripts was carried out to ensure that coding was appropriate and new codes were only created where a unit of meaning could not be captured by an existing code.

Individual codes were then assigned to higher- level categories, and sub-categories (appendix 12), before being finally developed into themes. Initially this was undertaken as a team approach with supervisors but subsequently the researcher working alone on this. Several *working* thematic frameworks were developed to contain and represent the data. Comparison continued within and across transcripts while developing the themes and thematic framework. The themes were continually checked and adapted to ensure that there was an appropriate *fit* between the data and the themes representing them. Transcripts were systematically checked to ensure that all relevant data could be assigned to the thematic framework and that this provided a logical and plausible representation of the data. Each of these main themes contained several sub-categories. After further analysis, the themes and sub-categories were reorganised, and in some instances combined.

A computer package, QSR NUD.IST (Non-numerical Unstructured Data Indexing, Searching and Theorising) version 4.0 was used to index the data using the thematic framework. This is a widely used computer software package for assisting in the analysis of qualitative data (Murphy et al., 1998). Indexing was initially undertaken with close supervision by academic supervisors and later independently. Once data had been indexed within NUD.IST a selection of reports were checked by a supervisor for accuracy of coding as a quality assurance step. Once the coding was complete, this held a comprehensive set of coded data indexed around the thematic framework. An example of a section of a report from NUD.IST can be seen in appendix 13.

3.8.1 Memo writing

Memos are an important part of the analysis of qualitative data, and have been described as the step between coding and final analysis (Charmaz, 2000). As the analysis was in

progress for the current study, memos were written in a separate book to the field journal and kept alongside it. These contained detailed notes on specific codes or themes and worked as an aide-memoir to interpreting data. In this study memos were also used to record higher-level theoretical concepts about the analysis or interview transcripts. Writing memos therefore assisted with the interpretation of the data and improved the quality of the analysis. An example of a memo is in appendix 14.

3.9 Summary

Qualitative research enables a systematic study of issues that are not reducible to numbers and has an important role to play in health services research. The study of "what?" and "why?", rather than "how many?" which is frequently the focus of quantitative research, is particularly useful in an area of study about which very little is known.

For the preliminary phase of this study, qualitative interviews were undertaken to characterise the health beliefs, experiences, risk perceptions and views on the potential of modern genetic technology, of a group of people who were not themselves affected by CHD but whose siblings had experienced a heart attack at a young age. The sample source for this study was a previous case-control study undertaken to identify genetic aspects of blood clotting mechanisms in premature heart attack. This is a limiting factor in the current study, because all the PRAMIS participants were white. Furthermore, PRAMIS cases were all recruited into a tertiary centre of cardiology, so their homes were distributed widely over the East Midlands (they had travelled to Glenfield Hospital, Leicester for participation in PRAMIS on one occasion). Since there were no resources available for travelling, and unaffected siblings of the PRAMIS cases needed to be interviewed in their own homes, this further reduced the sample population from which to recruit participants to this study. Utilising a previous study as a sample source therefore proved problematic, and perhaps a more robust method would have been to utilise an alternative sample either newly identified, or identified specifically for this purpose from an existing or previous study.

Chapter 4 The results of a qualitative study of unaffected siblings of people who have experienced a heart attack

4.1 Introduction

This chapter describes the results of the qualitative phase of the study, which was introduced in the previous chapter, and will outline the characteristics of participants, the qualitative results, and finally a summary. This qualitative phase was undertaken to explore the risk perceptions and health beliefs of individuals whose siblings had experienced a heart attack at a young age and had participated in a previous study (PRAMIS). A thematic framework was developed consisting of four main themes to organise, index and interpret the data. The thematic framework, analysis and data excerpts are presented in this chapter.

4.2 Characteristics of participants

Of the 63 PRAMIS cases identified as potential participants for this study, 20 unaffected siblings were interviewed. Ten males and ten females participated, with ages ranging from 30-58 years (30-57 years for males, and 41-58 years for females). The mean age was 46.9 years (43.8 years for males, and 50 years for females). Fifteen of the participants had manual occupations, with the remaining five participants either having clerical occupations or owning their own businesses and there was one business analyst. The majority (twelve) were former smokers, four had never smoked and four were current smokers (one of them male). Fifteen of the participants were married or co-habiting, three were divorced or separated, one was single and one was a widow.

Of the PRAMIS participants (i.e. the affected siblings of the participants in this study through whom we recruited our participants) the majority (15) were male. This is unsurprising since PRAMIS cases were people who had experienced a heart attack at a young age (under 50 years) and the majority (85%) were male (see chapter 2). The affected siblings (who had participated in PRAMIS) were aged from 36-59 years with a mean age of 49.2 years (49.5 years for males, and 48.2 for females). The age at which

they had their heart attack ranged from 30-49 years, with a mean age of 43.2 years (43 years for males, and 44.8 years for females). Therefore, participants in this study were young-middle aged people, with siblings who had experienced a heart attack at a young age. A full table of characteristics of participants is in appendix 15.

4.3 Results

4.3.1 Thematic framework

As described in chapter 2, the thematic framework for interpreting the qualitative results underwent substantial development during analysis. It was finally established that a framework consisting of four key themes and numerous sub-themes provided a satisfactory means of organising and representing the data. These themes are: the experiences of premature heart attack in a sibling, explanations for siblings' heart attack, inheritance and genetics and participants' experience with healthcare professionals (see appendix 16 for the full thematic framework). All of the qualitative data can be explained and interpreted by these themes, which form the structure for the rest of this chapter. Theoretical saturation was achieved in this study approximately by the tenth interview.

4.4 The experience of premature heart attack in a sibling

4.4.1 Surprise / shock

Overwhelmingly, participants described their initial reaction to their siblings' heart attack as one of shock and surprise. This frequently involved questioning why their sibling had experienced a heart attack at such a young age. Additionally some participants gave accounts that reflected on beliefs about coronary candidacy (Davison et al., 1991), and how their sibling's heart attack had violated such beliefs:

> "When I got the 'phone call to say it was a possible heart, and I just said well "why"?", because she's still young. I mean a heart attack to me is when somebody older has a heart attack" (A1, 49-year old female)[1]

[1] For a glossary of symbols used in quotes, see appendix 17.

> "The fact that he was so young I think that is what made it such a shock" (A12, 46-year old female)

> "Everybody thought if anybody were gonna get it, you start from the eldest, right, and work down like. But I mean he was relatively young, well all of us are, still are, and was then. So, yeah, I mean you just don't have heart attacks at our age, we've never heard of it anyway ... it shocked us and it made us think about things for a little while." (A13, 54-year old female)

> "It were just absolute, unbelievable shock, he were the last person on Earth you would have thought, you know, when people have heart attacks you always imagine big, fat people, red faced, beer drinkers, smokers, eating burgers, and, but [affected siblings name] were never like that, always had good food, as in, he weren't a heavy drinker, he's always had lots of exercise...he were just the last person on Earth to have had a heart attack" (A14, 55-year old female)

> "...obviously the younger you are the less you expect it. The older your get it's not quite so shocking, it's more usual I suppose, to expect someone to have a heart attack when they are older" (A20, 47-year old male)

Above and beyond the initial feeling of shock and surprise at the untimely nature of their siblings' health problems and how it ran counter to beliefs about coronary candidacy, the last quote highlights the introspection that many participants reported following their siblings' illness. This introspection may have implications for the health-related behaviour of individuals whose siblings have had a heart attack, with them either being more fatalistic (and therefore less likely to lead a "healthy" lifestyle), or being overly anxious about their own health.

Unsurprisingly participants often expressed a great deal of concern for their sibling and his/her well being, particularly following such a life transforming and unexpected event. This appeared especially true for the small number of people who were in their sibling's presence when the heart attack occurred, or for those who visited them in the period immediately afterwards:

> "I tell you to be honest, you know it was the most worrying thing I think has ever happened to me sitting in that er ambulance ... watching it was awful, and couldn't really do nothing, 'cause I wouldn't know how to" (A11, 47-year old male)

> "I was devastated, yes I was really devastated, he is just the last person that I would have thought would ...I didn't know if I was going to lose him or not ... it really upset me to see him with all the monitors, drips and wires, I couldn't believe how it did upset me" (A4, 41-year old female)

Participants appear to be voicing a sense of personal suffering as a result of their siblings' heart attack. This could be interpreted as concern for their sibling, or other members of their family, however it could also reflect their feelings of personal vulnerability.

4.4.2 Vulnerability

One of the key areas that was tactfully explored during the interviews for this study, was the extent to which participants felt vulnerable to developing heart disease because their siblings had experienced a heart attack. The number of people affected by CHD in a family appeared to be quite a significant factor for a number of participants. Participants' feelings of vulnerability seemed to be heightened if they came from families where there were multiple members affected with heart disease:

> "I mean, in your quieter moments or when you go to sleep at night these things go through your head and you do think, yes, I must admit I have felt sick, you know with my father having a heart attack, it really didn't bother me too much in terms of thinking you know, it could happen to me. Since [siblings name] has had hers yes, I have thought, perhaps I am vulnerable to the same sort thing." (A5, 43-year old male)

This is an interesting point and may indicate that if only one person in a family is affected, this does not necessarily lead people to presume that they are at increased risk themselves as the heart attack can possibly be discounted as an unfortunate but one-off event. Similar findings have been described in earlier research where, males particularly, often did not recognise or acknowledge the significance of a family history of CHD until more than one first-degree relative was affected (Hunt et al., 2001). Perhaps this means that people require more evidence than one affected sibling to consider that there may be a potential genetic influence on their personal risk. Other studies have similarly shown that there appears to be confusion regarding what actually constitutes a family history of heart disease, with adult offspring (especially males) being unlikely to interpret a parental

history of CHD as a family history, and therefore questioning the significance this poses to their own risk for heart disease (Watt et al., 2000).

The feeling of "closeness" to a family member has been described as a factor that may lead to feelings of susceptibility to the same illnesses or diseases that other family member has experienced (Davison et al., 1989, Richards, 1993). These feelings were explored during this study. When asked directly if they felt close to their sibling, and if as a consequence of that, they felt at increased risk of experiencing the same illnesses as their sibling (thereby giving an indication of feeling of vulnerability), most participants in this study appeared to reject the idea that physical resemblance or emotional closeness to a sibling who had experienced a premature heart attack would somehow increase the risk of them having a heart attack themselves. They often did so by indicating that though they resembled their sibling physically they were quite different in other aspects:

> "No, no I've never thought of it like that, no. You may well look the same, like, but we got little bits of different characteristics" (A11, 47-year old male)

4.4.3 Detachment

Therefore, there appeared to be a strong sense amongst some participants that their siblings' heart attack had little or no consequence for their own risk status, because they were completely different people and would therefore be unlikely to experience the same health problems. Additionally, some people offered a rationale as to why they would not be at increased risk of heart disease, often citing their much healthier lifestyle as a compensatory factor with the implication that this would protect them from any genetic predisposition. Similar findings have been highlighted before, where high levels of ambivalence, particularly amongst men in manual occupations have been observed (Hunt et al., 2001) though these have not been previously described as "detachment". Hunt (2001), for example described these phenomena as ambivalence to family history. It is perhaps significant in this context that the majority of participants in this study were from

manual occupations, and are therefore, according to the observations of Hunt et al., more likely to appear ambivalent to their own personal risk. The following quotes, however, illustrate the point of detachment well:

> "I've never really thought ooh, our [siblings name] had a heart attack, I'd better be careful, you know. I've never really equated it like that all." (A20, 47-year old male)

> "Whatever happened to him was totally different to me because I'm a different person" (A3, Male 53, brother had MI at 30)

There is an interesting paradox here: the shock and surprise participants expressed when talking about their feelings about their siblings' heart attack as discussed above, and then this pronounced sense of detachment that some of the same participants appeared to express when the discussion turned to their own individual risk. Perhaps this is some sort of defensive mechanism whereby individuals are so shocked by their siblings' heart attack that they develop compensatory mechanisms to counter-balance any fears they may have.

4.4.4 Complacency / indifference

When discussion moved away from participants' personal experience of their siblings' heart attack to more abstract beliefs about heart disease, many talked about heart disease in a rather complacent manner, particularly in comparison to other major diseases such as cancer. There appeared to be a belief that a heart attack was a condition that a person either recovered from completely or was almost instantly fatal, whereas, cancer was often discussed in terms of a condition that people invariably died from in a lingering and undignified manner. A general impression was therefore given that a heart attack was somehow a favoured mode of death because of the rapid nature of the condition. This belief about heart disease leading to a "quick death" has been described before in qualitative research. Having a heart attack was seen as a "good way to go", by participants interviewed by Emslie et al (2001 a) and similar feeling were expressed by participants in this study:

"I have seen a friend die from cancer, not pleasant. At least hopefully with [affected siblings name], if and when it happens and it possibly won't be long, it could be painful but then it will be quick. Cancer you linger. I think that is the frightening thing about it". (A12, 46-year old female)

"Now if there's something wrong it's a cancer of something, you know what I mean? And I know there's a lot of cancers can be cured and all that but that's the biggest worry in my eyes anyway, not a worry but it's in the back of my mind like, not a heart attack or anything like, cancer... heart attack don't worry me, no". (A13, 54-year old male)

"Yeah, yeah, cause, I think with heart, it's sort of like you know, you have a heart attack, you possibly live, you may die. But with cancer you don't die it's sort of like more it's such a long wait, as it were to actually die. I don't like wasting diseases" (A7, 32-year old male)

" I mean if you have one good heart attack you've gone, that's it, the end of it" (A14, 55-year old female)

Participants in the current study, therefore, appeared to be far more frightened of cancer than heart disease, presumably relating to beliefs about rapid nature of death as previously described, and to the belief that there was less suffering involved in a death following a heart attack.

The gendered nature of beliefs about heart disease was also apparent in participants' accounts. Heart attacks appeared to be viewed as a predominantly male condition, and some of the terminology used by participants was quite illustrative of these ideas in this study. Heart attacks in a male relative were frequently discussed as being a "massive heart attack" whereas, often referred to as merely a "heart attack" when speaking of female relatives. Previous evidence suggests that lay belief systems tend to view males as being more likely coronary candidates (Emslie et al, 2001b) and therefore heart disease is viewed as a condition that largely affects males.

4.4.5 Beliefs about moderating perceived risk

Despite the evident complacency about heart disease (at least relative to cancer), many participants talked about factors that might motivate them to moderate their personal risk for heart disease, including both positive and negative role modelling of health-related behaviour, the social acceptance of certain activities (particularly smoking) and other ideas about moderating risk.

> "Mum and dad are quite heavy smokers and er, you know it's always, I'm probably the one in the family what don't smoke ...she's [participant's mother] always been a heavy smoker, since I were a kid, all I'd seen were me mam with a fag in her mouth, just all through growing up. That's probably why I'm not smoking because I didn't like it and just couldn't be doing with all the smoke around me ... I'm pretty much anti-smoking to be honest." (A9, 30-year old male)

A range of factors, not just the experience of having one sibling who had experienced a heart attack, therefore influenced many participants' beliefs about the risk of heart disease. Therefore individuals appeared to conceptualise their personal risk alongside other factors.

4.4.6 Barriers to moderating perceived risk

While many participants acknowledged that they would like to modify their personal risk of heart disease, by altering their lifestyle, some indicated that there were often practical barriers preventing them from moderating their risk. Examples such as a lack of time, or being pre-occupied with other activities, or perceived cost of a healthy diet were frequently mentioned:

> "Now and again you do think I really must go, like all these jobs, I really must go [for medical checks], ...then something else comes along, and you don't bother... something more important always crops up." (A5, 43-year old male)

> "Well, you know er, I would actually like to quit the cigarettes, at the minute, but I can't. I don't actually eat a lot of fatty foods. I go to the gym at least once a week so as far as I know that'll keep me, I know I'm wrecking everything by smoking, but that's how it goes." (A7, 32-year old male)

> "Well, not everybody can afford proper food if you like, fruit, and fresh meat, fresh vegetables is expensive. Not everybody can afford it." (A12, 46-year old female)

Difficulties associated with individuals maintaining a healthy lifestyle, and confusion about health promotion information have been identified in previous research as barriers (Meillier et al., 1996, Frankel et al., 1991, Davision et al., 1992). Other work that used quantitative methods to assess whether individuals perceived themselves to have a family history of heart disease, highlighted that people who perceive themselves to have a family history of heart disease and view themselves at risk, are less likely to smoke (Hunt et al., 2000).

4.4.7 Information and support

Participants utilised a variety of sources for information about heart disease. This ranged from generally available sources of information such as newspapers and magazines to specialised sources, such as health information leaflets, which they accessed at their GP surgery. Some participants mentioned British Heart Foundation literature, and health and current affairs programmes on the television were also frequently mentioned as sources of information. Of the participants who had actively sought information, that is to say, had purposefully taken steps to obtain information, public libraries, health care professionals and the internet seemed to be the most commonly identified sources of information.

Some participants suggested that information about heart disease risk should be targeted at all families who might be at substantial risk. This demonstrates some insight into the genetic susceptibility, but was often tempered by the realisation that identifying a large proportion of the population for screening would be a large undertaking:

> "I mean, ... me and [affected siblings name] are only small, there's only two of us, but people from large families, you know, say there's ten of them, I think it would prove probably too expensive would it? ...I think it would be a good thing, even if they didn't contact all the brothers and sisters, just a few of them" (A14, 55-year old female)

However, other participants indicated that they personally would not want to be investigated to assess whether they had an elevated risk of developing heart disease (or cancer) because they felt that this might increase their anxiety levels, or that they would be worrying unnecessarily about their health.

Continuity of health-related information seemed to be a concern for many participants, with several discussing how changing trends in recommendations, particularly in relation to diet, were extremely confusing and disconcerting. Examples were mentioned of how dietary recommendations for fat intakes (types and quantities) had frequently changed over time, thereby leading to confusion and a loss of confidence in experts' advice and how this had had a detrimental effect on subsequent behaviour.

A small number of participants expressed a desire for some sort of structured psychological support mechanism or network that could be available to siblings of people who had experienced a premature heart attack. In cardiac rehabilitation, the needs of partners of people who have experienced a heart attack have been well documented (Thompson, 1989). However, participants in this study seemed to be requesting a service for other family members to access, whether this was some sort of counselling service, or specific clinical service to identify people at risk of CHD. The following quote, from one female participant, expressed well this desire for a structured support mechanism:

> "It would be nice to be able to go somewhere or pick up the 'phone and talk to somebody, especially in my position where I'm virtually on my own ... you need input from people that know, from professional people" (A17, 58-year old female)

Several participants put forward recommendations regarding the content of information and support that they felt should be made directly available to people whose siblings (or other close family members) had experienced a recent heart attack. This included suggestions for health information to be included in television adverts, in school education and in areas that are accessible to the general public (such as Citizens Advice) for people

who may not be registered with a GP, or lack access to the internet. One participant, who worked in a cardiac department of a hospital, indicated that there was considerable information about heart disease on coronary care, but then very little information in any of the other areas of the hospital which might therefore miss a large number of people, visitors for example. Some participants indicated that health-related information (health promotion literature) should include symptoms to be aware of in relation to heart disease and where and to seek assistance if such symptoms occurred.

4.4.8 Lifestyle changes following siblings' heart attack

Several participants reported that they had made changes to their lifestyle following their siblings' heart attack, including changes in their dietary or exercise habits. Some had clearly sought advice as a direct result of their siblings' heart attack, but more interestingly, some had examined their own lifestyle, and a few had even self-prescribed daily aspirin, as a preventative measure.

> "Well, I'm thinking to myself, now I'm a lot older than [brothers name] and if that happened to him, and you read about these incidents where if somebody has a heart attack they give you an aspirin don't they? And so I thought well I'll take one once a week and then it'll help me thin my blood... and obviously we don't have full fat milk now, we have skimmed milk and, you know, things like that, and everything's grilled" (A13, 54-year old male)

Other participants reported that their entire family had radically changed their lifestyle, particularly dietary intake of fats and stopping smoking as a direct consequence of heart attack in the family. Conversely, a small number of participants reported to have made little or no change to their lifestyle, often citing the feeling that their siblings' heart attack was a result of an excessively unhealthy lifestyle.

4.5 Explanations for siblings' heart attack

When questioned about the causes of their siblings' heart attack, unsurprisingly many participants were able to correctly identify the majority of well-established risk factors for heart disease, particularly smoking, a high dietary intake of fat, obesity and a lack of

that has investigated causal attributions for heart attacks which has generated similar findings (French et al., 2005). Interestingly, however, participants often required prompting to consider genetic aspects as a specific risk factor for heart disease. For example, when asked about the causes of heart attacks generally, one participant gave the following response:

> "Bad diet, lack of exercise, smoking...stress. That's all I think. I don't know anymore." (A16, 58-year old female)

Other people talked about how risk factors often added to each other to make the situation worse, and how increasing age was normally associated with heart disease.

> "I would say, probably a combination for the fact that she had always been a smoker, the fact that she was overweight and also the fact that I think for the past few years she has had quite a lot of stress" (A5, 43-year old male).

Stress as a cause of heart attacks, was frequently mentioned in participants' accounts. Accounts of siblings' (i.e. PRAMIS participants) experiencing different or unusually high levels of stress, or from different sources (personal, financial, occupational) were very prominent. This seemed especially convincing to participants as an explanation for the period leading up to their siblings' heart attack, with frequent references to marital breakdown, difficulties with work and other stressors.

> Interviewer: "What do you think caused his heart attack?"
> "Stress. I honestly do. Unadulterated stress" (A10, 34-year old male)

> Interviewer: "What do you think caused his heart attack?"
> "Well I think personally it was stress with his job"(A14, 55-year old female)

> "You often hear there's more stress going around now, and you always assume that probably heat attacks and things like that is it more to do with stress side, because they do say that as years to come, you know stress will be your killer, in other words more than anything else." (A18, 53-year old female)

Another dimension of this included comments about siblings' strategies for coping with stress. This often led to accounts about how their siblings became easily worked-up, or to

depictions of them as being pre-occupied with worries and concerns. For example, when referring to his brother, one participant said:

> "He's a "whittler", what I put our [siblings name] down as a whittler ... a whittler, you know, always whittling about something, I suppose you could say that's related to the stress factor." (A19, 57-year old male)

When this participant was asked to elaborate what he meant by the term "whittler", he explained that his brother was continually worried and moaning about things. Another man, also talking of his brother said:

> "He gets stressed out very easily, like I say we all get stressed out it just depends on how you handle it." (A9, 30-year old male)

Perhaps it is unsurprising in this study that stress as a cause for heart attacks was given such prominence, particularly amongst a group of people whose siblings have had a heart attack at such a young age. In the literature, much reference is made to stress, particularly chronic stress; a recent systematic review of the literature for causal attributions for heart disease found that stress was one of the most commonly cited causal attributions for heart attacks (French et al., 2001). It is also important to note that male siblings were particularly likely to be seen as very stressed.

References to quite specific day-to-day habits of their siblings were also prominent as an explanation for their heart attack. For example, it was not just that siblings had a poor diet, but that the timing of eating may have been a causal factor in their health difficulties. Some participants, for example, commented that their siblings frequently ate take-away food late at night, which they felt had significantly contributed to their heart attacks.

> "I think sometimes we're all living hand-to-mouth sort of thing, we could just say slow down for half an hour, have a bit of time to eat us meals, or eat sensible meals ... I don't believe in eating late at night ...overindulge, that's the biggest problem " (A3, 53-year old male)

> "I mean eating late at night, sitting on your heart is not good for you, you know, if you can eat during the day ... not after eight o'clock" (A9, 30-year old male)

Participants also frequently referred to fate as an explanation for their sibling's heart attack. This included the belief that their sibling had somehow been "chosen" to have a heart attack, or it was simply the luck of the draw, or for some random reason that was inevitable or unavoidable. Some of the following quotations illustrate these beliefs:

> "It's a one-off... I can't recall any part of the family having heart problems, ... sorry [siblings name] you're just a one-off and that's unfortunate, but you're the one that was chosen, and that's the way that I've probably thought." (A3, 53-year old male)

> "If it happens, it happens" (A7, 32-year old male)

> "I am very much a person that, what will be will be" (A16 58-year old female)

As with stress, fate as an explanation for heart attacks, is a well documented in the literature as an aspect of the lay belief systems, especially the work on coronary candidacy (Davison et al., 1989, Hunt et al., 2000). On the other hand, not all participants expressed these views. One male rejected the notion of fate quite forcefully:

> "I mean fate's another word for luck in my eyes. No, you make your own luck, don't you? So you make yourself, you can make your own health." (A13, 54-year old male)

4.5.1 Life event

There appeared to be a broad consensus amongst participants that their siblings' heart attack was a major life event with considerable psychological implications and which had changed the life of the affected sibling. Depression was commonly reported as a consequence of the affected siblings' heart attack:

> "I think she got a little depressed. Erm, I think it was worse when she came out, because obviously she couldn't go back to work". (A1, 49-year old female)

> "He gets very depressed. Really, very depressed. We were talking on Sunday and he said that what he misses, his work mates, now he's at home all day he doesn't see anybody as such. The kids come home from school and he gets ever so depressed and he's very short tempered" (A4, 41-year old female)

4.6 Inheritance and genetics

4.6.1 Nature of genetics

There appeared to be a belief among some participants about the fundamental and unique nature of genetics as a risk factor for heart disease. That is, risk that operated via genetic pathways was seen as distinct from other risk factors. Genetic information had a special significance because genes were regarded as the essence of life, the basis of one's very being, and accounts centred on how genetics sets you apart from others:

> "...you are talking about the basic building blocks of life and the genetic code that builds your body.." (A5, 43-year old male)`

> "I mean it's your being your body entirely, you know, it is in the code for your body." (A5, 43-year old male)

It is not surprising therefore, that genetic problems were often portrayed, or discussed, in terms of being serious, unalterable or "nasty" conditions:

> "It seems, the science, seems more involved and more interesting and, I don't know, when somebody mentions genetics, having a genetic problem you tend to think that's a root problem with you, it could be a very serious problem." (A5, 43-year old male)

Other participants talked about certain genetic conditions, or chromosomal abnormalities, particularly Down's Syndrome, cystic fibrosis and cervical cancer. Much of this discussion appeared to be leading towards preventing conditions such as these through prenatal screening:

> "...make sure we don't breed sort of people like with um, you know, like Downs Syndrome." (A11, 47-year old male)

While this view with its eugenic overtones and clumsy expression is distasteful, it does nevertheless reflect participants' sense that conditions with a genetic aetiology, were often referred to with a sense of dread. Additionally, there appeared to be some understanding

about the nature of inheritance, and how certain illnesses were clustered in families. Referring to cervical cancer, one participant commented:

> "...in their particular family, a couple of sisters have had cancer..." (A13, 54-year old female)

Another participant that requires special consideration, within the context of this sub-theme of the nature of genetics, is case A12. This is a 46-year old woman whose daughter (who was present during the interview), has Turner's Syndrome. This led a discussion about her daughter's condition, which may illustrate the experiences of families where there are people who have chromosomal disorders. The quote below appears to portray an over-simplified view of the role of genetics in heart disease, which current literature suggests is a complicated inter-play of inheritance, environment and behaviour.

> "I think genetically we have, it's 50:50. Some of us have got the gene where you can have heart problems, some of us can't. It's like anything else I suppose. Um, I mean we have got genetic problems, [daughters name] has got Turner's Syndrome, that's chromosome disorder. I think if there is a family trait there, if you are one that's unlucky enough to inherit it, you're the one that can get the problem" (A12, 46-year old female)

Heart disease, unlike for example Huntingdon's, is not a single gene disorder and the role of genetics is not yet fully understood. The above quote therefore seems to contrast dramatically with the scientific views of genetically conferred risk in heart disease.

4.6.2 The nature of genetic testing

Exploring the nature of genetic risks with participants often led to a discussion about genetic testing. This included discussions about the nature of genetic testing, what these tests might reveal and consequences of genetic testing. There appeared to be a belief amongst some participants that genetic testing was somehow fundamentally different from other testing procedures, or great uncertainty about what a genetic test would involve:

> "...if somebody mentions some genetic test I think it seems to jump into a, in my perception it jumps into a different league than the normal, what I call the normal tests, you can go to your surgery and have." (A5, 43-year old male)

> "I don't think I thoroughly understand what a genetic test would be". (A20, 47-year old male)

Other participants expressed ideas about what a genetic test could reveal, particularly in relation to other more conventional tests. There appeared to be a belief that a genetic test was far more specific and would therefore lead to much more detailed or informative results. This is presumably related to the previously mentioned views about genetic information:

> "I would think that genetic ones [tests] would be far more appropriate to be honest. Because like you can only, they're using all these tests for ...check if you like, the things that they think which is cholesterol, and high blood pressure and thick blood, I think the genetic one [test] it would show you a hell of a lot more". (A7, 32-year old male)

The potential consequences of genetic testing were discussed by many participants, both in terms of the potentially alarming or distressing nature of the information, and its broader social and financial implications, such as limited employment opportunities or reducing availability of insurance for affected individuals. Regarding the psychological consequences of this information, there was an acknowledgement that information generated by genetic testing may lead people to be overly anxious or depressed about their future health:

> "It could be very depressing for some people to find they are predisposed to very nasty diseases, or some people won't be able to do anything about it perhaps". (A5, 43-year old male)

Others acknowledged that depending on the outcome of the result, there may be some pressure to disclose this information to other family members.

> "I think if one of your family has a particular problems and that genetic tests turns up something, it may put pressure on other members of your family to think that they should go and be tested as well ... If [siblings name] had gone for some sort

> of genetic test and found that yes she has got some sort of genes that predisposes her to heart disease, then yes I would feel even more, I would probably even feel more likely to go and have the tests. (A5, 43-year old male)

The responsibility of disclosure about inherited risk to other members of a family has been highlighted before, in the context of women undergoing genetic counselling for breast and ovarian cancer (Hallowell, 1999), as has the whole notion of kinship and families (Richards, 1993, Petersen, 1998, Richards, 1996). Participants in this study, as illustrated by the quote above, appeared to acknowledge this responsibility to their kin by claiming that they too would inform other family members of the potential need for genetic testing.

Regarding the broader social consequences of genetic testing, participants frequently required prompting to mention insurance or employment, but when they did, this was often discussed in a negative manner. In terms of insurance, there appeared to be a belief that a genetic test, if it revealed an individual to be at an elevated risk of developing a certain disease, would lead to them being unable to obtain insurance:

> "I think with insurance companies taking more note of genetic tests, ... um, we are going to end up with a situation where you won't be able to get insured for anything. Only the healthiest person with no genetic abnormalities or defects is the person who is going to get life insurance". (A5, 43-year old male)

Fears about discrimination from life insurers, based on genetic factors, have been described before (Low et al., 1998) amongst support groups of individuals with specific and well-known genetic conditions (including cystic fibrosis and Huntingdon's disease). In this context it is worth noting that the insurance industry has agreed a moratorium on the use of genetic information for life insurance purposes up to the value of £500,000 for the next five years (Mayor, 2001, Warren, 2001).

The potential impact of genetic testing on employment, and employment opportunities, were also discussed by a number of participants in a similar fashion to insurance opportunities. Some participants felt that if an individual was able to undertake the duties required of them by their employer then a genetic test should be largely irrelevant. Others,

however, stressed that a genetic test might lead an employer to consider the amount of time a person is likely to require off work due to sickness:

> "I think it would be more likely to deter companies from employing people like that ...[they] are going to spend certain amount of time off sick, so I think in that situation, um, I think most of the effects would be negative" (A5, 43-year old male)

4.6.3 Views about future susceptibility genetic testing

The views of participants' about the possibility of future genetic susceptibility testing were one of the key areas of discussion in this study. When specifically asked the hypothetical question about whether they (the participants) would be willing to undertake a test that could demonstrate they were at elevated risk of developing heart disease, a very broad range of responses were voiced. This question was posed carefully, and was preceded by an explanation that such a test was not available at the moment, but may be developed in the future. Superficially there seemed to be a full range of responses from the extremely positive:

> "I would yes. Yes. I would like to have had it when I was younger". (A11, 47-year old male)

> "I think I would. OK there is no cure as such, but forewarned is forearmed if you like". (A12, 46-year old female)

To other responses which expressed considerable resistance to genetic susceptibility testing:

> "No, because I don't want to live under that shadow, if someone would tell me I'm a high risk, I'd sooner not know, definitely". (A20, 47-year old male)

Following close examination of the transcripts and NUD.IST reports to attempt to understand the source of this variation, there was some evidence that beliefs about genetic testing were gendered. Of the three participants who were strongly resistant to genetic susceptibility testing, all were men. With the exception of the first two interviews (both with female participants), where the question was not specifically asked, all of the female participants expressed a positive view about the possibility of future genetic

testing. The three men who did not want to have any form of genetic testing, appeared concerned about how they would live their life with such information, which was clearly expressed by the following quote:

> "If somebody told me I'd got a high risk of having this, I'd say "Oh shit! – Thank you very much, now how do I live my life?" I'm 53 and now you've told me I'm 90% gonna die from heart problems..." (A3, 53-year old male)

4.6.4 Public interest in genetic research

Participants appeared interested in genetics and genetic research, with the majority speaking in positive terms about the potential impact that genetic research might have on future health:

> "I think they should do genetic research. Definitely...if there is a way forward where genetically these diseases and chromosome disorders and genetic faults, if you like, can be remedied and find a cause for them, it has got to be good. Got to be..." (A12, 46-year old female)

Other participants expressed more sceptical views about genetic research and referred to the need for controls to be placed on such research to ensure that it was conducted within an ethical framework:

> "I think we should be concerned, but were not gonna be able to stop it are we? ... I suppose we have to move forward don't we? And without this sort of thing we can't move forward [pause]. Where does one draw the line however? (A16, 58 year-old female)

No clear gender differences were found regarding participants interest in genetic research, although two participants (both males) were equivocal and indifferent in their response to this line of questioning. The only participant who was quite resistant to genetic research was a woman:

> "I don't know, I'm a little bit sceptical on these, this sort of thing, you know. I don't know, I think humph (exclamation), you're dabbling in things that you don't really know about, to be honest with you". (A1, 49-year old female)

Another dimension that some participants discussed was whether there should be an authority or agency established to ensure the guardianship of genetic information. This is interesting on two levels, firstly that genetic information is viewed as special, requiring careful handling and attention to confidentiality (see above and below in other themes), but specifically the expressed desire for guardianship of this information:

> "Who should make the decision...That's difficult. Um, cause obviously there would be social and political implications whoever made the decision...Somewhere along the line the doctors have got to be given the credit of being able to judge what's in the patient and family's best interest". (A12, 46-year old female)

It is interesting that participants are therefore echoing some of the recommendations of the Human Genetic Advisory Commission report *Inside Information* (Human Genetic Advisory Commission, 2002) and general concerns regarding the security of genetic information (Petersen, 1998). These reservations were expressed in a variety of forms. For example, while seeking some sort of guardianship for genetic information some participants were more concerned about confidentiality, questioning whether local GP practices could meet this requirement:

> "Anyone who works in one of the offices as a reception for example, they've got access to the information of their client...I mean you don't have to be a degree to be a receptionist and they haven't signed anything, so there's no confidentiality. I mean I go down, or ring up for an appointment at the Health Centre, for example, and the receptionist is asking you, "what's wrong with you?" you know, "is it important?"...Scuse me, this is between me and my doctor." (A16, 58 year-old female)

Some participants therefore, discussed beliefs about inheritance and genetics with reference to the fundamental nature of inheritance, the perception that genetic problems are often serious or unalterable conditions and many expressed an interest in current genetic research. Previous research has highlighted concerns, however, about the public understanding of the new genetics (Durant et al., 1996, Richards, 1993, Richards, 1996).

4.7 Experiences with health care professionals

An important dimension of an individuals' readiness to accept recommendations to modify their lifestyle is in their ability to approach and trust health care professionals. However, some participants in this study gave examples of their GPs either being unaware of their family history (their siblings' heart attack), or simply having little interest in following up this line of enquiry when participants had themselves taken the initiative in broaching the subject.

Some participants offered a variety of explanations for consulting their GP (or other health care professionals) in the aftermath of their siblings' heart attack, however, others felt that either there was no need to go and see a GP, or there was always some reason to delay the consultation. The following quotations illustrate the range of these accounts, from the barriers (always busy), to the direct link between siblings' heart attack and consultation:

> "You know I ring to go for some sort of tests or cholesterol test and things like that but I have to put my hands up and say I have not done it ...I mean I do have good intentions of doing that, ... but it's always one of things that gets put to the back of your mind so many other things you have to do in a busy lifestyle. Yes, it would take a lot to lead me up there." (A5, 43-year old male)

> "I mean, since this business with [affected siblings name] I mean I go to the well man clinic now." (A13, 54-year old male)

The experiences of participants with health care professionals following their siblings' heart attack was an important aspect of this study and probably has significant implications for primary care in the future, especially in the context of identifying individuals who are probably at elevated risk of CHD. There appeared to be a group of people who were inhibited from seeking help, because of previous experiences with their GP where they felt their concerns had been brushed aside or trivialised, and some participants appeared particularly reluctant to visit their GP to discuss the implications of

their siblings' heart attack. One female participant, for example, when asked if she had considered visiting her GP following her brothers' heart attack, said:

> "... I don't feel that you can go to the doctor and say well, you know, I would like you to check this out ... you're wasting their time ... I feel that they feel I'm wasting their time, so I won't go" (A2, 51-year old female)

Participants who did visit their GP following their siblings' heart attack, either for reassurance or investigation, reported a variety of experiences. One 32-year old male, whose sister had experienced a heart attack at 36 years of age, and whose father had died following a heart attack and triple coronary artery by-pass graft (and who had specifically been advised to seek medical advice by hospital staff when his sister was admitted), commented on his visit to his GP as follows:

> "... my doctor, to be truthful, that I had at the time, didn't seem to be all that interested ... it was almost like he didn't give a toss, and I was asking him something totally pointless" (A7, 32-year old male)

His, unfortunately, was not a unique experience. Another 34-year old male, whose brother had had a heart attack at 36-years old, had visited the practice nurse at the GP surgery for a cholesterol check following his brothers' heart attack and clearly felt that the nurse was not taking him seriously, or was trivialising his situation:

> "[The nurse asked] "What are you having this done for?" They make you feel ... [like] a bit of an idiot really...[you ask yourself] "What have I had this done for, I'm alright, I'll be alright, so why bother?" (A10, 34-year old male)

There is some evidence from previous studies to suggest that GPs and primary care staff lack the knowledge and skills to effectively manage patients' concerns about their family history, even though they may consider genetics as an increasingly important part of primary care (Watson et al., 1999). The experiences of some participants in this study suggest there may be some room for improvement in this aspect of service provision. Some participants, however, had more neutral or constructive experiences with health care professionals, notably on having cholesterol and blood pressure checks:

> Interviewer: "So, he was quite willing to sort of check your blood pressure and your cholesterol?"
> "Yeah, he just checked everything. I've had them checked recently and I'm fine" (A9, 30-year old male)

> "I think they must have took my blood pressure and cholesterol (A20, 47-year old male).

It is also important to note that participants' experiences of interacting with and consulting their GP prior to their siblings' heart attack had clearly impacted on their consulting behaviour in the aftermath of the heart attack. The prior experience of people with health care professionals is important to understand an individual's consulting behaviour and their expectations of the health services.

> "...the female doctor gives me an inferiority complex, and going to talk to her now I always feel as if I'm wasting her time ... [the doctors think] I've got one hour to see the patients, and you're coming to me with that, when I've got people out there that are ill, you know and that's the attitude..." (A2, 51-year old female)

> "All the doctors I've seen in the last 10 years not one of them have asked me about any illnesses in the family, other than what I have suffered from" (A3, 43-year old male)

Experiences, however, were not confined to attitudinal differences, but often encompassed structural and organisational systems within the GPs surgery particularly relating to appointment arrangements:

> "...if you've got anything wrong with you, they [GP surgery] say "well is it an emergency?" ... so you've either got to say yeah, and be a liar, or go down on the four week waiting list, see sometimes it can be as long as six weeks, if you want to see one specific doctor". (A15, 50-year old female)

4.8 Summary

This qualitative phase of the study involved 20 individuals (ten male and ten female) whose siblings had experienced a heart attack under the age of fifty years. Some of the findings from the interviews are perhaps not surprising: participants would be expected to discuss the common risk factors for heart disease and to describe sources of health

information. Other findings, on the other hand, are extremely interesting. The strong sense of detachment that some participants felt from their siblings' heart attack, and that it was therefore of little consequence to their own risk status is a particularly interesting finding given the known strong genetic components of a multi-factorial condition such as heart disease (especially heart attacks).

Participants' views about genetic technologies and the possibility of some sort of genetic susceptibility testing were also of interest. If this type of technology is to be utilised in the future, as the Government plans (Department of Health, 2003), then these technologies need to be acceptable to the service-users of tomorrow. The findings of this study would suggest that future service users might not be particularly receptive to this prospect. The gender difference suggested by this study needs to be confirmed by future quantitative work. If it is confirmed, it is of particular concern, because males are more likely to experience heart attacks at a younger age.

The apparent indifference and complacency towards heart disease, especially in relation to other diseases, for example cancer, was interesting, although has been described before. Indeed, some participants indicated their assumption that this was a commonly held belief amongst the general public. For such a major public health concern as CHD, the degree of indifference continues to be worrying.

The experience of participants with health care professionals is another interesting, and quite concerning finding of this study. As highlighted in chapter 1, according to the National Service Framework for CHD, primary care groups (now Primary Care Trusts) are responsible for identifying individuals at substantial risk of developing CHD and offering appropriate treatment and advice (Department of Health, 2000). Judging by the accounts of participants in this study, there is considerable room for progress, before PCTs are in a position to undertake this adequately. Considering the importance of family history of CHD and the finding that many of the participants in this study had actively sought help

regarding their concerns, the reported behaviour of primary care staff represents a lost opportunity.

Chapter 5 The methods of a questionnaire based comparative study of a group of unaffected siblings and a group where there is no family history of CHD

5.1 Introduction

The quantitative phase of this study was a postal questionnaire survey that investigated further some of the key findings of the earlier qualitative phase, participants' attitude to risk and belief about developments in genetic technologies. The questionnaires were posted to participants in the PRAMIS study (unaffected siblings of the cases) and the control group, who were a comparison group for this study. Questionnaires and surveys are probably one of the most utilised research methods, and health services research is no exception, however good practice suggests that qualitative development work can inform the design of questionnaires (Murphy et al., 1998). Therefore the qualitative phase of this study was used to influence and inform the development of the questionnaire. Prior to undertaking the qualitative phase of this study, it was anticipated that the follow-on postal questionnaire would focus on issues such as health-related behaviour (especially risk factors) and quality of life. However, the qualitative findings suggested that issues such as causal attributions for their siblings' heart attack, vulnerability, a certain degree of indifference to heart disease (particularly in relation to conditions such as cancer) and views about genetic research and future genetic susceptibility testing were important for siblings of people who had experienced a premature heart attack. These topics were explored further with the postal questionnaire during the second phase of this study. This therefore represents a logical progression and a means of investigating further some of the key findings from the qualitative phase.

5.2 Aim of quantitative phase of study

The aim of the quantitative phase of the study was to explore further some of the key findings of the qualitative phase amongst a larger sample size and also to compare these findings with a group of people where there is no family history of heart disease (comparison group). The sample for this phase of the study was the unaffected siblings of

the PRAMIS cases and the PRAMIS controls (with the PRAMIS controls being the comparison group). Since the sample source for this postal survey was PRAMIS, a relatively small case-control study investigating the genetic components of blood clotting mechanism in people who have experienced a heart attack under the age of 50 years, compared with a control group where there is no family history of heart disease (see chapter 2), a major consideration in the development of the questionnaire was to maximise the potential response rate. To achieve this the questionnaire was kept as short as possible, as this has been shown to be an important factor in increasing the response rates in postal surveys (McColl et al., 2001, Edwards et al., 2002). This is very important for this study as it involved siblings of participants in a previous study, who are a group that are very difficult to contact. Consequently very careful consideration had to be given to the inclusion of each item, with sufficient justification for the inclusion of each. Furthermore, questionnaire design is often a compromise; requiring a balance between including measurements that are valid and reliable but avoiding a questionnaire which is overly long thus placing an excessive burden on participants' (McColl et al., 2001). Therefore, the questionnaire that was designed for this study was developed following consideration of these two important issues.

5.2.1 Themes from qualitative phase of study

The postal questionnaire developed for this study was follow-on from the preliminary qualitative interview work undertaken, as described in chapters 3 and 4. Qualitative research is particularly useful as a means of researching an area about which very little is known, or as a precursor to quantitative research (Murphy et al., 1998). The aim was that the postal questionnaire would benefit considerably by the time invested to undertake the qualitative phase. Prior to undertaking the qualitative interviews, very little was known about how this group of people viewed their individual risk, their health beliefs, or how they viewed genetic research in this area. By undertaking the qualitative interviews, it was possible to devise and design a questionnaire that was more suited to the concerns and

priorities to this group. This was therefore the overarching aim of undertaking the qualitative phase of the study as a pre-curser to the quantitative phase.

5.2.2 Ethical considerations

The study protocol was revised (appendix 18) and an application was made to the Leicestershire Research Ethics Committee in April 2002. Ethics project reference number 7825 was granted approval to undertake this study in May 2002 (letter of approval, appendix 19). The Directorate of Research and Development from University Hospitals of Leicester also approved trust indemnity for this phase of the study (letter of approval, appendix 20).

5.2.3 Administering the questionnaire

The questionnaire was administered by post to both groups. Because of a variety of reasons (including the researcher starting a new job), there was a delay in starting posting until August 2003. From August 2003 to November 2003 all of the PRAMIS controls were sent a questionnaire and associated information. From September 2003 to November 2003 the majority of the PRAMIS cases were sent information and letters to forward to their siblings.

5.2.4 Maximising response rate

Given the relatively small sample (PRAMIS) from which participants for the current study were recruited, it was important to maximise the response rate for the questionnaire. This was particularly important for the siblings of the PRAMIS cases (participant group in this study). A recent systematic review about maximising response rates to postal questionnaires has shown that some relatively simple steps can maximise the response rates of postal questionnaires (Edwards et al., 2002). Therefore, the following were all incorporated as part of this process:

5.2.5 Letters

In accordance with requirements of the local research ethics committee, all letters were on University Hospitals of Leicester headed paper, as opposed to University of Leicester paper, and were signed by Professor Samani (Consultant Cardiologist and Principal Investigator of PRAMIS). All postage out was in brown A4 envelopes, franked for postage. Wherever possible, the letters were personalised with a handwritten name of the recipient and dated. Clearly this was not possible in the letter that the PRAMIS cases forwarded to their unaffected siblings. All return correspondence was in smaller, brown envelopes that were stamped (second class) and had printed return addresses on them.

5.2.6 Information

Information about the study was included with correspondence, as were contact details of the researcher and supervisors. In the letter to PRAMIS cases asking them to forward the sealed envelope to one of their unaffected siblings, it was recommended that they contact their sibling to inform them that the questionnaire was being forwarded.

5.2.7 Other considerations

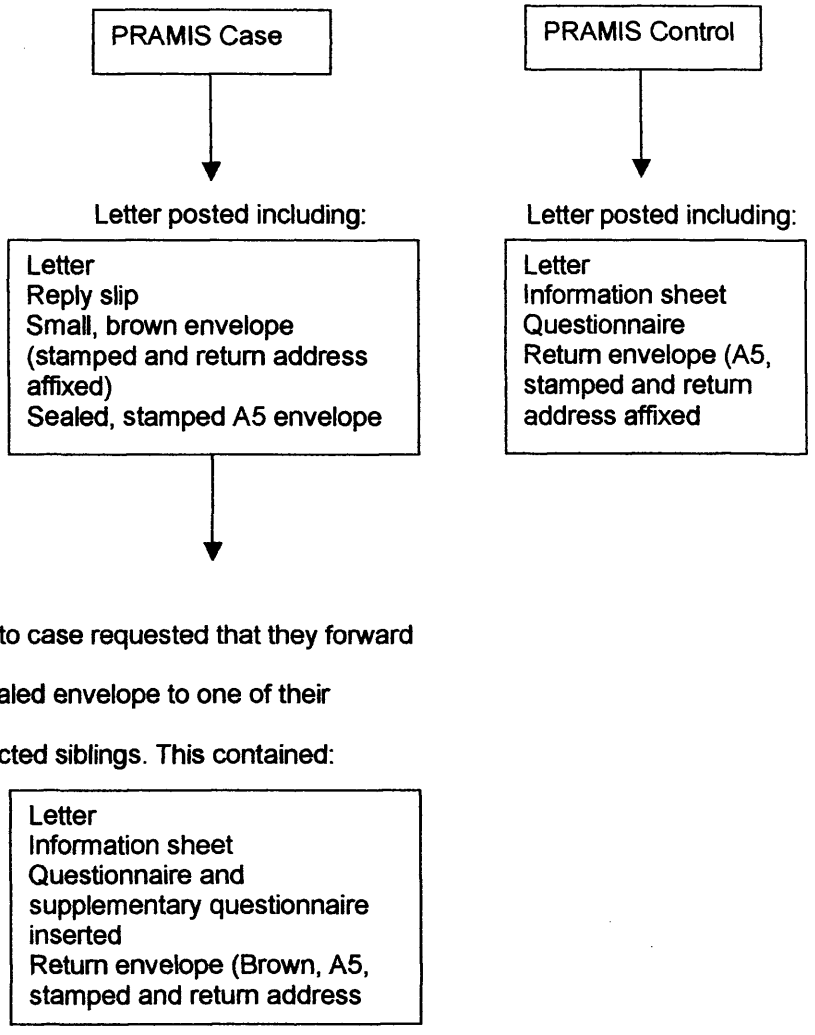
The majority of the recommendations from the systematic review for maximising questionnaire response were addressed by the above measures, with the exception of an incentive. No monetary or gift incentives were possible because of the financial constraints of the study. However, the importance and unique nature of the study was emphasised in correspondence.

5.3 Process

The process of administering the questionnaires was somewhat complicated in that the two groups had to be accessed in a slightly different manner. The PRAMIS controls (comparison group) could be contacted directly using the PRAMIS dataset. However, the unaffected siblings of the PRAMIS cases had to be contacted via the PRAMIS cases because of ethical reasons and because the contact details (or permission to contact

them) was not know from PRAMIS. This proved to be a very laborious task, which is illustrated by figure5.1.

Figure 5.1 An illustration of the process of administering the questionnaire



Copies of the above documents (letters, information sheets) are in appendices 21-25.

5.4 Questionnaire

The questionnaire utilised in this study (appendix 26) was developed over several months and piloted with a group of colleagues and friends before being finally printed. The questions were based either on previously published work, or developed following results from the qualitative phase. The questionnaire was kept as short as possible and was in

two parts: a single A3 sheet folded to make a two page booklet printed on pale green paper and a single A4 sheet printed on pale pink paper. The green booklet was posted to the entire sample (i.e. the PRAMIS cohort (controls and siblings of the cases) and the pink sheet was only posted to the siblings of the PRAMIS cases. Furthermore, many of the attitudinal statements included an option for "don't know / neither" in a further attempt to enhance response rates (Edwards et al., 2002), and efforts were made to ensure the questionnaire had an interesting layout and used a variety of response styles. The format of the questionnaire, source of questions and the rationale for utilising them are detailed below.

5.4.1 Demographic details

Questions 1 to 3 regard the demographic details of the individual, including gender, date of birth and postcode.

5.4.2 Occupation

Occupation is related to income and is frequently used as an indicator of social class and therefore question 4 uses the format of the Office for National Statistics to assess occupation (Office for National Statistics, 1997). Question 5, was based on the Registrar General's classification of occupation with six options, from professional to unskilled. Whilst this may have been superseded by more sophisticated measures of employment (which categorise occupations into far more subdivisions, and include newer forms of employment, such as call centre personnel), this question remains adequate for the purposes of this questionnaire, without being as lengthy as the more recent question formats typically used to measure social status.

5.4.3 Education

There are many options available to measure an individual's education, for example the highest qualification obtained or age at completion of full-time education. The age at which the participant completed full-time education was asked in question 6. The format of this

question is the same as that utilised by the Office for National Statistics to ascertain an individual's educational levels (Office for National Statistics, 1997). Additionally, this question requires a smaller number of responses than the alternative, which involves asking about the highest qualification achieved.

5.4.4 Health-related behaviour

Health-related behaviour is undoubtedly important as a concept and as a measurement of an individual's risk of developing CHD. However, the tools to measure health-related behaviour (which includes physical activity, smoking, drinking and drug use) are very lengthy and complex; for example the physical activity questionnaire used in the European Prospective Investigation of Cancer (EPIC) is eleven pages long. To use a tool such as this in this study would be inappropriate and might reduce the response rate significantly. There are also elaborate instruments to measure diet, such as the number of daily servings of fruit and vegetables (Thompson et al., 2000), but again incorporating these would make the final questionnaire too lengthy.

Smoking is perhaps the one modifiable risk factor for CHD that appears to be easily measurable with a questionnaire. Current smoking status is therefore used in question 7, (in a standard format utilised by the Office for National Statistics) (Office for National Statistics, 1995) as a proxy for health-related behaviour. Although this may be lacking in sensitivity, for example it does not measure frequent changes in smoking habit (quitting and restarting) (Corrigan et al., 2002), it does allow a measurement of current smoking in a simple format.

5.4.5 Perception of current health status

Validated tools are available to measure perceived current health status and quality of life, for example the Short-Form 36 (SF-36) and the Nottingham Health Profile (Bowling, 1997), and many of these have been utilised in the context of CHD (Thompson et al., 1998). However, to incorporate these within this questionnaire would have created a large

and unwieldy questionnaire, particularly as these issues appeared of secondary importance from the qualitative phase of the study. Therefore, the compromise opted for was to incorporate two questions (questions 8 and 9) that had previously been adapted from the SF-36 by Marteau (Marteau et al., 1996). These two questions will provide important information, whilst avoiding the questionnaire becoming too lengthy and cumbersome.

5.4.6 Causal attributions about heart disease

One of the key aspects that emerged from the qualitative phase of the study was the priority siblings of MI patients gave to causal attributions for heart disease. Question 10 therefore, asks participants whether a particular risk factor *increases* an individual's risk of heart disease. This question is based on the work of Weinman et al (2000), who investigated the causal attributions of heart disease in patients with first-time heart attacks and their spouses. Importantly for the present study this item has been used within the context of a family setting. Question 10 is further divided into ten sections, one for each of the following risk factors of heart disease:

Diabetes

Genetic factors

Stress or worry

Eating fatty foods

Smoking

Lack of exercise

High blood pressure

Chance or fate

Being overweight

High cholesterol

Question 11 listed the ten risk factors highlighted in question 10 and asked the participants to pick the five most important, and rank them in order, with one being the

most important. This was also based on the work of Weinman et al (2000). Question 12, an open-ended question, and asked participants to list any other things that they felt were important risk factors for heart disease.

5.4.7 Knowledge of heart disease

Clearly much health promotion information is aimed at increasing knowledge about a particular condition, and ultimately altering behaviour as a consequence of that. Question 13 aims to assess the level of knowledge of participants regarding heart disease. This is adapted from the work of Wilcox & Stefanick (1999) who investigated the knowledge of mortality, perceived risk (general and personal) and the level of control felt in relation to heart disease and various cancers. Question 13 is divided into four sub-sections. The first two (part a and b), are concerned with mortality rates for heart disease in relation to cancer, and compares men over the age of 65 with women over the age of 65. The second two (part c and d), ask about genetic risk. These three main disease groups (heart disease, stroke and cancer) were referred to during 13-16 and also in the context of genetic susceptibility testing.

5.4.8 Perception of risk, and ability to reduce the risk of heart disease

Question 14 and 15 measured the perceived personal risk of heart disease and the ability to reduce personal risk, in comparison to stroke and cancer. This was adapted from the work of Wilcox & Stefanick (1999), but required adapting for the current study, as the previous authors specified which type of cancer they were referring to (breast, colon or lung). For the current study therefore, cancer was used as a general term, as specifying a particular type would have over complicated the issue. The three disease groups of heart disease, stroke and cancer were therefore used as comparators as they represented the major causes of mortality in the UK. The perceived ability of the individual to reduce the risk of heart disease (as well as stroke and cancer) was measured by question 15, which was identical to a question used by Marteau et al., (Marteau et al., 1996) to measure ability to reduce risk. In an earlier draft of the questionnaire, another question was

included to measure the control that an individual felt over their health, which was adopted from the work of Wilcox & Stefanick (1999). However, after the questionnaire was piloted with colleagues and friends that particular question was removed as participants (to the pilot) felt that these two questions were repetitive.

5.4.9 Fear of heart disease

The qualitative interview phase of this study suggested that many people, who may be at high risk themselves of developing heart disease, tend to be less concerned about heart disease, relative to other conditions such as cancer. To measure this concern an approach was taken which required participants to compare which of the three diseases (heart disease, stroke and cancer as described above) they feared the most. This particular question was not based on any previously published items, but was developed as a means of indicating the comparative fear of these three diseases. Certainly the degree of ambivalence expressed by participants in the qualitative phase was surprising, and therefore quantifying which disease category participants in a larger study fear, was worth investigating.

5.4.10 Health as a value

The value or weight individuals give to health may give an indication of the importance participants place on health for a good quality of life and is likely to be a key factor in understanding the beliefs of participants in this study. Question 17, utilizes three sub-sections to measure health as a value. This question was based on a methodological paper that presented a scale to measure the value placed on health (Lau et al. 1986).

5.4.11 Attitudes to genetic research

One of the main areas of discussion for participants in the interview phase of this study was their attitude to genetic developments. This was particularly focused on modern genetic research aimed at identifying the genetic components of common multi-factorial conditions. As many of the participants in the qualitative phase had highlighted this topic

of some importance to them, question 18 was selected to measure participants' attitudes to genetic research and developments. The first two questions (sections a and b) have been reproduced from the Human Genetics Commission (Human Genetics Commission, 2001), while the final question (section c) has been developed for the purposes of this questionnaire. Question 19 (section a) enquires about the attitudes of respondents to genetic developments. This is a more general item, rather than the question 18 c, which is specifically focused on the individual.

5.4.12 Attitudes to potential genetic susceptibility testing

A key feature of participants' accounts during the qualitative interview phase were their views about possible moves to develop some form of genetic susceptibility testing for common multi-factorial conditions. Participants' views on this varied (during the qualitative phase); with some indicating they would be keen to undertake susceptibility genetic testing if it were available, whilst others were strongly opposed to this possibility. A key aim of the questionnaire, therefore, was to quantify particiapants' views in relation to genetic susceptibility testing for specific conditions. Question 19, sections b-d therefore enquires about participants' attitudes to genetic testing in the future. This item has been specifically developed for this study.

5.5 Supplementary questionnaire

The supplementary questionnaire was issued only to unaffected siblings of the PRAMIS cases, and was not given to the comparison (control) group. This was largely based on issues that emerged as important aspects in phase 1 of the study. This was printed on different coloured paper to the main questionnaire. The main areas included in this supplementary questionnaire are:

Consultation with GP

Sources of information

Desire for access to health care professionals

Adequacy of services available

Smoking behaviour

5.5.1 Consultation with GP

These questions were not based on previously published data, but developed for the purposes of this study in the light of findings of the qualitative phase of the study. Some participants in that phase of the study had visited their GP, with some being investigated regarding their personal risk, and others feeling that their fears were dismissed. The postal survey was therefore a good opportunity to explore this further.

5.5.2 Sources of information

Participants in the qualitative phase seemed to draw upon a number of different sources for information about health-related topics; this was investigated further in the quantitative phase by question 21, which was based on sources identified by participants in the interviews:

The internet

Television

Public library

Books

Newspapers

Magazines

British Heart Foundation literature

Didn't know where to go

Other health care professionals (asked to specify)

5.5.3 Desire for access to health care professionals

A very strong desire expressed by many participants in the qualitative phase of the study, was the need to speak to someone in a professional capacity about their sibling's heart attack. This was explored in a question 24.

5.5.4 Adequacy of services available

An important aspect of Government health policy is to identify individuals at risk of developing CHD, as highlighted in the National Service Framework (Department of Health, 2000). Advances in molecular technology may raise the profile of inherited elements of CHD and lead to new services for individuals with a family history of CHD. Question 25 aims to measure the needs of this group of people in relation to services, and was developed for this study.

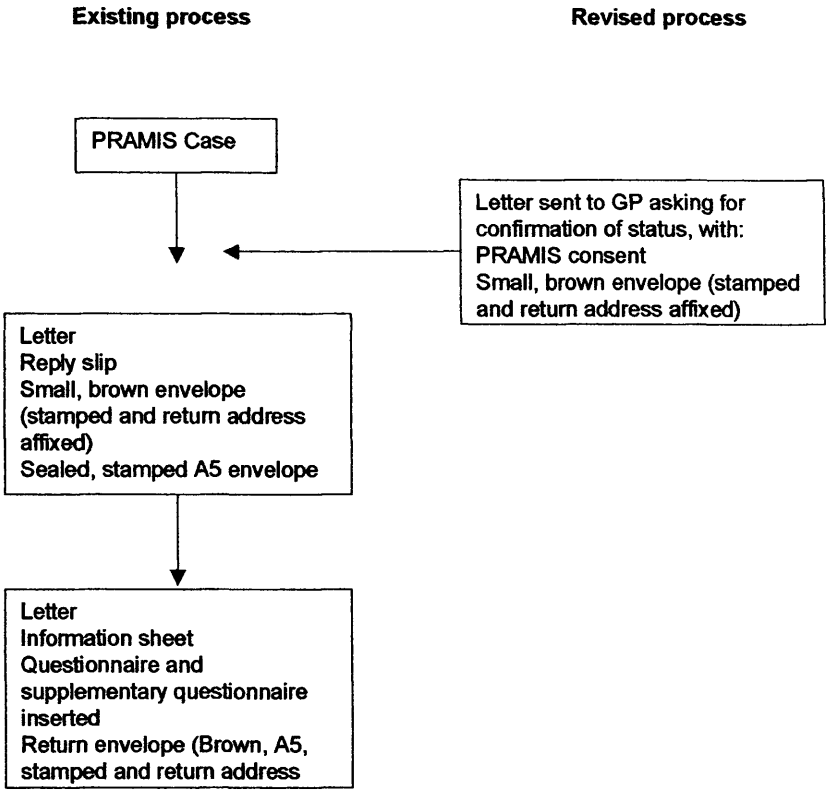
5.5.5 Smoking behaviour

In the same way that current smoking status is utilised as a proxy for health-related behaviour in the main questionnaire, smoking is also used as a mechanism to assess any changes in health-related behaviour following a sibling's heart attack. A question for current smokers was adapted from a question used by the Office for National Statistics, where the topic under investigation was alcohol consumption in young mothers (Office for National Statistics, 1995).

5.6 Problems encountered with administering questionnaires

In November 2003, a spouse of one of the PRAMIS cases contacted the researcher informing him that her husband had deceased since his participation in PRAMIS. Following this, no more questionnaires were issued until written confirmation had been sought from GPs that their patient was still alive. This was undertaken by writing a letter to the GP (appendix 27) explaining that their patient had participated in a previous study and that a further questionnaire based study was being undertaken. The letter was sent from, and signed by Professor Samani (Principal Investigator on PRAMIS) and the researcher on UHL headed paper. Copies of the original PRAMIS consent and a return paid envelope were included in this letter to GPs. This therefore added another stage to the recruitment process, as illustrated in figure 5.2.

Figure 5.2 Modified process of administering the questionnaire to include a letter to GP of PRAMIS participant



In retrospect, this process should have been incorporated into the main protocol for all of the participants, particularly as recruitment for the PRAMIS study had been undertaken from June 1999 to May 2002. This is an important lesson from this study.

5.7 Data entry

All data were entered into a purpose written database using Microsoft Access 2000. An example of a data entry form can seen in appendix 28. Numerical values were assigned to all data, apart from the small number of open-ended questions, where data were entered as free text. For many of the form fields, drop-down options were incorporated in an attempt to reduce data entry errors. Furthermore, only two individuals entered data – the researcher and his partner. Random selections of fields were later checked for data entry errors, but none were found.

5.8 Power

Power calculations are important in quantitative research as a means of establishing how likely the study is to detect a particular effect, for a given sample size, effect size and level of significance (Altman, 1991). In the protocol and ethics application for the quantitative phase of this study, two power calculations were undertaken. Firstly, it was estimated that with a response rate of 60% for the unaffected siblings, and a percentage of the unaffected siblings having a particular risk perception or health belief of 60%, this percentage would be accurate to within 9% of its true value. Secondly, for comparing two proportions, it was estimated that there would be 80% power to detect a difference between the two groups of 18% (at the 5% significance level) (study protocol for quantitative phase, appendix 18).

All power calculations were undertaken with a computer programme PS: Power & Sample Size Calculator version 2.1.31:
(www.mc.vanderbilt.edu/prevmed/ps/index.htm)

Table 5.1 shows a selection of power calculations assuming different proportions for the comparison group. The proportion in the comparison group gives a range of detectable alternatives in the unaffected sibling group. For example, if the proportion in the comparison group is 0.3, the detectable alternative in the unaffected sibling group ranges from 0.125 (if less likely), and 0.5 (if more likely).

Table 5.1 Power calculations for this study

Proportion in comparison group	**Detectable alternative**	
	Less likely	More likely
0.1	0.0024	0.253
0.3	0.125	0.50
0.5	0.29	0.71

The Detectable alternatives in the above table were calculated with the following variables:

Sample size (cases)	59
Significance	0.05
Power	0.8
Ratio of controls to cases	3:1

5.9 Analysis methods used

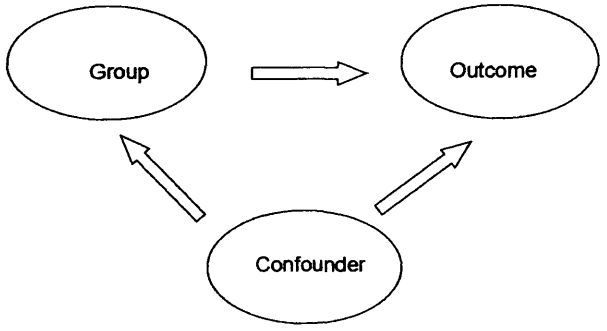
All data were electronically imported to SPSS (Statistical Package for the Social Sciences) for Windows, version 12.0, and a systematic statistical analysis was undertaken of all data. For binary data, the Chi-squared test was used, using cross-tabulations with differences expressed as differences in proportions with 95% confidence intervals. For ordinal data from the Likert scales, a comparison of means was calculated using the two independent samples t-test. The t-test is often applied to Likert scales, and has been utilised in other studies (Wilcox & Stefanick, 1999). However, this test does not meet the textbook assumptions about normality as it is on a 5-point Likert scale. Nevertheless, the test is known to be robust to departures from normality and a recent methodological paper demonstrated it to be particularly useful for ordinal data on scales of three, four or five points by using simulation methods to demonstrate that the test was unbiased and had the appropriate power (Sullivan & D'Agostino, 2003). Therefore, the t-test was appropriate to be utilised in this situation, and calculated differences in mean with 95% confidence intervals. Additionally, the t-test does not collapse data into dichotomous variables. This can be demonstrated by calculating power for continuous data (in this instance from a 5-point Likert scale), with a standard deviation that was frequently found (0.9), gives a detectable alternative value of 0.38, which represents under a half a "Likert unit". This is therefore another justification for utilising this analysis. For the supplemental questionnaire, which was only completed by unaffected siblings of PRAMIS participants, frequency tables were calculated.

5.10 Confounding and adjustment

Large differences were observed between the unaffected siblings and comparison groups for some of the base-line characteristics, particularly in relation to gender and occupation. Since these are important characteristics that may affect some of the attitudinal measurements built into the questionnaire, it was necessary to consider confounding. Confounding is important, and essentially a situation where an outcome will be affected by an external factor (Rothman, 1996). A representation of this can be seen in figure 5.3: the group being the unaffected siblings or the comparison group, the outcome being a variable (for example a belief about how important a particular risk factor is), and the confounder (being the difference in base-line characteristics). The confounder needs to be related to both the group and the outcome to distort the relationship. If only related to one of ether the group or the outcome, then the relationship will not be distorted.

Therefore, in the analysis regression methods were utilised. Adjustment was performed by incorporating terms for gender and occupational group into the regression model. For data with binary outcomes, this was undertaken with logistic regression, and for ordinal data this was undertaken with linear regression. Both of these methods calculated a p-value adjusted for gender and occupation, as well as adjusted effect sizes (Altman, 1991). For the fear of disease question where there are 3 unordered categories, adjustment was carried out using a log linear model incorporating terms for gender and occupation.

Figure 5.3 An illustration of how confounding factors need to affect both the group and the outcome



5.11 Summary

This postal questionnaire was developed following the results of the earlier qualitative phase of the study, which influenced the design of the questionnaire in terms of content and specific topics included. The questionnaire was deliberately kept as short as possible in an effort to maximise response rates (Edwards et al., 2002). There are two important considerations here that influenced the development of the questionnaire; the influence of the qualitative phase of the study, and the necessity to ensure that everything was undertaken to ensure that the response rate was maximised.

Chapter 6 The results of a questionnaire based comparative study of a group of unaffected siblings and a group of individuals where there is no family history of heart disease

6.1 Introduction

This chapter will present the quantitative results of the study, from the postal survey. The following sections will be included: response rates to the questionnaire (section 6.2) results of the main questionnaire (section 6.3), the results of the supplementary questionnaire (section 6.4) and a summary of results (section 6.5).

6.2 Response rates

Figure 6.1 A flow chart illustrating the response rate from unaffected siblings of PRAMIS cases

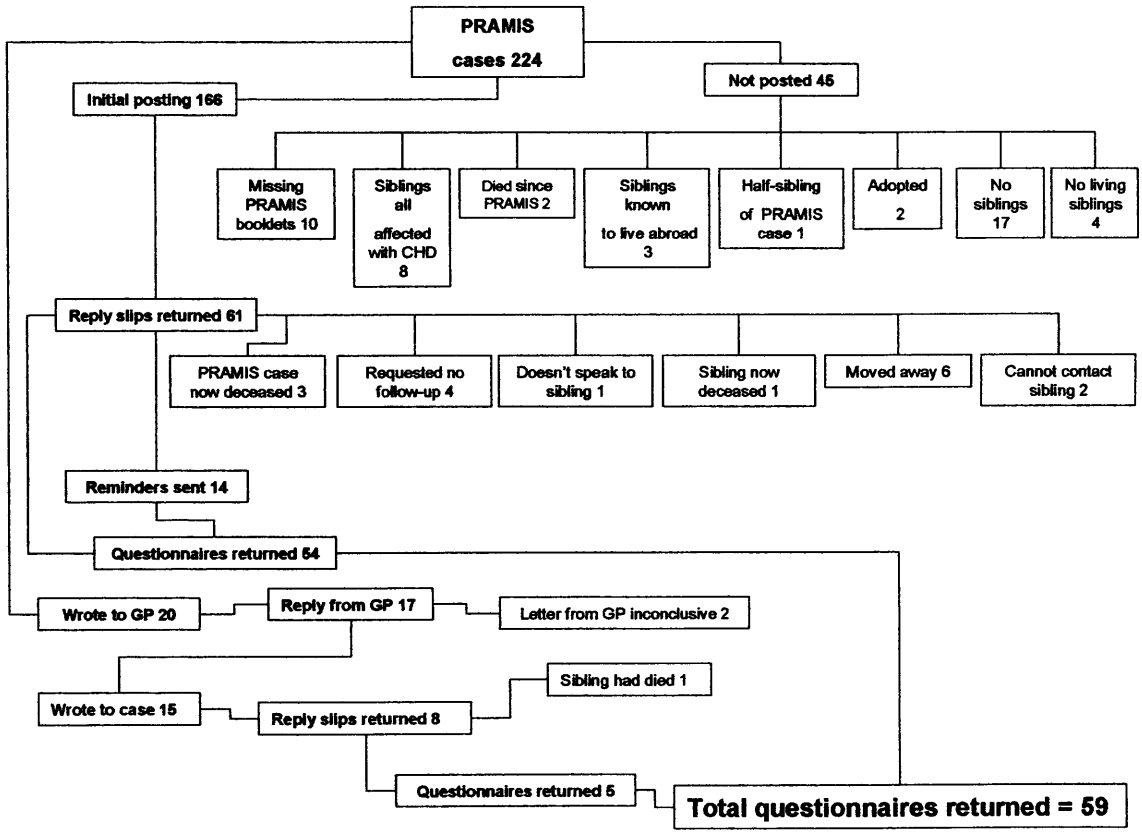


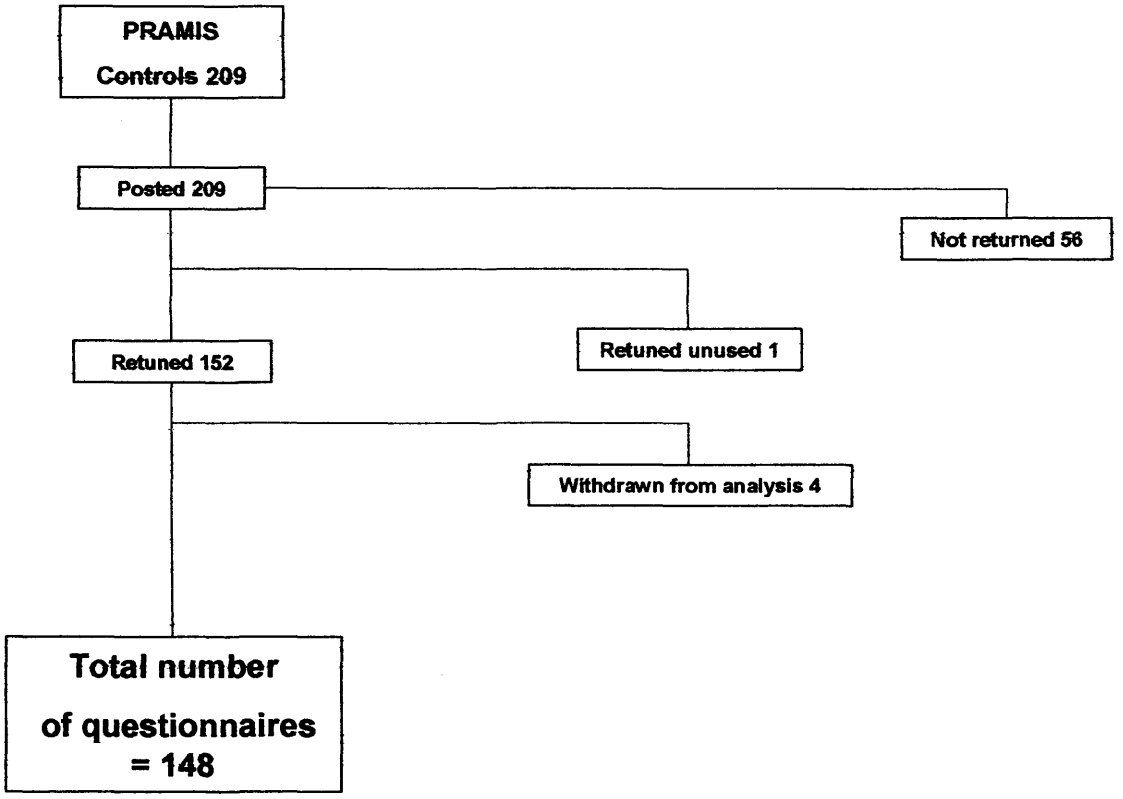
Figure 6.1 demonstrates the complexities involved when contacting siblings in an attempt to recruit them into a study, via their sibling who had participated in a previous study. This will be discussed further in the conclusion of this chapter and in the final chapter. As can be seen from the above figure, of the 224 PRAMIS cases, 166 were initially sent letters

and packs to forward to their siblings. Forty five PRAMIS cases were not included in this initial mail-shot for a variety of reasons, including people who were known to have no siblings (from the PRAMIS database), and other PRAMIS cases for whom there was missing data (the 10 individuals with missing booklets). Sixty-one reply-slips were returned, with 17 PRAMIS cases declining further contact at this stage, or stating that they could not contact their sibling. Three of the PRAMIS cases that were sent letters had deceased since they participated in the PRAMIS study; their spouses notified the researcher of this development. This was when the protocol amendment described in the previous chapter, of writing to the General Practitioners' (GP) of the PRAMIS cases for confirmation that they were still registered with the practice, was implemented. The GPs of 20 PRAMIS cases were written to, with 17 replying. Of these, the details were inconclusive on 2 letters and 15 further PRAMIS cases were contacted. A further 8 reply-slips were returned and 5 questionnaires. The total number of questionnaires from unaffected siblings was therefore 59. The response rate for postal questionnaires from the unaffected siblings of the total number of PRAMIS cases is 26.3%, and of those who received an initial posting (at least to the PRAMIS case), was 32.6%. Although this is low, some participants may not have responded anyway, for reasons unknown, such as having a difficult relationship with their sibling. Participants from the qualitative phase of the study were not excluded from inclusion in this postal survey. In an ideal situation, they would all have been excluded, but because the sample source (unaffected siblings of PRAMIS cases) was such a small group to start with, and there were such significant complexities and difficulties recruiting participants (detailed above), it was decided to include them in the study. This will be discussed in the final chapter.

Recruiting the comparison group from the PRAMIS controls directly was far more straight-forward, as detailed in the figure below. All of the 209 PRAMIS controls were sent questionnaires directly. One hundred and fifty two were returned, and 1 was returned unused. Fifty-six were not returned, and no attempt was made to contact them again. Four questionnaires were withdrawn from the analysis because they indicated on this

questionnaire that they had a first-degree relatives (either a parent or a sibling) who was affected with CHD, although it is unknown whether this was the case at the time of PRAMIS.

Figure 6.2 A flow chart illustrating the response rate from PRAMIS controls (the comparison group for this study)



The response rate therefore is 70.8%, considerably higher than the response rate for the unaffected siblings. However, as these people had already participated in research as the PRAMIS controls, they were probably more likely to respond to the invitation to participate in further research. Since no attempt was made to send reminders to the PRAMIS controls, an even higher response rate might have been achieved if more time and resources had been available to dedicate to this study.

6.3 Main questionnaire

Tables below correspond to sections, questions or groups of statements in the main questionnaire.

6.3.1 Demographic details of study participants

Table 6.1 shows the demographic details of the study participants from the first six questions of the questionnaire

Variable	Unaffected sibling n=59	Comparison group n=148	P-value**
Gender			
Male	23 *(39.0%)**	126 *(85.1%)*	0.001
Female	36 *(61.0%)*	22 *(14.9%)*	
Age (years)			
Mean	50.44	50.96	0.61+
Standard deviation	8.758	5.461	
Minimum	33	31	
Maximum	66	62	
Range	33 years	31 years	
Description of occupation			
Professional	14 *(24.1%)*	52 *(35.4%)*	0.001
Managerial	8 *(13.8%)*	37 *(25.2%)*	
Skilled non-manual	5 *(8.6%)*	13 *(8.8%)*	
Skilled manual	9 *(15.5%)*	31 *(21.1%)*	
Partly skilled	14 *(24.1%)*	6 *(4.1%)*	
Unskilled	8 *(13.8%)*	8 *(5.4%)*	
Age finished education			
16 or under	45 *(77.6%)*	82 *(55.4%)*	0.008
17	2 *(3.4%)*	12 *(8.1%)*	
18	5 *(8.6%)*	9 *(6.1%)*	
19 or over	6 *(10.3%)*	45 *(30.4%)*	

Footnote:
* Figures are counts, with percentages in parentheses. ** Calculated by Chi-Square + Calculated by t-test

There is a very surprising difference in the gender distribution, especially considering similarities in PRAMIS regarding gender, as detailed in table 1 of chapter 2. This is possibly due to the methodology of sending the postal questionnaires via the PRAMIS

cases and then being reliant upon them to forward to one of their unaffected siblings. The mean age of participants was similar for both groups at 50.44 years (unaffected siblings) and 50.96 years (comparison group), ranging from 33-66 (unaffected siblings) and 31-62 (comparison group).

Another interesting aspect of the demographic characteristics of participants in this study is the large and significant difference in occupation. Of the unaffected siblings, 37.9% describe their occupation as either professional or managerial, as compared to 60.6% of the comparison group. Conversely, 37.9% of the unaffected siblings and 9.5% of the comparison group describe their occupation as partly skilled or unskilled. This is statistically significant, with a p-value of 0.001. The age at which participants left school is also very different, with 77% of the unaffected siblings and 55% of the comparison group leaving school at 16 years or younger; 10% of the unaffected siblings and 30% of the comparison group leaving school at 19 years or over. This is possibly a reflection of the differences in recruitment for PRAMIS cases and controls, with PRAMIS cases being recruited via Coronary Care Units in Leicester, and the controls via two GP practices in Leicestershire.

Nevertheless, these differences are problematic as they could interfere with any other differences observed between the groups. It was therefore necessary to adjust for these differences during analysis. This was undertaken by the regression methods described in chapter 5.

6.3.2 PRAMIS: some considerations

Having identified some of these large differences in the base-line characteristics of the participants to this survey, it became necessary to compare these data with the original PRAMIS data. The following tables were created after merging some of the PRAMIS data, with data for this study.

Table 6.2 Gender of PRAMIS cases (whose unaffected siblings completed a questionnaire), compared with the gender of the comparison group

	Affected individuals (PRAMIS Cases)	Comparison group
Male	48 *(87.3%)*	126 *(85.1%)*
Female	7 *(12.7%)*	22 *(14.9%)*

Table 6.2 shows a cross-tabulation between the gender of PRAMIS participants, whose unaffected siblings completed a questionnaire, and the comparison group for the current study. As detailed in chapter 2 (page 25), approximately 14% of PRAMIS cases and controls were female. In this study, 14% of participants in the comparison group were female, and 12.7% of the PRAMIS cases whose siblings completed a questionnaire were female. However, the table 6.3 shows the gender of the unaffected sibling to whom the PRAMIS cases forwarded the questionnaire. This analysis was a cross-tabulation of the gender of participants in the current study with the gender of PRAMIS participants, selecting only the unaffected siblings.

Table 6.3 Gender of unaffected sibling to whom the PRAMIS case forwarded the questionnaire

Gender of sibling questionnaire posted to	Gender of PRAMIS case	
	Male	**Female**
Male	19 *(39.6%)*	0 *(0%)*
Female	29 *(60.4%)*	7 *(100%)*

As can bee seen, all females sent the questionnaire to a sister, and 60% of males sent the questionnaire to a sister. This may reflect an important methodological issue with this study, since PRAMIS cases were asked to forward the information and questionnaire to a sibling of their choice, rather than specifically a sibling of the same gender. A premature heart attack is more common in males than females and PRAMIS cases and controls were matched for gender, with approximately 14% of each group being female. The PRAMIS cases could have been asked to forward the research information to a sibling of the same gender, but that would probably have reduced the sample size even further. Additionally, some imbalance could have been expected for this study. If 50% of PRAMIS cases forwarded the research materials to a sister and 50% to a brother, this would not reflect the gender distribution observed in PRAMIS.

6.3.3 General health of study participants

Table 6.4 shows the general health of study participants from questions 7 to 9 of the main questionnaire.

Table 6.4

Variable	Unaffected sibling	Comparison group	P-value**
Current smoker?			
Yes	11 *(18.6%)**	17 *(11.5%)*	0.17
No	48 *(81.4%)*	131 *(88.5%)*	
Of current smokers, number of cigarettes smoked per day			
1-10	5 *(45.5%)*	7 *(41.2%)*	0.82
11 or more	6 *(54.5%)*	10 *(58.8%)*	
Number of visits to GP in last 2 years			
None	4 *(6.8%)*	21 *(14.2%)*	0.018
1-3 times	24 *(40.7%)*	82 *(55.4%)*	
4-6 times	19 *(32.2%)*	24 *(16.2%)*	
7 times, or more	11 *(18.6%)*	21 *(14.2%)*	
Description of health over the last 12 months			
Good	27 *(46.6%)*	104 *(70.7%)*	0.005
Fairly good	27 *(46.6%)*	36 *(24.5%)*	
Not good	4 *(6.9%)*	7 *(4.8%)*	

Footnote:
* Figures are counts, with percentages in parentheses. ** Calculated by Chi-Square

There are very few current smokers amongst participants, with 18.6% of unaffected siblings and 11.5% of the comparison group identifying themselves as smokers. As there were so few current smokers, these groups were collapsed, for the purpose of analysis, into 2 categories (participants who smoked up-to 10 cigarettes per day and participants who smoked over 10 cigarettes per day). Of current smokers, 54.5% of unaffected siblings smoked more than 10 cigarettes per day, whereas 58.5% of the comparison group smoked more than 10 cigarettes per day. This is a non-significant difference.

The number of participants visiting their GP more than 3 times in the last 2 years, shows a ten percentage point difference: with 40% of unaffected siblings, compared to 30% of the

comparison group. This is a significant difference with a p-value of 0.018. This is quite surprising and may demonstrate that the unaffected siblings experience more illness, or are more concerned about their health than the comparison group. Participants' description of their health is interesting and shows a significant difference, 70% of the comparison group stating their health as "good" in the last 12 months, but only 47% of the unaffected sibling group. The description of health as "fairly good" is 47% for the unaffected siblings, but only 25% of the comparison group. This is statistically significant with a p-value of 0.005.

6.3.4 Sub-analysis by gender: description of health over the last 12 months

As there was such a significant difference in the description of health over the last 12 months, a sub-analysis by gender was undertaken to examine the possible influences of gender on reporting of health. As can be seen from the following tables, although numbers are reduced dramatically, the trend does follow the general pattern for the whole sample. The only exception to this observation being females who describe their health as being "not good", but there were only 2 participants thus.

Table 6.5 Male

	Good	Fairly good	Not good	p-value **
Unaffected siblings	10* *(43.5%)*	10 *(43.5%)*	3 *(13.0%)*	0.028
Comparison group	89 *(71.2%)*	30 *(24.0%)*	6 *(4.8%)*	

Footnote: * Figures are counts, with percentages in parentheses. ** Calculated by Chi-Square

Table 6.6 Female

	Good	Fairly good	Not good	p-value **
Unaffected siblings	17* *(48.6%)*	17 *(48.6%)*	1 *(2.9%)*	0.279
Comparison group	15 *(68.2%)*	6 *(27.3%)*	1 *(4.5%)*	

Footnote: * Figures are counts, with percentages in parentheses. ** Calculated by Chi-Square

Since there are very small numbers involved in these sub-analyses, it is important to consider the trend of the results in relation to the main results displayed in table 6.4 previously. For both males and females the results are similar to the main result.

6.3.5 Risk Factors

Table 6.7 displays the results for views about risk factors for heart disease. Participants in both groups held broadly similar views about risk factors for heart disease. For example, participants in both groups "strongly agree" that the following factors increase the risk of heart disease: 25% of each group for diabetes; 55% of each group for high blood pressure; 48% of each group for genetic factors; and more than 70% of each group for smoking. There are some surprising results, for example in the comparison group 4% disagree (or strongly disagree) that smoking is a risk factor and 5% disagree (or strongly disagree) that eating fatty foods is a risk factor.

There is an interesting difference in the two groups for stress or worry. Stress was discussed by many of the participants in the qualitative phase as being a risk factor or cause of their siblings' heart attack. As can be seen from the table, 16.4% of the comparison group disagree that stress (or worry) is a risk factor, compared to only 1 of the unaffected siblings (1.7%). The difference in the mean is 0.29 (95% confidence interval 0.02-0.56), and a p-value of 0.034. After adjustment, the difference is no longer statistically significant (p-value 0.069), but it is the largest difference in table 6.7.

6.3.6 Ranking of risk factors

Table 6.8 gives the results of the ranking of risk factors. Participants were asked to rank the risk factors that they thought to be most important from the list provided from 1- 5 (1 being the most important). This table contains the details of those risk factors that participants considered to be the top 3 most important risk factors. Ranking of risk factors in the top 3, showed broadly similar results for the two groups. For example, approximately 60% of each group ranked smoking in the top 3 most important risk factors,

and 30% in each group placed eating fatty foods in the top 3. Lack of exercise shows a surprising significant difference (p=0.003), with 23% of the comparison group ranking it in the top 3 risk factors, but only 5% of the unaffected siblings. After adjustment for gender and occupation, this is still significant (p=0.027). However, this result for this risk factor does demonstarte the largest difference in proportions of the 2 groups. Stress or worry was ranked in the top 3 by 46% of the unaffected sibling group and 20% of the comparison group, with a p-value of 0.022 and a difference in proportion of 0.15 (0.01 – 0.30 95% confidence interval). Adjusted for gender and occupation, the p-value of this difference is 0.024. Interestingly, chance or fate was placed in the top 3 risk factors by 2% of the comparison group (3 participants) and none of the unaffected siblings, the p-value was calculated with a Fisher's Exact test, because of small numbers, but was not significant. The difference in proportion and adjusted p-value were not calculated, also because of the small numbers involved.

Table 6.7 Description of views of unaffected siblings and comparison group regarding beliefs about risk factors for heart disease

		Strongly Disagree*	Tend to disagree	Neither	Tend to agree	Strongly agree	Mean**	Difference in mean (95% CI)	p- value ***	Adjusted difference in mean	Adjusted p-value ****
Diabetes increases the risk of heart disease	Unaffected Sibling	0 (0%)	5 (8.6%)	20 (34.5%)	19 (32.8%)	14 (24.1%)	3.72	-0.12 (-0.41 – 0.17)	0.40	-0.07 (-0.40 – 0.27)	0.69
	Comparison group	2 (1.4%)	13 (8.8%)	31 (21.1%)	63 (42.9%)	38 (25.9%)	3.85				
Genetic factors increase the risk of heart disease	Unaffected Sibling	0 (0%)	1 (1.7%)	2 (3.4%)	28 (47.5%)	28 (47.5%)	4.41	0.05 (-0.19 - 0.28)	0.70	0.02 (-0.26 – 0.29)	0.89
	Comparison group	1 (0.7%)	8 (5.4%)	2 (1.4%)	65 (43.9%)	72 (48.6%)	4.36				
Stress or worry increases the risk of heart disease	Unaffected Sibling	1 (1.7%)	0 (0%)	1 (1.7%)	25 (42.4%)	32 (54.2%)	4.47	0.29 (0.02-0.56)	0.034	0.29 (-0.02 – 0.61)	0.069
	Comparison group	2 (1.4%)	10 (6.8%)	12 (8.2%)	57 (38.8%)	66 (44.9%)	4.19				
Eating fatty foods increases the risk of heart disease	Unaffected Sibling	0 (0%)	1 (1.7%)	2 (3.4%)	18 (30.5%)	38 (64.4%)	4.58	0.17 (-0.08 - 0.41)	0.18	0.10 (-0.19 – 0.40)	0.48
	Comparison group	3 (2.0%)	5 (3.4%)	5 (3.4%)	52 (35.1%)	83 (56.1%)	4.41				
Smoking increases the risk of heart disease	Unaffected Sibling	0 (0%)	0 (0%)	1 (1.7%)	16 (27.1%)	42 (71.2%)	4.69	0.05 (-0.17 - 0.27)	0.65	0.01 (-0.25 – 0.27	0.94
	Comparison group	3 (2.0%)	3 (2.0%)	3 (2.0%)	26 (17.6%)	113 (76.4%)	4.64				
Lack of exercise increases the risk of heart disease	Unaffected Sibling	0 (0%)	1 (1.7%)	2 (3.4%)	36 (61.0%)	20 (33.9%)	4.27	-0.06 (-0.30- 0.17	0.60	0.002 (-0.28 – 0.29)	0.99
	Comparison group	3 (2.0%)	4 (2.6%)	7 (4.6%)	63 (41.4%)	75 (49.3%)	4.34				
High blood pressure increases the risk of heart disease	Unaffected Sibling	0 (0%)	1 (1.7%)	2 (3.4%)	24 (40.7%)	32 (54.2%)	4.47	0.06 (-0.17 - 0.29)	0.61	0.006 (-0.27 – 0.28)	0.97
	Comparison group	2 (1.4%)	4 (2.7%)	6 (4.1%)	56 (37.8%)	80 (54.1%)	4.41				
Chance or fate is involved in the development of heart disease	Unaffected Sibling	9 (15.3%)	13 (22.0%)	17 (28.8%)	19 (32.2%)	1 (1.7%)	2.83	0.20 (-0.15 - 0.55)	0.26	-0.005 (-0.41 – 0.40)	0.98
	Comparison group	29 (19.6%)	40 (27.0%)	42 (28.4%)	29 (19.6%)	8 (5.4%)	2.63				
Being overweight increases the risk of heart disease	Unaffected Sibling	0 (0%)	1 (1.7%)	1 (1.7%)	24 (40.7%)	33 (55.9%)	4.51	0.04 (-0.18- 0.27	0.72	-.004 (-0.30 – 0.23)	0.79
	Comparison group	3 (2.0%)	2 (1.3%)	3 (2.0%)	57 (37.5%)	87 (57.2%)	4.47				
High cholesterol increases the risk of heart disease	Unaffected Sibling	0 (0%)	1 (1.7%)	1 (1.7%)	20 (33.9%)	37 (62.7%)	4.58	0.13 (-0.10- 0.36)	0.27	0.007 (-0.27 – 0.28)	0.96
	Comparison group	2 (1.4%)	4 (2.7%)	7 (4.7%)	50 (33.8%)	85 (57.4%)	4.45				

Footnote to table 6.7

* Numerical values were assigned to the responses, for example, Strongly Disagree being 1 → Strongly Agree, being 5

** Therefore, the Mean figure is the mean score on the scale described above

*** Calculated using a T-test

Table 6.8: Ranking of risk factors: those ranked in the top 3

	Unaffected sibling	Comparison group	p-value**	Difference in proportions (95% CI)	Adjusted p-value ***
n =	**56**	**148**			
Diabetes	6* *(10.7%)*	15 *(10.1%)*	0.90	0.006 (-0.09 – 0.10)	0.46
Eating fatty foods	17 *(30.4%)*	46 *(31.1%)*	0.92	-0.007 (-0.15 – 0.13)	0.57
Lack of exercise	3 *(5.4%)*	35 *(23.6%)*	0.003	-0.18 (-0.27 – 0.09)	0.027
Genetic factors	22 *(39.3%)*	54 *(36.5%)*	0.71	0.03 (-0.12 – 0.18)	0.65
Smoking	33 *(58.9%)*	89 *(60.1%)*	0.88	-0.01 (-0.16 – 0.14)	0.75
High blood pressure	26 *(46.4%)*	55 *(37.2%)*	0.23	0.09 (-0.06 – 0.24)	0.19
High cholesterol	26 *(46.4%)*	58 *(39.2%)*	0.35	0.07 (-0.08 – 0.22)	0.83
Stress or worry	20 (35.7%)	30 (20.3%)	0.022	0.15 (0.01 – 0.30)	0.024
Being overweight	14 (25.0%)	59 (39.9%)	0.048	-0.15 (-0.29 - -0.01)	0.042
Chance / fate	0 (0%)	3 (2.0%)	0.56 +	#	#

Footnote:

* Figures are counts, with percentages in parentheses. ** Calculated by Chi-Square

\+ Due to very small numbers, this was calculated with Fisher's Exact test

\# Not calculated due to small numbers

*** Adjusted for gender and occupation with Logistic regression

6.3.7 Knowledge of unaffected siblings and comparison group about heart disease

Table 6.9 displays the results for the four statements regarding knowledge of heart disease in the context of cancer, gender and risk in relation to having a parent or sibling affected with heart disease. While these were based on previously published work, the topics for the statements were developed from the qualitative phase of the study. During the interviews, many participants seemed unaware of the seriousness and significance of heart disease for quality of life, particularly in relation to cancer, gender differences and how important family history is as a risk factor (in relation to either parents or siblings). In relation to the first statement, CHD remains the single largest cause of death in the UK, accounting for the deaths of 24% of men and 17% of women in the UK (Petersen, Peto & Rayner, 2004).

Broadly similar results are found between the groups for the statement "more people die from cancer than heart disease", with 37.9% of the unaffected siblings and 36.3% of the comparison group agreeing (or strongly agreeing) with the statement. The difference in mean is 0.13, 95% confidence interval = -0.22 – 0.47 (Adjusted difference in mean = 0.07, 95% confidence interval = -0.33 - 0.48).

In the qualitative phase of this study, and indeed in the general population, CHD is viewed as a predominantly male condition, which resulted in the inclusion of the second statement about gender differences of CHD. While deaths from CHD are higher in younger men, death rates among women post-menopause rapidly catch-up and are almost equal by the age-group 65-74 years. Therefore, the statement "A man over age 65 is much more likely to die from heart disease than a woman over age 65" is inaccurate, but has been used in previous studies (Wilcox & Stefanick, 1999) as a method of measuring knowledge about heart disease. Interestingly, there are significant differences between the two groups regarding this statement. 39.7% of unaffected siblings and 80% of the comparison group agree (or strongly agree) with this statement. Similarly 32.8% of

unaffected siblings and 11.5% of the comparison group disagree (or strongly disagree), with the statement. The difference in mean is –0.64 (95% confidence interval = -0.93 - -0.35), with a p-value of 0.001. Adjusted for gender and occupational class the difference in the mean is -0.48 (95% confidence interval = -0.82- -0.15), with a p-value of 0.005.

The final two statements relate to genetic risk for CHD, and having a first-degree relative (parent or sibling) increases an individual's relative risk of CHD by up-to ten-fold (Samani & Singh, 2001). Regarding the statement "having a parent with heart disease, increases my risk of developing the disease", 15% of the unaffected siblings disagreed (or strongly disagreed), compared to 6.8% of the comparison group. However, overall for this statement there were broadly similar results as approximately 80% of each group agreed (or strongly agreeing) with the statement. Broadly similar results are found for the statement "having a sibling with heart disease increases my risk of developing the disease", with approximately 75% of participants in each group agreeing (or strongly agreeing) with the statement (difference in mean = -0.02, 95% confidence interval = -0.30 – 0.26) (adjusted difference in mean = -0.12, 95% confidence interval = -0.44 – 0.03). Interestingly, over 10% in each group disagreed (or strongly disagreed) with the statement.

Table 6.9 Knowledge of unaffected siblings and comparison group about for heart disease

		Strongly Disagree *	Tend to disagree	Neither	Tend to agree	Strongly agree	Mean **	Difference in mean	p- value ***	Adjusted difference in mean	Adjusted p-value ****
More people die from cancer than heart disease	Unaffected Sibling	3 *(5.2%)*	19 *(32.8%)*	14 *(24.1%)*	21 *(36.2%)*	1 *(1.7%)*	2.97	0.13 (-0.22 - 0.47)	0.47	0.07 (-0.33- 0.48)	0.78
	Comparison group	14 *(9.5%)*	62 *(41.9%)*	18 *(12.2%)*	42 *(28.2%)*	12 *(8.1%)*	2.84				
A man over age 65 is much more likely to die from heart disease than a woman over age 65	Unaffected Sibling	4 *(6.9%)*	15 *(25.9%)*	16 *(27.6%)*	20 *(34.5%)*	3 *(5.2%)*	3.05	-0.64 (-0.93 - -0.35)	0.001	-0.48 (-0.82- -0.15)	0.005
	Comparison group	4 *(2.7%)*	13 *(8.8%)*	27 *(18.2%)*	85 *(57.4%)*	19 *(12.8%)*	3.69				
Having a parent with heart disease increases my risk of developing the disease	Unaffected Sibling	2 *(3.4%)*	7 *(12.1%)*	2 *(3.4%)*	31 *(53.4%)*	16 *(27.6%)*	3.90	-0.12 (-0.38 - 0.14)	0.38	-0.21 (-0.51 - 0.09)	0.18
	Comparison group	0 *(0%)*	10 *(6.8%)*	12 *(8.1%)*	92 *(62.2%)*	34 *(23%)*	4.01				
Having a sibling with heart disease increases my risk of developing the disease	Unaffected Sibling	3 *(5.1%)*	4 *(6.8%)*	9 *(15.3%)*	31 *(52.5%)*	12 *(20.3%)*	3.76	-0.02 (0.30- 0.26)	0.88	-0.12 (-0.44 - 0.03)	0.47
	Comparison group	3 *(2.0%)*	12 *(8.1%)*	22 *(14.9%)*	88 *(59.5%)*	23 *(15.5%)*	3.78				

Footnote to table 4:

* Numerical values were assigned to the responses, for example, Strongly Disagree being 1 → Strongly Agree being 5

** Therefore, the Mean figure is the mean score on the scale described above

*** Calculated using a T-test

**** Adjusted for gender and occupation with linear regression

6.3.8 Chance of getting heart disease, stroke or cancer amongst unaffected siblings and comparison group

Table 6.10 shows the results for participants' perception of their personal risk for three main diseases (CHD, cancer and stroke), compared to people of their age and sex. There are some important and highly significant results. There are very large differences between the two groups for both heart disease and stroke (p=0.001 for both), but broadly similar results for cancer. The results are a response to a statement regarding a chance of getting the diseases, and if the results for high and very high chance are combined, approximately 39% of unaffected siblings felt that they were at increased risk of getting heart disease, compared to approximately 7% of the comparison group. Similarly, 28% of unaffected siblings, compared to 7% of the comparison group felt that they had a higher risk of having a stroke. For both of these diseases, approximately 55% of respondents felt that they were at average risk. When these results are compared to the findings for cancer, a very different picture is seen, as there are broadly similar results for the two groups regarding their chances of getting cancer, with the difference in mean being –0.02 (95% confidence interval = -0.24 –0.19). Adjustment for gender and occupation made very little difference for this result.

A particularly interesting observation in relation to these results is that in the qualitative phase of the study, participants appeared indifferent to their personal risk of having a heart attack, and there appeared to be a strong sense of detachment from their siblings' heart attack (see chapter 4). Furthermore, many participants expressed far more concern about cancer than they did about heart disease. This result, however, was probably expected as participants in previous studies have viewed heart attacks as a favoured mode of death, particularly in relation to cancer (Emslie et al., 2001).

Table 6.10 Compared with other people of *your* age and sex, what do you think are the chances of you getting the following diseases in the next 15 years?

		Very low*	Low	Average	High	Very high	Mean **	Difference in mean	p-value ***	Adjusted difference in mean	Adjusted p-value ****
Heart disease	Unaffected Sibling	2 *(3.5%)*	1 *(1.8%)*	32 *(56.1%)*	19 *(33.3%)*	3 *(5.3%)*	3.35	0.80 (0.55- 1.05)	0.001	0.94 (0.65- 1.22)	0.001
	Comparison group	16 *(10.9%)*	46 *(31.3%)*	75 *(51.0%)*	8 *(5.4%)*	2 *(1.4%)*	2.55				
Stroke	Unaffected Sibling	2 *(3.4%)*	6 *(10.3%)*	34 *(58.6%)*	16 *(27.6%)*	0 *(0%)*	3.1	0.54 (0.55- 1.05)	0.001	0.63 (0.36- 0.91)	0.001
	Comparison group	14 *(9.5%)*	47 *(32.0%)*	76 *(51.6%)*	9 *(6.1%)*	1 *(0.7%)*	2.56				
Cancer	Unaffected Sibling	2 *(3.4%)*	9 *(15.5%)*	42 *(72.4%)*	5 *(8.6%)*	0 *(0%)*	2.86	-0.02 (-0.24 – 0.19)	0.84	-0.04 (-0.29 - 0.22)	0.78
	Comparison group	6 *(4.1%)*	29 *(19.7%)*	91 *(61.9%)*	18 *(12.2%)*	3 *(2.0%)*	2.88				

Footnote to table 5:

* Numerical values were assigned to the responses, for example, Very Low Disagree being 1 → Very High being 5

** Therefore, the Mean figure is the mean score on the scale described above

*** Calculated using a T-test

**** Adjusted for gender and occupation

6.3.9 Disease most feared

Given the previous results about participants' perception of their chance of getting any of these three diseases (heart disease, stroke and cancer), the results for the item which enquired about the disease that participants feared the most is extremely interesting. The following table shows the results for the question "which one of the following illnesses do you fear the most?"

Table 6.11

	Unaffected sibling	Comparison group	p-value**	Difference in proportions (95% CI)	Adjusted p-value +
Heart disease	12* *(20.7%)*	9 *(6.3%)*	0.001	0.14 (0.03 - 0.26)	0.001
Stroke	18 *(31.0%)*	27 *(18.8%)*		0.12 (-0.01 – 0.26)	
Cancer	28 *(48.3%)*	108 *(75.0%)*		-0.27 (-0.41 - - 0.12)	

Footnote: * Figures are counts, with percentages in parentheses. ** Calculated by Chi-Square
+ Calculated from log linear regression model

This is a very important and highly significant result with a p-value of 0.001. Particularly in light of results from the previous table statement about the chance of getting one of these conditions, where there were very similar results between the two groups for cancer, but significant results for heart disease and stroke (see table 6.10). It is interesting therefore that the results in this table show that 75% of the comparison group fear cancer the most, compared to 48% of unaffected siblings. There are also big differences with heart disease and stroke. It is very surprising that far more of the unaffected siblings fear stroke than the comparison group (31%, compared to 19%). Indeed, more of the unaffected siblings fear stoke than they do heart disease. Heart disease sees a three-fold difference, with 21% of the unaffected sibling group fearing heart disease, compared to 6% of the comparison group.

In the qualitative phase of this study, participants overwhelmingly stated that they were more frightened of cancer than heart disease. It is interesting therefore, that just under 50% of the unaffected sibling group stated that they feared cancer more than either stroke or heart disease. The adjusted p-value was calculated in SPSS using a log linear model incorporating terms for gender and occupation.

Sub-analysis by gender: Disease most feared

When these results were sub-analysed by gender, very similar results are found, although naturally the numbers within the groups are dramatically reduced. Importantly, the trends remain the same as the results in the main table.

Table 6.12 Disease most feared: males

	Heart disease	Stroke	Cancer	p-value **
Unaffected siblings	5* *(22.7%)*	8 *(36.4%)*	9 *(40.9%)*	0.005
Comparison group	8 *(6.6%)*	24 *(19.7%)*	90 *(73.8%)*	

Footnote: * Figures are counts, with percentages in parentheses. ** Calculated by Chi-Square

Table 6.13 Disease most feared: females

	Heart disease	Stroke	Cancer	p-value **
Unaffected siblings	7* *(19.4%)*	10 *(27.8%)*	19 *(52.8%)*	0.073
Comparison group	1 *(4.5%)*	3 *(13.6%)*	18 *(81.8%)*	

Footnote: * Figures are counts, with percentages in parentheses. ** Calculated by Chi-Square

6.3.10 Views of unaffected siblings and comparison group about their ability to reduce their risk of certain diseases

Participants' views on their ability to reduce their personal risk of developing three disease groups (heart disease, stroke and cancer) were assessed. Interestingly, fewer participants in either group thought that they could reduce their risk of cancer compared to other conditions. Table 6.14 shows broadly similar results across the two groups for both stroke and cancer. The mean score for stroke was 1.93 in both groups (difference in mean = -0.001, 95% confidence interval = -0.24 – 0.24), and for cancer = 2.33 (unaffected siblings) and 2.32 for comparison group), difference in mean = 0.01 (95% confidence interval = -0.25 – 0.27). There is a small difference in the views of the two groups regarding heart disease. 49% of the unaffected siblings, compared to 64% of the comparison group stated that "yes, definitely" they could reduce their risk of heart disease. The overall difference in mean was 0.16 (95% confidence interval =-0.3- 0.35), and p-value = 0.097. Adjusted for gender and occupational class this reduced to a difference in mean of 0.04 (95% confidence interval =-0.18 –0.26), and a p-value of 0.71.

Table 6.14 Do you think it is possible for <u>you</u> to do anything to reduce your risk of the following diseases?

		Yes, definitely	Yes, maybe	No, probably not	No, definitely not	Mean	Difference in mean * (95% CI)	p-value	Adjusted difference in mean (95% CI)	Adjusted p-value ****
Heart disease	Unaffected Sibling	29 *(49.2%)*	25 *(42.4%)*	5 (8.5%)	0 (0%)	1.59	0.16 (-0.3 – 0.35)	0.097	0.04 (-0.18- 0.26)	0.71
	Comparison group	94 (63.5%)	44 (29.7%)	10 (6.8%)	0 (0%)	1.43				
Stroke	Unaffected Sibling	16 (27.6%)	30 (51.7%)	12 (20.7%)	0 (0%)	1.93	-0.001 (-0.24- 0.24)	0.99	-0.10 (-0.39- 0.18	0.46
	Comparison group	52 (35.1%)	57 (38.5%)	36 (24.3%)	3 (2.0%)	1.93				
Cancer	Unaffected Sibling	9 (15.5%)	24 (41.4%)	22 (37.9%)	3 (5.2%)	2.33	0.01 (-0.25- 0.27)	0.94	0.000 (-0.31- 0.31)	0.99
	Comparison group	31 (20.9%)	50 (33.8%)	56 (37.8%)	11 (7.4%)	2.32				

Footnote to table 6:
* Numerical values were assigned to the responses, for example, Yes Definitely being 1 → No, Definitely Not, being 4
** Therefore, the Mean figure is the mean score on the scale described above
*** Calculated using a T-test
**** Adjusted for gender and occupation

6.3.11 Health as a value

Table 6.15 shows the results for the statements regarding health as a value. For the first two statements: "If you don't have your health, you don't have anything" and "Good health is only of minor importance in a happy life", there are broadly similar results for the two groups. For the first statement, the difference in mean is 0.31 (4.14 for unaffected siblings, and 3.83 for the comparison group) (95% confidence interval = –0.04 – 0.65). Adjusted for gender and occupation, the difference in mean is 0.23 (95% confidence interval = -0.18 – 0.63). For the second statement, results are also broadly similar between the groups, with approximately 80% in each disagreeing (or strongly disagreeing) that health is only of minor importance.

Interestingly, however, there is a statistically significant difference between the two groups for the statement "there are few things more important than good health". 37% of the unaffected siblings disagree (or strongly disagree), as opposed to 15% of the comparison group. The overall difference in mean is –0.67 (95% confidence interval = -1.06 - -0.28) and a p-value of 0.001. Adjusted for gender and occupation, the results are still significant, with the difference in the mean of –0.66 (95% confidence interval = -1.11 - -0.20), and p-value = 0.005. It is unclear why there should be such a difference for this one statement, when the results for the others are so similar. However, all of these statements have been used in previous research (Lau et al., 1986).

Table 6.15 Views of health as a value among unaffected siblings and comparison group

		Strongly Disagree	Tend to disagree	Neither	Tend to agree	Strongly agree	Mean	Difference in mean (95% CI)	p-value*	Adjusted difference in mean (95% CI)	Adjusted p-value ****
If you don't have your health, you don't have anything	Unaffected Sibling	0 *(0%)*	8 *(13.6%)*	5 *(8.5%)*	17 *(28.8%)*	29 *(49.2%)*	4.14	0.31 (-0.04 - 0.65)	0.082	0.23 (-0.18 - 0.63)	0.27
	Comparison group	3 *(2.0%)*	28 *(18.9%)*	11 *(7.4%)*	55 *(37.2%)*	*51 (34.5%)*	3.83				
Good health is only of minor importance in a happy life	Unaffected Sibling	29 (49.2%)	17 (28.8%)	1 (1.7%)	7 (11.9%)	5 (8.5%)	2.02	0.22 (-0.14 - 0.55)	0.195	0.06 (-0.33 -0.45)	0.77
	Comparison group	68 *(45.9%)*	61 *(41.2%)*	3 *(2.0%)*	13 *(8.8%)*	3 *(2.0%)*	1.80				
There are few things more important than good health	Unaffected Sibling	13 *(22.0%)*	9 *(15.3%)*	3 (5.1%)	11 (18.6%)	23 (39.0%)	3.37	-0.67 (-1.06 - -0.28)	0.001	-0.66 (-1.11 - -0.20)	0.005
	Comparison group	5 *(3.4%)*	17 *(11.5%)*	8 *(5.4%)*	55 *(37.2%)*	63 *(42.6%)*	4.04				

Footnote to table 8:

* Numerical values were assigned to the responses, for example, Strongly Disagree being 1 → Strongly Agree being 5

** Therefore, the Mean figure is the mean score on the scale described above

*** Calculated using a T-test

**** Adjusted for gender and occupation

6.3.12 Genetic developments

Participants' views about advances in genetic technology were one of the key themes from the qualitative phase of this study. This was further explored in the postal survey, the results of which are detailed in table 6.16. Broadly similar results were found between each group. For example, 75% of the unaffected siblings and 83% of the comparison group agreed (strongly or tend to) that genetic developments would bring cures for many diseases. Similarly, only 12% of unaffected siblings and 7% of the comparison group agreed that research on human genetics was tampering with nature and unethical.

Interestingly, 15% of the comparison group disagreed that genetic research has the potential to improve their health, compared to only one participant (1.7%) of the unaffected sibling group. Overall, the mean scores for this statement were 3.97 (unaffected siblings) and 3.76 (comparison group), the difference in mean = 0.20 (95% confidence interval = -0.08 – 0.49), adjusted for gender and occupational class, mean = 0.12 (95% confidence interval = -0.22 – 0.46)

The two groups had similar beliefs for the idea that genetic research would lead to improvements in the health of future generations. Only 5% of the comparison disagreed, compared to one participant (1.7%) of the unaffected sibling group. Overall the mean scores were 4.26 (unaffected siblings) and 4.22 (comparison group), with the difference in mean being 0.04 (95% confidence interval = -0.21 – 0.29), adjusted for gender and occupational class, mean = 0.003 (95% confidence interval = -0.29 – 0.30).

Table 6.16 Views about developments in genetic technology among unaffected siblings and comparison group

		Strongly Disagree	Tend to disagree	Neither	Tend to agree	Strongly agree	Mean	Difference in mean (95% CI)	p-value*	Adjusted difference in mean (95% CI)	Adjusted p- value ****
New genetic developments will bring cures for many diseases	Unaffected Sibling	0 *(0%)*	4 *(6.8%)*	11 *(18.6%)*	30 *(50.8%)*	14 *(23.7%)*	3.92	-0.12 (-0.34 - 0.12)	0.30	-0.17 (-0.43- 0.10)	0.21
	Comparison group	1 *(0.7%)*	5 *(3.4%)*	13 *(8.8%)*	98 *(66.2%)*	31 *(20.9%)*	4.03				
Research on human genetics is tampering with nature and is therefore unethical	Unaffected Sibling	19 *(32.2%)*	19 *(32.2%)*	14 *(23.7%)*	5 *(8.5%)*	2 *(3.4%)*	2.19	0.17 (-0.14- 0.47)	0.28	-0.03 (-0.38 - 0.32)	0.87
	Comparison group	50 *(33.8%)*	58 *(39.2%)*	30 *(20.3%)*	7 *(4.7%)*	3 *(2.0%)*	2.02				
Genetic research has the potential to improve *my* health	Unaffected Sibling	0 *(0%)*	1 *(1.7%)*	14 *(23.7%)*	30 *(50.8%)*	14 *(23.7%)*	3.97	0.20 (-0.08 - 0.49)	0.17	0.12 (-0.22 -0.46	0.47
	Comparison group	3 *(2.0%)*	19 *(12.8%)*	22 *(14.9%)*	70 *(47.3%)*	34 *(23.0%)*	3.76				
Genetic research will lead to improvements in the health of future generations	Unaffected Sibling	0 *(0%)*	1 *(1.7%)*	3 *(5.2%)*	34 *(58.6%)*	20 *(34.5%)*	4.26	0.04 (-0.21- 0.29	0.78	0.003 (-0.29 - 0.30)	0.98
	Comparison group	3 *(2.0%)*	5 *(3.4%)*	11 *(7.4%)*	66 *(44.6%)*	63 *(42.6%)*	4.22				

Footnote to table 9:

* Numerical values were assigned to the responses, for example, Strongly Disagree being 1 → Strongly Agree being 5

** Therefore, the Mean figure is the mean score on the scale described above

*** Calculated using a T-test **** Adjusted for gender and occupation

6.3.13 Genetic susceptibility testing

In the context of genetic developments, one of the key findings of the qualitative phase of this study was participants' views about the possibility of genetic susceptibility testing. While this was explored in some depth during the interviews, the questionnaire allowed for quantification of these views. Overall, for the three disease categories, participants' appeared to be amenable to genetic susceptibility genetic testing. For example, the majority of both groups (86% of unaffected siblings, and 83% of the comparison group) agree (or strongly agree) that they would take a genetic susceptibility test for heart disease. Similarly, for stroke and cancer, the participants who agree that they would have a test, is in the region of 80%. There are however, a small number of the comparison group who appear resistant to this type of testing.

As some gender differences were observed in the qualitative phase, a sub-analysis of this data by gender was undertaken. The results were broadly similar to those presented in the table 6.17, and can be viewed in appendix 29. Although there are some differences between the unaffected sibling and comparison groups in these results, caution must be used when interpreting them due to small numbers.

Table 6.17 Views of unaffected siblings and comparison group on the possibility of genetic susceptibility testing for common diseases

		Strongly Disagree	Tend to disagree	Neither	Tend to agree	Strongly agree	Mean	Difference in mean (95% CI)	p-value*	Adjusted difference in mean (95% CI)	Adjusted p-value ****
If a genetic test were available, that could indicate that I was at increased risk of developing heart disease, I would want to have such a test	Unaffected Sibling	0 *(0%)*	6 *(10.2%)*	2 *(3.4%)*	22 *(37.3%)*	29 *(49.2%)*	4.25	0.18 (-0.13 - 0.49)	0.25	0.20 (-0.16- 0.55)	0.28
	Comparison group	6 *(4.1%)*	10 *(6.8%)*	8 *(5.4%)*	67 *(45.3%)*	57 *(38.5%)*	4.07				
If a genetic test were available, that could indicate that I was at increased risk of having a stroke, I would want to have such a test	Unaffected Sibling	0 *(0%)*	5 *(8.5%)*	2 *(3.4%)*	25 *(42.4%)*	27 *(45.8%)*	4.25	0.18 (-0.13 - 0.49)	0.26	0.20 (-0.17- 0.56)	0.29
	Comparison group	7 *(4.7%)*	10 *(6.8%)*	9 *(6.1%)*	61 *(41.2)*	61 *(41.2%)*	4.07				
If a genetic test were available, that could indicate that I was at increased risk of developing cancer, I would want to have such a test	Unaffected Sibling	0 *(0%)*	5 *(8.5%)*	1 *(1.7%)*	21 *(35.6%)*	32 *(54.2%)*	4.36	0.29 (-0.03- 0.58)	0.079	0.26 (-0.12- 0.63)	0.18
	Comparison group	8 *(5.4%)*	10 *(6.8%)*	10 *(6.8%)*	56 *(37.8%)*	64 *(43.2%)*	4.07				

Footnote to table 10: * Numerical values were assigned to the responses, for example, Strongly Disagree being 1 → Strongly Agree being 5** Therefore, the Mean figure is the mean score on the scale described above

*** Calculated using a T-test **** Adjusted for gender and occupation

6.4 Supplementary Questionnaire for unaffected siblings only

Table 6.18 shows the all of the data from the supplementary questionnaire that was included with the main questionnaire for completion by the unaffected sibling group only. These questions and statements were all developed from the qualitative phase of the study, and represent some of the topics that participants in that phase of the study highlighted. Of particular interest from that phase of the study, and which had not been explored further in the main questionnaire (because it would have been inappropriate to do so with the comparison group), are the participants' experiences of consulting their GP following their siblings' heart attack, sources of health-related information, their views about service provision for unaffected siblings of people who had a heart attack and whether they had modified their cigarette smoking habits (for those who smoke). As discussed before, smoking was utilised as a proxy for health-related behaviour in this study. Additionally, because of the problems involved with accessing unaffected siblings, and the disappointingly low response rate, there is a limited sample size.

As can be seen, only 32% of participants visited their GP or other health care professional following their siblings' heart attack. Of those participants who did visit their GP or other health professional, all found the consultation helpful. This would seem a small percentage of unaffected siblings who were concerned enough by their siblings' heart attack to seek advice from their GP or other health professional.

The list of options, for source of information regarding inherited aspects of heart disease, was directly linked to the qualitative phase of the study as participants had mentioned many of these as sources of information that they had used. For this question, participants were provided with a list of sources asked to tick all that apply. From the questionnaires it is apparent that unaffected siblings in the quantitative phase of the study also utilise a variety of information sources. Fifteen percent reported accessing British Heart Foundation information (leaflets) as a source, and 18% reported that they were unaware of suitable information sources. Six percent reported that they had used the internet as a

source of information, and only 6% stated that they had sought information from other health care professionals. When asked to specify which healthcare professionals they had sought information from, 3 participants stated their GP. All other responses were singular and are listed in the table with examples including family nurse, diabetic clinic.

Participants were asked to respond to the statement "I needed to speak to a health professional in the months following my brother or sister's heart attack", 30% agreed (or strongly agreed) with this statement, and 24% disagreed (or strongly disagreed). Thirty-nine percent, however, would not commit themselves either way. Responding to the statement "adequate services are available for understanding inherited aspects of heart disease" 43% of participants agreed (or strongly agreed), whilst 32% disagreed (or strongly disagreed), with 20% not committing themselves either way.

Of current smokers, which only represent 13 participants in the unaffected sibling group, 7 (12% of the group, or 53% of smokers) stated that they smoked less than before their siblings' heart attack, six participants (10% of the group, or 47% of smokers) stated that they smoked about the same.

Table 6.18 Supplementary questionnaire for unaffected siblings only

In the months following your brother or sister's heart attack, did you visit your family doctor or a health profession specifically to discuss any concerns you might have had as a consequence of this?	Yes 18 *(32%)* No 39 *(68%)*	
If yes: I found the consultation with my family doctor very helpful:	Strongly disagree Tend to disagree Neither Tend to agree Strongly agree	0 *(0%)* * 0 *(0%)* 1 *(5.5%)* 10 *(55.5%)* 8 *(44.4%)*
Did you seek ***information*** about inherited aspects of heart disease from any of the following sources? (tick all that apply)	The internet Television Public library Books Newspapers Magazines BHF literature Didn't know where to go Other health care professionals	4 *(6.8%)* 3 *(5.1%)* 1 *(1.7%)* 4 *(6.8%)* 1 *(1.7%)* 1 *(1.7%)* 9 *(15.3%)* 11 *(18.6%)* 4 *(6.8%)*
For other health care professionals, GP was cited three-times, otherwise the following list includes all other single responses:	Diabetic clinic Doctor Family Family Nurse Leaflets Pharmacist	
I needed to speak to a health professional in the months following my brother or sister's heart attack:	Strongly disagree Tend to disagree Neither Tend to agree Strongly agree	9 *(15.3%)* 5 *(8.5%)* 23 *(39.0%)* 10 *(15.3%)* 9 *(15.3%)*
Adequate services are available for understanding inherited aspects of heart disease:	Strongly disagree Tend to disagree Neither Tend to agree Strongly agree	3 *(5.1%)* 16 *(27.1%)* 12 *(20.3%)* 20 *(33.9%)* 6 *(10.2%)*
If you smoke, please answer the following: Compared to **before** your brother or sister's heart attack, would you say you smoke more, less or about the same nowadays?	I smoke **much more** nowadays I smoke **more** Nowadays I smoke **about the same** nowadays I smoke **less** Nowadays I smoke **much less** nowadays	0 *(0%)* 0 *(0%)* 6 *(10.2%)* 3 *(5.1%)* 4 *(6.8%)*

* The percentages here refer to the 18 people who visited a health professional following their siblings' heart attack

6.5 Summary

The quantitative phase of this study enhanced and built upon the findings of the qualitative phase. The topics included in the questionnaire were strongly influenced by the earlier qualitative study although many of the questions or statements had been utilised in previously published work, because it was important to use items from previous questionnaires where possible so that comparisons with previous work could be drawn. There were considerable difficulties in recruiting unaffected siblings of the PRAMIS participants, particularly in relation to the controls, (who were recruited directly for the comparison group). It was considered to be the only ethical way to undertake this form of work as the siblings of PRAMIS participants had not agreed, or consented, to have any direct contact with PRAMIS or any subsequent research. This issue is discussed further in the next chapter. However, because of the nature of the recruiting process, an imbalance in the gender of the two groups was found in this study, which was not found in PRAMIS. The impact of this for the current study was that nearly all of the results required adjustment for gender and occupational class differences. However, in virtually all instances this did not affect interpretation of the results. Nevertheless, some interesting and important findings were observed from this postal questionnaire.

Knowledge of heart disease, particularly gender attribution, the relationship to other diseases and the risk associated with having parents or siblings affected with the disease were all measured in the questionnaire. The perception of heart disease in relation to cancer has been utilised in much previous research (Shepherd et al., 1998, Wilcox & Stefanick, 1999) and the findings of this study regarding the perception of the number of people who die from heart disease, is in line with national and international findings (Shepherd et al., 1998). The perception of gender differences in heart disease is quite profound, with heart disease being viewed as a predominantly male condition, which was also found in the study from which the statement was also utilised (Wilcox & Stefanick, 1999). Participants in the questionnaire phase of this study appeared to acknowledge the risk associated with having a parent or sibling affected with the condition, which often

required prompting in the qualitative phase of this study, and in previous research (Hunt et al., 2001).

Important, and highly significant differences were found in the two groups regarding perception of ability to reduce personal risk of various diseases. For this, and some subsequent statements / questions three disease categories of heart disease, stroke and cancer were utilised as comparative disease processes. This was again based on Wilcox & Stefanick (Wilcox & Stefanick, 1999), but they had specified heart disease in relation to breast, lung and colon cancer. For this study, cancer was used as a generic term, which may have been a limiting factor particularly for female participants. However, in this study far more unaffected siblings than the comparison group stating that they felt to be at higher risk than the general population for heart disease and stroke, but appeared to express similar feelings of personal risk for cancer. This is particularly interesting when the fear of a certain disease is explored, as very high proportions of both groups (48% of unaffected siblings and 75% of the comparison group) feared cancer more. Certainly in the qualitative phase of the study cancer was discussed by many participants as a disease that they were far more frightened of than heart disease, which was often discussed in terms of being a favoured mode of death, as has been described before (Emslie et al., 2001). The perception of ability to reduce the risk of the three disease categories, (heart disease, stroke and cancer) was measured in this questionnaire, adapted from Wilcox & Stefanick, 1999, which demonstrated similar results for both groups.

The attitudes of participants to genetic developments and the possibility of genetic susceptibility testing was measured in the questionnaire, and was developed both from the findings of the earlier qualitative work and the MORI poll conducted on behalf of the Human Genetics Commission (Human Genetics Commission, 2001). Specifically, the statements about genetic developments and research on human genetics were directly

lifted from the Human Genetics Commission, and showed broadly similar results as the ones reported there.

Genetic susceptibility testing, however, is a very complex proposition. In a systematic review of the literature of the psychological consequences of predicative genetic testing, it has been suggested that emotional state of people seeking genetic testing should be included into testing protocols (Broadstock et al., 2000). Additionally, there have been arguments that how favourably individuals view genetic susceptibility testing is dependant on how treatable the condition that the test is for (Motulsky, 1999). Some of these, and other issues are further discussed in the final chapter.

Chapter 7 Discussion

7.1 Introduction

This study aimed to examine the health beliefs and risk perceptions of individuals currently unaffected by heart disease who have a sibling who has had a heart attack under the age of 50 years. CHD is a leading cause of premature death in the UK, and family history of CHD is an independent risk factor for CHD, and an important predictor of future disease. Siblings share 50% of their genetic makeup and having a sibling who has experienced a heart attack prematurely increases an individual's risk by up-to tenfold (Samani & Singh, 2001). Previous studies have demonstrated that having a close family member affected with heart disease under the age of 50 years does not necessarily mean people will perceive themselves to be at risk of developing the disease (Hunt et al., 2001). Therefore, examining the risk perception and health beliefs of people currently unaffected by CHD, but who are possibly at higher risk of developing the disease than the general population is important if this group of people are to be targeted for primary prevention interventions as stated in standard four of the National Service Framework (NSF) for CHD. However, neither the NSF nor a recent review about progress towards achieving the standards, mentions the importance of family history (Healthcare Commission, 2005). This is probably because family history is not included in the Framingham equation to calculate CHD risk (Kardia et al., 2003), but there is significant evidence that family history could be utilised as a tool for identifying those at high risk of developing CHD (Yoon et al., 2002, Hunt et al., 2003 McCusker et al., 2004).

Only one study has previously investigated the health beliefs and risk perceptions of people who have a sibling who has CHD (Becker & Levine, 1987). However, there were some fundamental problems with that study; the index case from which the unaffected siblings were recruited included a diverse mix of patients with angina, and those who had had interventional surgery. The interviews were conducted over the telephone, and moreover the study is now nearly 20 years old. In the intervening time, particularly the last few years, considerable advances have been made in genetic technologies. Examples of

this can be found in the announcement of cloning the first human embryo (Vass, 2001), and advances in stem cell research (Braude et al., 2005). The context of the current study, therefore, includes the wide publicity given to rapid development of genetic technologies, and which may provide new mechanisms for prevention and management for a number of complex, multi-factorial conditions such as heart disease and the wide publicity given to this work (Epstein, 2004).

A two-stage design was devised for this study; preliminary qualitative interviews with unaffected siblings of the PRAMIS participants (cases), followed by a postal survey of a larger sample to quantify some of the findings of the earlier qualitative phase. As noted above, the postal survey also included a comparative element with the PRAMIS control group. The study therefore employed a mixed methodology design involving qualitative and quantitative approaches. This chapter will summarise some of the key findings of the study (section 7.2), discuss methodological limitations (7.3), highlight some of the ethical considerations about contacting siblings (7.4), explore possible clinical implications (7.5), evaluate the quality of the qualitative phase (7.6), an assessment of the overall impact of the findings (7.7), make recommendations for future research (7.8) and conclusions (7.9).

7.2 Summary of main findings

The qualitative phase of this study involved 20 semi-structured interviews with unaffected siblings of PRAMIS cases. Data were analysed using the constant comparative method. Four main themes were identified from the analysis: the experience of premature heart attack in a sibling, explanations for siblings' heart attack, inheritance / genetics and participants' experiences with healthcare professionals. The quantitative phase of this study enhanced and built upon the findings of the qualitative phase and involved a postal questionnaire developed specifically for the current study utilising questions or statements from previously published studies, and some items developed solely for this questionnaire. The questionnaire was broadly divided into sections about risk factors, knowledge of heart disease in relation to other conditions, participants' beliefs about their

control of their own health, their beliefs about chances of getting diseases, which disease they feared the most, their beliefs about heath as a value, views about advances in genetic technology and views about future susceptibility testing. Additionally, there was a supplementary questionnaire for the unaffected sibling group only. Chapters 4 and 6 detail the results of the qualitative and quantitative phases of this study respectively. However, to present an overall summary of the findings, a thematic approach for the results will be adopted to allow an integration of the qualitative and quantitative findings.

Many participants in the qualitative phase expressed a strong sense of detachment from their siblings' heart attack, providing defensive rationalisations and explanations to explain their siblings' heart attack. This finding that has not been reported before, and may have implications for how people perceive their personal risk to CHD and therefore how receptive or ready they may be to modifying their lifestyle. High levels of ambivalence to heart disease, particularly amongst men in manual occupations have been observed before (Hunt et al., 2000), but these have not been described as "detachment". In the current study, participants in the qualitative phase often referred to a heart attack in a sibling as something that was of little or no consequence to their own health. However, in the quantitative phase of the study large differences were observed between the two groups. When asked the question "compared with other people of *your* age and sex, what do you think are the chances of you getting the following disease in the next 15 years?", 1 in 20 of the unaffected siblings felt that they had a low chance of getting heart disease, compared to over a third of the comparison group. Conversely, over a third of the unaffected siblings felt that they had a high chance of getting heart disease, compared to only 1 in 20 of the comparison group. This question was developed from previous research, where participants (all women) rated their personal lifetime risk of getting heart disease significantly higher than cancers (Wilcox & Stefanick, 1999). These differences between the findings of the qualitative and quantitative phases are likely to reflect the choice of methodologies. During an in-depth interview a rapport is developed with participants and they may be more likely express their feelings freely.

How CHD was viewed in relation to other conditions, particularly cancer, is an interesting finding from both phases of the study. In the qualitative phase of the study, some participants appeared very complacent or indifferent to heart disease, with most appearing to fear cancer far more than heart disease, whereas cancer was discussed by many participants as a disease that was more frightening than heart disease, and siblings often referred to cancer as being associated with a long, lingering and undignified death. This indifference, particularly in a group of people who are probably at high risk of developing CHD, may have implications for how receptive they might be to preventative strategies or advice. This is an interesting contribution, but similar findings have been described before (Emslie et al., 2001). Similarly, in the quantitative phase, when participants were asked to indicate which disease they feared most, nearly half of the unaffected siblings feared cancer most, compared to two-thirds of the comparison group. However, 1 in 5 of the unaffected siblings feared heart disease most, compared to only 1 in 20 of the comparison group. These differences between the two groups were statistically significant. Interestingly, when participants were asked in the questionnaire what they thought their chances of getting cancer were, compared to people of their age and sex, both groups perceived themselves to have a similar risk, with over 2 thirds of both groups indicating that they felt they had an average risk of developing cancer. Highly significant differences were found between the two groups regarding their perceived chance of having a stroke. These results were statistically significant ($p=0.001$,adjusted for gender and occupational class).

Beliefs about the gendered nature of CHD were evident in both phases of this study. In the qualitative phase, participants frequently talked of a heart attack in a male relative being "massive", but as being "merely a heart attack" when discussing a heart attack in a female relative. Beliefs about gender differences in heart disease appear to be quite profound, as this finding is in line with other with (Emslie et al., 2001b). In the quantitative phase, participants were asked to respond to the following statement "a man over 65

years is much more likely to die from heart disease than women over age 65". This statement is not true, but has been previously utilised in other research as a means of assessing knowledge about gender and heart disease (Wilcox & Stefanick, 1999). In the quantitative phase of the current study, less than half of the unaffected siblings agreed with the statement, compared to nearly three-quarters of the comparison group. Similarly, a third of the unaffected siblings disagreed as opposed to 1 in 10 of the comparison group. This was statistically significant (p=0.005, adjusted for gender and occupation). This could suggest that the unaffected siblings were better informed about heart disease than the comparison group.

Beliefs about the causes of CHD, and risk factors for heart disease were explored in both phases of the study. In the qualitative phase, participants frequently mentioned stress or fate as explanations for their siblings' heart attack, but often needed prompting to consider family history as being an important risk factor, or appeared to require more than one relative affected with heart disease to acknowledge their personal risk. Stress has been highlighted in previous research as a common explanation for heart attacks (French et al., 2005), as has fatalism (Davison et al., 1989, Frankel et al., 1991, Davison, 1992. Hunt et al., 2000). Additionally, there is prior evidence that interpretation of family history of CHD is often ambiguous (Watt et al., 2000, Hunt et al., 2001), but it is important to note that these studies were not undertaken with siblings of people who had experienced a heart attack. Therefore, it might be difficult to draw conclusions from this previous work that is relevant to unaffected siblings.

Beliefs about risk factors were incorporated into the questionnaire for the quantitative phase. For the majority of risk factors broadly similar results found between the two groups, with the somewhat minor exception of stress or worry. Nearly all of the unaffected siblings agreed that stress or worry increased the risk of heart disease, as did over three-quarters of the comparison group. Furthermore, lack of exercise was ranked in the top 3 risk factors by only a very small number of the unaffected siblings, but by a fifth of the

comparison group. Considering how much publicity is dedicated to highlighting the potential preventative benefits of exercise, this is very surprising and may indicate how difficult promoting exercise may be to this particular *high-risk* group. If the mean scores for risk factors from the current study are compared to the source of these questions (Weinman et al., 2000), some interesting differences are found. For example, while the scores for stress or worry were higher in this study than in the Weinman study, participants in this study were also asked to rank the risk factors, and stress was only ranked in the top 3 most important risk factors by a third of unaffected siblings and a fifth of the comparison group. In the Weinman study, the mean ratings for risk factors placed stress as the most important risk factor, which may indicate that participants in this study were more informed.

Some participants in the qualitative phase expressed a desire for a formal support service where they could access information, lifestyle advice and an assessment of their individual risk following their siblings' heart attack. It appeared that some did not feel that they receive this sort of service from their GP. Moreover, the supplementary questionnaire, which was only sent to the unaffected sibling group, asked participants to respond to the following statement: "I needed to speak to a health professional in the months following my brother or sister's heart attack", with a third agreeing, while a fifth disagreed. Perhaps this highlights the need to offer unaffected siblings the chance to be incorporated into cardiac rehabilitation programmes as a primary prevention strategy. There is clear evidence of the benefit of cardiac rehabilitation to partners of heart attack patients has been demonstrated (Thompson, 1989), but extending this formal support and inclusion in cardiac rehabilitation for genetically related individuals could be explored. Perhaps now that home-based cardiac rehabilitation programmes are becoming more popular (Dalal et al., 2004), this would be particularly relevant to explore.

A finding of the qualitative phase of the study that was of particular concern were the experiences of participants with primary health care services if they sought advice

following their siblings' heart attack. Some suggested that their concerns were trivialised or brushed aside by their GP or practice nurse. This is a worrying finding and it may have important clinical implications, as it is the responsibility of GPs and Primary Care Trusts (PCTs) to identify individuals at substantial risk of developing CHD (Department of Health, 2000). This finding would suggest that this aim is not being addressed when people seek help following their sibling's heart attack. Siblings of people who have had a heart attack are known to be a largely neglected group for primary prevention purposes (Hengstenberg et al., 2001), but family history could be used to identify a small group of people at very high risk of developing CHD (Yoon et al., 2002, Kardia et al, 2003, McCusker et al., 2004). This perhaps demonstrates an educational need for primary care staff, especially in relation to interpreting risk factor information (Watson et al., 1999, Bankhead et al., 2001). In the quantitative phase of the study, unaffected siblings were asked if they had discussed any concerns they may have following their siblings' heart attack with a health care professional. Only 18 (32%) reported that they had visited a healthcare professional following their sibling's heart attack, but all felt that the consultation had been helpful. Ideally, more of the unaffected siblings would have sought help or advice following their siblings' event from a health care professional, but some reported in the qualitative phase that they felt inhibited from visiting their GP because of previous bad experiences.

The sources that people utilised to access information were also explored. In the qualitative phase, some participants suggested possible areas where health-related information could be accessed, including the television, public libraries, visitors areas in hospitals and places such as Citizens Advice for people who are not registered with a GP, or do not have access to the internet. This was further explored in the supplementary questionnaire where unaffected siblings were asked to identify if they had sought information about inherited aspects of heart disease. A variety of sources of health information were utilised by unaffected siblings, such as British Heart Foundation literature, books and the internet. Surprisingly, nearly a fifth of the unaffected siblings indicated that they did not know where to access information and only 1 in 20 went to

another health care professional, which included the practice nurse and pharmacist. Additionally, when asked if adequate services were available for understanding inherited aspects of heart disease nearly a half agreed and a third disagreed. This would seem to suggest that more information should be routinely made available to unaffected siblings, particularly as only a third had sought any help following their siblings' heart attack.

One of the key aims of the current study was to identify beliefs about genetic research and the possibility of genetic susceptibility testing for heart disease in the future. In the qualitative phase, views about hypothetical genetic susceptibility testing for heart disease were explored, and the majority of participants appeared to view this positively. However, there were some indications of gender differences in the readiness to accept advances in molecular technologies. All female participants (10) expressed a positive view about future genetic testing, but 3 (of the 10) men interviewed were highly resistant to the possibility of this in the future. Although this finding may be explained by the relatively small number of participants in the qualitative phase, the hostility that they expressed was quite marked, especially when compared to female participants. Admittedly, however, the majority of men were willing to undertake a genetic test (if available), but it was nevertheless only men who were resistant to the idea of such a test. In the questionnaire, participants were asked to a score on a 5–point Likert scale their responses to the following statement: "if a genetic test were available that could indicate that I was at increased risk of developing [heart disease, stroke or cancer], I would want to have such a test". Regarding a test for heart disease, over three-quarters of the unaffected siblings and the comparison group agreed that they would want to have a test. However, only 1 in 10 of the unaffected siblings and the comparison group indicated that they would not want to have such a test. For stroke, similar results were observed as the heart disease question. The same question, but for cancer, demonstrated broadly similar results to heart disease and stroke, with slightly over a half of the unaffected siblings and slightly less than half of the comparison group agreeing. Because some gender differences were observed in the qualitative phase of the study regarding genetic susceptibility testing, a sub-analysis of

these data by gender was undertaken. There was little evidence in the quantitative phase of any gender differences with the results being broadly similar to the main results. However, the questionnaire may not have been sensitive enough to measure extreme levels of hostility, which were observed in the qualitative phase.

Nevertheless the finding of the qualitative phase of the study, where some men were very resistant to the possibility of genetic susceptibility testing requires further quantitative investigation. This is potentially very important and, if confirmed, could have important implications for health services, and perhaps illustrates the need for more efforts to promote a greater understanding about the role of genetics amongst the general public. Based on the findings of a systematic review on the literature of the psychological consequences of predicative genetic testing, an assessment of the psychological state of people seeking genetic testing should be included in pre- testing protocols (Broadstock et al., 2000). Additionally, it has been argued that how favourably individuals view genetic susceptibility testing is dependent on how treatable the condition is, that the test is for (Motulsky, 1999). Indeed, the unaffected siblings of people who have experienced a heart attack are likely to have conventional as well as genetic risk factors (Hengstenberg et al., 2001), and the former may be treatable with existing therapies, for example high blood pressure, high cholesterol, or even as an absolute minimum, to be offered aspirin.

On the whole, participants in the qualitative phase expressed considerable interest in genetic research, and genetic information was viewed as being unique and something that sets an individual apart from others. Conditions with a genetic aetiology were often discussed in the context of being serious or sinister conditions. Therefore, the attitudes of participants to genetic developments were investigated using items developed from the findings of the qualitative phase and from a MORI poll conducted on behalf of the Human Genetics Commission (HGC) (Human Genetics Commission, 2001). The results of the current study are broadly in line with those of the HGC. For example, when asked to respond to the statement "new genetic developments will bring cures for many diseases",

approximately three quarters of participants in the HGC study agreed, as did a similar amount of unaffected siblings and the comparison group in the current study. Furthermore, when asked to respond to the statement "genetic research will lead to improvements in the health of future generations" the vast majority of both unaffected siblings and the comparison group agreed to the statement. This therefore demonstrates a very positive view to genetic research and development amongst this sample.

Health-related behaviour is clearly an important part of preventative health and self-management. Several participants in the qualitative phase of the study reported that they had modified their lifestyle following their siblings' heart attack, for example changing their dietary or exercise habits, although this certainly did not include all of the participants. Nearly a fifth of unaffected siblings smoked, compared to only 1 in 10 of the comparison group. Of those who did smoke, more than half of both groups smoked more than 11 cigarettes per day. The change of smoking habits, (assessed in the supplementary questionnaire), indicated that of the 11 unaffected siblings who currently smoked, over half smoked about the same amount of cigarettes now, and the remainder smoked less now. However, this does not measure those individuals who stopped smoking following their siblings' heart attack, and it only applies to the 11 unaffected siblings who currently smoke. Of these individuals, the majority had not changed their smoking habits since their siblings' heart attack. This seems rather disappointing and probably indicates the need for a larger more specific study to assess this fully.

7.3 Methodological limitations

One of the key limitations for this study concerns the sampling. The sample source for the PRAMIS study was limited to white Europeans. Therefore all members of ethnic minorities were excluded from this study, so the results cannot be generalised to ethnic minority groups. In addition, as little funding was available for travel costs, participants for the qualitative phase were primarily selected for the characteristics of age and gender and living within travelling distance of Leicester. This limited the extent to which occupational

criteria could be used for quota sampling and the majority of participants in the qualitative phase had manual occupations.

The quantitative phase of the study was also subject to some additional methodological limitations, which require further discussion. This phase of the study involved a postal questionnaire, copies of which were sent to the unaffected siblings of the PRAMIS cases and to the PRAMIS controls as the comparison group. Direct access was possible for posting the questionnaire to the former group, as their details were all available on the PRAMIS database, and a very high response rate (71%) was achieved. Furthermore, the PRAMIS controls had already participated in previous research and were possibly more likely to participate in subsequent research. By contrast, however, to reach the unaffected siblings of the PRAMIS cases, all research materials had to be sent via the PRAMIS cases, who then forwarded the questionnaire to one of their siblings. This proved to be an elaborate and convoluted process and led to a relatively disappointing response rate for the unaffected sibling group (32%), despite every effort being made to maximise response, in line with recommendations of Edwards et al. (Edwards et al., 2002). The only previous study of unaffected siblings, had a response rate of 91% for contacting them, although this was all by telephone, and it is unclear if the index contact individual (with the heart attack) was involved in the approach to participants (Becker & Levine, 1987). It is unlikely that this process would gain ethical approval now.

This method of contacting siblings led to large differences between the unaffected sibling and comparison groups in terms of number of participants, but also to surprising gender differences between the two groups. In the unaffected sibling group, 61% of participants were female, whereas in the comparison group 15% were female, a highly significant difference. Although problematic for interpreting the results, this is nevertheless a very interesting phenomenon. In the PRAMIS study there were equal numbers of females (approximately 15%) for the case and control groups. Examination of data for the current study in conjunction with some PRAMIS data reveals that of the unaffected siblings who

participated in this study, 13% of their affected siblings (that is to say the PRAMIS cases) were female. On further analysis of the combined data (this study and some PRAMIS data) it is apparent that the PRAMIS cases had predominantly sent the research materials to a female sibling. Sixty percent of male PRAMIS cases sent the research materials to a female sibling, while all PRAMIS females sent the materials to a female sibling. This methodological problem was not anticipated during the design and planning of the study. In hindsight, it would have been possible to request PRAMIS cases forward the research material to a sibling of the same gender, but this may have reduced the response rate and sample size even further. In retrospect, some imbalance should have been anticipated with the design of the current study. This is because the sample for the current study were the unaffected siblings of the PRAMIS participants (cases), and it is conceivable that (with a sample source of 15% female) if all PRAMIS participants had only one sibling, 50% of them would be expected to be female. Therefore, this imbalance should perhaps have been anticipated and prevented. Ideally a new sample would have been recruited for the current study, which would have prevented many of these problems. Utilising a previous study as a sample source inevitably leads to limitations since the restrictions, bias and limitations from the previous research will also affect a new study. However, recruiting a new sample would have had serious resource implications for this study.

Thus, in the quantitative phase of the study, there were significant differences in the base-line characteristics of the two groups, especially in relation to gender and occupational class. To control for the possibility that the findings observed in the quantitative phase of the study were not due to these potential confounding factors (demographic differences between the two groups), regression analysis was undertaken (linear regression for ordinal data, logistic regression for binary data and log-linear regression for the question about fear of disease). The results therefore are adjusted for the differences in base-line characteristics. For the most part, however, very little difference was found between the original and re-calculated p-value.

There are several ways in which this study could have been improved. For example, in the qualitative phase, participants from a broader spread of occupational groups would have helped to maximise the transferability of the findings. Some of the findings from the quantitative phase of the study appear to contradict findings in the qualitative phase of the study. For example, during the qualitative phase, some participants requested quite strongly, a structured programme of support, which was not necessarily observed in the questionnaire. This could be for a number of reasons, for example the participants in the qualitative phase being mostly from manual occupations, or the sample size in the quantitative phase may well have been too small to detect such differences. Perhaps more likely, is that these apparent discrepancies are a reflection on the methodological approaches used. Undertaking an in-depth interview in a participant's own home enables a rapport between the researcher and the participant to develop, which allows the exploration of ideas to a depth that would not be possible with a questionnaire. Moreover, some of the questionnaire items may not be sensitive to the strength of participants' beliefs, for example about genetic testing, which can be explored and probed during interviews. A previous study that adopted a mixed methodological approach also found some apparently contradictory findings. This investigated the quality of life of patients undergoing cancer drug trials (Cox, 2003) using qualitative interviews in conjunction with previously validated quantitative tools. Some profound difference were found in the results of this study, for example the questionnaires indicated that participating in the trial had minimal impact on the participant, whereas, the interviews indicated that participating in the trial often had dramatic effects on the individuals (Cox, 2003). Although the topic of the Cox study is very different to that of the current study, this is nevertheless a useful illustration of an important methodological issue, which needs to be considered when interpreting results. One possible reason for the differences observed in the two phases of this study could be explained by the different philosophical underpinnings of qualitative and quantitative research (Constructivism and Positivism respectively). Although it is now acknowledged that combining research methods can be helpful (Bryman, 1988, Bryman, 2005), these two methods do nevertheless have different assumptions.

Additionally, a sub section of unaffected siblings were participants in both phases of the study, which could have led to research fatigue. However, because of the relatively small sample source it was necessary to attempt to recruit as many of the unaffected siblings as possible. Excluding participants from the qualitative phase to the quantitative phase would have reduced the sample further.

Lastly, an editing error with the questionnaire was noted after they had been posted to participants; this is a typographical error on the supplementary questionnaire resulting in the question numbers being out of sequence (question numbers 22 and 23 are missing).

7.4 An evaluation of quality in the qualitative phase

A recent report on evaluating quality in qualitative research has provided a framework for assessing quality, which includes the following criteria: defensibility of approach, rigour of conduct, the relationship of the researcher to the researched (reflexivity), credibility of claims and the broader impact of the study (Spencer et al., 2004). The current study clearly fulfilled many of these criteria. For example, there were clear research questions (what are the risk perceptions, health beliefs and views about genetic developments amongst people whose siblings have experienced a premature heart attack?); there was a clear and logical line of inquiry, and a choice of method (semi-structured interviews) that was "fit for purpose". Sampling was limited because of the choice of the sample source (PRAMIS participants) and the necessity to interview participants at home that were geographically accessible for this un-funded study. The study was conducted with rigour, demonstrated by careful recording of data, systematic analysis using the constant comparison method, facilitating in-depth investigation, which was initially undertaken in a group setting with supervisors. To some extent the claims of the qualitative phase are supported by an element of respondent validation as the interview prompt guide was developed during the data collection to reflect findings from early analysis. The

development of categories and themes can be clearly seen from analysis of the emerging data and a balanced selection of data is presented.

The broader impact of the study is detailed below, but reflexivity requires some discussion. Spencer et al, define reflexivity as "showing awareness of the impact of the researcher on the researched" (Spencer et al., 2004, page 71). The researcher has a clinical background, from the nursing profession with experience in cardiology, acute medicine, community nursing and research (having worked on the British Heart Foundation Family Heart Study). There is a possibility that this previous experience could have influenced the values and assumptions with which the researcher approached this study. If at all, this is perhaps likely to have made the researcher sensitive to clinical implications of this work. Considering the rigour with which the study was undertaken, and the quality assurance mechanisms incorporated (analysis initially undertaken with continual advice and discussion from supervisors), it seems unlikely that a different researcher undertaking this study would have produced drastically different findings.

However, since no new theory was developed from the qualitative phase of the study, it is questionable whether Grounded Theory is the most accurate description of the method undertaken for this study.

7.5 Ethical considerations about contacting siblings

This study raised some important ethical considerations. These concern both the recruitment process for the unaffected siblings, and the nature of the research topic. In each phase of the study, unaffected siblings of PRAMIS cases were accessed via their sibling who had participated in PRAMIS. While the Local Research Ethics Committee approved this method of approaching participants, it is nevertheless worth considering the ethical issues that this raises.

For the qualitative phase of the study the approach to unaffected siblings was relatively trouble free as the research fellow on PRAMIS was still working in the same department as the researcher. He was able to facilitate access to these individuals as he was in contact with many of them to complete the PRAMIS data collection. However, there was a delay of two years before material for the postal questionnaire was sent out to PRAMIS participants, which resulted in deceased PRAMIS participants being contacted. In hindsight, a further step to check the existing registration of the PRAMIS case with their GP should have been included in the design of this study from the outset. This would have added another stage to the recruitment process and further diminished response rates, and might have had cost implications, (as some GPs may have required payment for providing this information). However it would have helped to prevent questionnaires being sent to deceased patients and the distress this is likely to cause.

A lesson from this study regarding the ethical implications of contacting individuals that had participated in previous research, especially people who have an established disease process would be to ensure that the dataset for the primary study is up-to-date, particularly if there is a time delay. As many factors influence the timing of studies, including funding, research ethics applications and so on, maintaining the original dataset is vitally important. One method of maintaining a more up-to-date database would be to *track* participants on the original study via the Office for National Statistics (ONS), though there is a financial cost, both in terms of registering participants with the ONS and the administrative work associated with maintaining the database. Specific written permission on the consent form of the original study would also be required for this and there can be a three-month delay from the time of death to the ONS report. ONS tracking therefore would reduce the number of potential participants that would require written confirmation from their GP, rather than replace the process altogether, as ONS reports would only identify those individuals who had deceased more than 3 months previously. Therefore written confirmation would still be required from a GP in most cases.

There is another ethical dimension to this work, which relates to the possibility that exploring an individual's perception of their risk status in relation to their siblings' heart attack could exacerbate concerns or even raise concerns and anxieties that were not there before. Following the interviews in the qualitative phase, participants were asked if they had any concerns, or if any of the topics discussed had raised any specific worries. A telephone contact number was left with all participants in the qualitative phase, which was also reproduced in the study information sheet. None of the participants in the qualitative phase contacted the researcher, but this facility was available.

7.6 Clinical implications

This study raises some important clinical considerations particularly about identifying people at risk of developing heart disease and the possible future use of genetic technologies in clinical settings. As highlighted previously, standard 4 of the NSF for CHD requires GPs and PCTs to identify people at significant risk of developing the disease, but who are currently unaffected. Since siblings share 50% of their genetic material, and CHD is known to have such a strong genetic component, unaffected siblings of people who have experienced a heart attack are a group who should be identified and have their risk of developing CHD assessed. Hengstenberg et al. have argued that unaffected siblings have been largely ignored by primary prevention strategies, but are a readily identifiable group, who frequently have more than one modifiable risk factor (for example high cholesterol or high blood pressure) and would benefit from preventative measures, either educational, or targeting for specific therapies (Hengstenberg et al., 2001). Perhaps an appropriate means of doing this would be to systematically approach people who were known to have had a heart attack and enquire about their other family members (Yoon et al., 2002). Logistical problems would also need to be addressed if such an identification process were to be implemented, as unaffected individuals may be living at some distance from their siblings and would therefore be unlikely to be registered with the same GP. Busy GPs may be unable to spend time identifying people at risk who are not registered with their practice. With Practice Based Commissioning (Department of Health, 2004),

Payment by Results and the General Medical Services contract (GMS2), GPs already appear to be stretched. There may also be ethical difficulties associated with this proposition. Some of the participants in the current study indicated a strong sense of detachment from their siblings' heart attack and may not, therefore, be prepared to acknowledge their elevated risk status, and steps which identify them as being *at risk* may be harmful to their emotional well-being. Additionally, the process of contacting siblings (and identifying who would be the most appropriate person / people to do this) would need to be considered. This should probably include the index individual (the person who had experienced the heart attack) and relevant health care professionals, to ensure that data protection standards and guardianship of information is maintained.

A focused mechanism of identifying people at high risk of developing heart disease would help partially achieve standard 4 of the NSF for CHD. However, a recent report by the Healthcare Commission on progress towards achieving the national standards of the NSF makes no mention of family history (nor indeed does the NSF) and highlighted that the majority of activities addressing this standard of the NSF consisted of general schemes to reduce smoking and promote exercise, or healthy eating (Healthcare Commission, 2005). Undoubtedly these are important measures, but will not help to identify groups who are at high-risk by virtue of family history or provide interventions tailored to the particular needs of these groups. Specifically, unaffected siblings could have their modifiable risk factors assessed and treated as appropriately, for example, perhaps even having cholesterol lowering therapy at a lower threshold than would normally be prescribed. Prophylactic aspirin, unless there were any known contra-indications, could also be prescribed as regular low dose in the light of evidence demonstrating that aspirin is beneficial in primary prevention of CHD (Hayden et al., 2002, Elwood et al., 2005).

A particularly worrying finding of the study were the experiences of the participants in the qualitative phase of the study, who reported that when they visited their GP for an assessment of their personal risk or reassurance regarding their own health they were

dismissed or trivialised by their GP or practice nurse. This is unacceptable practice, but it also represents a missed opportunity in primary prevention, particularly considering the importance placed on primary prevention in the NSF. However it must be borne in mind that in the quantitative phase of the study, only 18 of the unaffected siblings (32%) visited their GP following their siblings' heart attack, and all reported the consultation to be helpful. As discussed in the section on methodological limitations, there are other examples of apparent contradictions between the two phases of the study. The most likely explanation in this case is methodological; developing a rapport with a participant during an in-depth interview where their feelings and experiences can be discussed, prompted by a researcher, is likely to generate different reports compared to completing a postal questionnaire. Nevertheless, this finding from the qualitative phase would seem to warrant further investigation.

Those participants who did not seek help from a health care professional following their siblings' heart attack may be experiencing the feeling of detachment that many participants in qualitative phase alluded to. For some participants, there appeared to be little or no consequence for their interpretation of their own risk status, and some mentioned that they were totally different people to their sibling and therefore felt unlikely to experience the same health problems that their siblings had. This is possibly an explanation for the finding that 68% of the unaffected siblings did not consult their GP. Additionally, some participants in the qualitative phase indicated that their prior experiences with GPs and other primary care staff were influential in their consulting behaviours, often in a negative manner. Whatever the reason, it would be hoped that more unaffected siblings could seek help following a heart attack in a sibling. This also raises the issue of the training needs of primary care staff in relation to interpreting family history for multi-factorial conditions. (Fry et al., 1999). The current study would suggest that this is certainly the case, with potential topics including the importance of family history, mechanisms for identifying individuals at substantial risk of CHD by virtue of their family history, and possibly the impact of genetic developments for the future.

Furthermore, there is also the need to consider policy implications for health services, particularly in relation to delivering genetic advice/ screening for common multi-factorial conditions, which will be possible in the near to intermediate future. Perhaps there is a future role for Nurse Consultants to be developed in primary care to undertake this role.

7.7 Overall impact

The broader impact and contribution that a study makes is the final component of assessment of quality in qualitative research (Spencer et al., 2004), and it is important to consider the overall impact that this study has / will make. So, what new theories has this work generated? What contribution will be made to the body of knowledge, and what will be the overall impact? The qualitative phase of the study was based within a grounded theory principle, (although utilising the constant comparison method), which aims to generate theories and concepts that are "grounded" in the emerging data (Pidgeon, 1998). Although no new theories have been developed from this study, there are several original contributions that this work makes. For example, the finding that unaffected siblings experience feelings of detachment from their siblings' heart attack has not been described before; nor have the experiences of these individuals when they sought help or reassurance from health care professionals specifically following their siblings' heart attack. The apparent reluctance of a small number of men to consider genetic susceptibility testing in the future may have important clinical consequences in the future. Therefore, while it cannot be claimed that theoretical advancement has taken place with this study, there are some important findings that may have an impact on future clinical practice. Perhaps the most significant of these are the experiences of people seeking help from health professionals and the views of genetic susceptibility testing. Some of the qualitative findings have been presented at local and national conferences (appendices 30 –32, 34).

Since this is a mixed-methodology study, incorporating qualitative and quantitative findings, it is also necessary to consider the impact and contribution that the quantitative phase has made. By virtue of the mixed-methodology approach, the key aspect of the questionnaire was that the topics for inclusion had been guided by the qualitative interviews of the unaffected siblings. This is an important strength and contribution of this study, because tools validated on another sample population might have provided findings that were not relevant to the sample in this study. The questionnaire enabled the quantification of some of the findings of the qualitative interviews and also comparison with a group of people where there is no family history of CHD. Although some of the quantitative findings appear to contradict certain aspects of the qualitative findings, this is probably methodological and has been reported before (Cox, 2003). Importantly, some of the main findings of the questionnaire were statistically significant. For example, unaffected siblings were more likely to believe that they had a higher chance of developing CHD than the comparison group ($p=0.001$), and 20% of unaffected siblings feared heart disease most, compared to only 6% of the comparison group ($p=0.001$). Findings from the quantitative phase have been presented at a local conference (appendices 33-34), where an award was won for Best Conference Presentation (appendix 35).

7.8 Recommendations for future research

This study has raised a number of important issues that could be addressed by further research. The following points detail these issues:

- What are the views of primary care professionals and policy makers about family history of CHD? The experiences of some participants in the qualitative phase indicated that when they sought help or reassurance from health professionals, they often felt that their concerns were dismissed or trivialised. To investigate the beliefs of primary care staff regarding family history of CHD would be a natural next step and enable this topic to be investigated from a service perspective. This topic would seem suitable for qualitative research.

- Is there a practical and ethical method of identifying people at risk by accessing individuals affected by CHD? For example, rather than a healthcare professional enquiring of an individual what their family history is, perhaps there is scope to start with the affected person and work from them to identify siblings or off-spring who may be at elevated risk because of their family history. This would have potentially very serious ethical implications, and would have to be very carefully planned and piloted.

- How could unaffected siblings be incorporated into a clinical support service? In the qualitative phase, some participants expressed a desire for a formal support service, which would be essential if routine identification methods were to be implemented.

- What are the training needs for primary care staff, regarding family history of CHD, and advances in genetic technology? The experiences of some participants in the qualitative phase indicated that current knowledge about risk factor status for CHD is not being used effectively in primary prevention. Furthermore, as genetic technologies develop, it is likely that some sort of genetic susceptibility testing might be developed for CHD and other common multi-factorial conditions. Therefore, assessing the training needs of primary care staff, would be an extremely useful study to complete in preparation for these developments

- A feasibility and pilot study could be considered for the role of Nurse Consultant in the genetics of common multi-factorial conditions. Ideally this post-holder in this role would work in both primary and secondary care.

Furthermore, an interventional trial could be considered with unaffected siblings of first-time heart attack patients, who could receive lifestyle advice, education and perhaps

certain drug therapies, for example aspirin and cholesterol lowering drugs. This could be compared to a non-interventional group who could be monitored. This might provide valuable evidence for judging the effectiveness of identifying and supporting at risk siblings and assess the feasibility of such a service.

7.9 Conclusions

This study investigated the risk perceptions, health beliefs and views of genetic developments amongst unaffected siblings of people who have experienced a heart attack at a young age. Prior to this study, there was only one previous study in this area (Becker & Levine, 1987), which is now nearly 20 years old, and pre-dates many of the genetic developments that form the contextual backdrop to the current study. This is an important topic of research because CHD is a leading cause of death in the UK and family history of CHD is an important risk factor; having a sibling affected with a heart attack under the age of fifty increases an individual's risk substantially and genetic technology may lead to genetic susceptibility testing for CHD in the future. The current study was undertaken as a two-phase mixed methodology study. Preliminary qualitative interviews with unaffected siblings of people who have experienced a heart attack, followed by postal survey to quantify some of the findings of the qualitative phase, amongst a larger group of unaffected siblings and a comparison group where there was no family history of CHD. This integrated qualitative and quantitative methodologies to improve understanding of the subject.

Participants in the qualitative phase expressed a strong sense of detachment from their siblings' heart attack and often talked about heart disease in a very complacent manner. A small number of the men that were interviewed appeared very resistant to the idea of genetic susceptibility testing in the future. Of particular concern, were the experiences of participants when they visited health professionals for advice following their siblings' heart attack and many felt that their concerns were dismissed or trivialised. In the quantitative phase, significantly more of the unaffected siblings felt that they had a higher risk of

developing CHD than the comparison group, but nevertheless nearly a half of unaffected siblings feared cancer more than any other disease.

This study has raised a number of important concerns and questions, particularly about the identification and clinical management of unaffected siblings, and the impact that advances in genetic technology may have. All of these issues merit further investigation to help improve the care of this group of people who are at very high risk of developing CHD, but are largely ignored at the moment.

Appendix 1

Qualitative phase
Study Protocol

Study Protocol:

Health perceptions of people who have a sibling who has experienced a heart attack

Investigators: Mr. Julian Stribling, Dr. Bridget Young, Mr. Paul Lambert, Professor NJ. Samani.

Objectives

- To describe the risk perceptions of individuals currently unaffected by CHD, who have a sibling who has experienced a heart attack, before the age of 50.
- To describe the health beliefs of individuals currently unaffected by CHD, who have a sibling who has experienced a heart attack, before the age of 50.
- To inform the design of a future postal survey to quantify the risk perceptions and health-related behaviour of people currently unaffected by CHD, who have a sibling who has experienced a heart attack.

Background

Coronary Heart Disease (CHD) is a leading cause of morbidity and mortality in the western world. In England alone, some 110,000 annual deaths are attributable to CHD, and this is one of the key areas of Government health policy (Department of Health, 2000). CHD has a complicated and multi-factorial aetiology (Tunstall-Pedoe et al., 1997), and whilst conventional risk factors (smoking, hypercholesteraemia, hypertension and diabetes mellitus) can certainly account for a large proportion of events, it has been estimated that approximately 50% of "risk" remains unexplained (Nora, 1983).

Genetic predisposition is rapidly emerging to account for much of the unknown risk of CHD (Marian, 1999). Family history (or familial aggregation) of CHD is an "independent" risk factor for CHD (Hopkins et al., 1988), and an important predictor for premature disease (Eaton et al., 1996). Much of the criticism of early studies into familial clustering of CHD argued that a "shared"

adverse environment between family members could be responsible, but adoption studies (Sorenson et al., 1988) and twin studies (Marenberg et al., 1994), have seriously undermined these criticisms. Having a first-degree relative affected by CHD under the age of 65 increases the individual's risk by at least twofold, and a maternal history of MI (myocardial infarction) under the age of 50 increases the risk tenfold (Samani & Singh, 2000). The rapidly advancing area of molecular biology and the Human Genome Project may revolutionise medicine (Collins, 1999) by providing clear indications of how individuals can modify their risks through behavioural change, but whether the health-related behaviour of individuals will be affected positively is unclear (Marteau & Senior, 1997).

Among the general public, lay beliefs or perceptions about the aetiology and causation of CHD (Davison, et al., 1991), may emphasise notions of "coronary candidacy" and "fatalism", which individuals may feel absolve them from personal responsibility for their own health and encourage them to believe that their own behaviour is not relevant to their risks of developing CHD. Defensive biases in health-related judgements, particularly in relation to information about risk factor status have also been observed (Croyle, et al., 1997). The extent to which such beliefs and judgements are shared by those with a family history of CHD is unknown. There is some evidence that individuals who currently do not have CHD seem to have a poor understanding of their own relative risk and appear resistant to behavioural change even when their siblings (Becker & Levine, 1987), or mother is affected (Allen & Blumenthal, 1998). For example, individuals may persist in smoking even when they perceive that they have a family history of CHD (Hunt, et al., 2000). A much better understanding is needed of perceptions of risk, among those with a higher risk of CHD by virtue of family history, and how these perceptions affect behaviour.

Setting

Among the current genetic studies based at Glenfield Hospital and the University Division of Cardiology is PRAMIS (**P**latelet **R**eactivity **A**nd polymorphisms in **M**yocardial **I**nfarction **S**tudy) a case-control association

study of people (under the age of 50) with premature myocardial infarctions. Having access to the participants of this study, and their unaffected siblings, places the Division of Cardiology in an ideal position (in collaboration with the Department of Epidemiology and Public Health) to investigate the perception of risk of CHD and health-related behaviour of unaffected individuals whose siblings have experienced a heart attack.

Design

A qualitative approach will be adopted, involving semi-structured interviews, within a grounded theory framework. Qualitative work is particularly useful as a precursor to quantitative research (Murphy et al 1998), especially in helping to define most pertinent issues to investigate, and suggesting the most appropriate ways of phrasing questions. Originally developed in the social sciences, grounded theory is now widely recognised as being especially useful in health services research (Smith & Biley, 1997; Cutcliffe, 2000; Pidgeon, 1996) as a means of accessing the lay beliefs that underlie health-related behaviour.

The interview sample will attempt to represent the diversity of social class and familial history of CHD, to broadly reflect the population from which the sample will be drawn. Approximately 15 participants will be selected using quota sampling. Interviews will investigate participants beliefs' about: i) familial risk and susceptibility to CHD and ii) preventative health-related behaviour to mediate this risk. A semi-structured checklist of topics will be used to guide the interviews to ensure the same basic issues are covered, but questions will be open-ended in style to enable previously unanticipated areas of interest to be explored. With permission from participants all interviews will be audio tape-recorded, and these will be transcribed verbatim so that accurate records of interviews are maintained. Data collection and analysis will proceed simultaneously until all key concepts have been identified, no new themes are emerging and theoretical "saturation" is achieved (Smith & Biley, 1997). The resulting data will be analysed using the constant comparison method (Green, 1998) to discover recurring and patterned ways of talking about CHD risk, familial patterns of CHD and the impact of these on health-related behaviour.

Analysis of the interviews and a literature review will generate a series of themes and other issues to be investigated in the survey phase of the study.

It is planned that this qualitative study will inform the design of a future postal survey to quantify the risk perceptions and reported health-related behaviour of unaffected individuals whose siblings have CHD. This planned quantitative study will draw upon the themes identified in the current qualitative study to investigate risk perceptions, health beliefs and psychological well being in a larger sample of participants.

Source of Subjects:

Unaffected individuals who have 1 sibling who has experience a heart attack (i.e. siblings of PRAMIS cases which comprises approximately 200 people).

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Appendix 2

Qualitative phase

Research ethics letter of approval

Leicestershire NHS
Health Authority

Melanie Sursham
Direct Dial 0116 258 8610

18 January 2001

Gwendolen Road
Leicester
LE5 4QF

Tel: 0116 2731173
Fax: 0116 2588577
DX 709470 Leicester 12

Mr J Stribling
Research Nurse
Division of Cardiology
Glenfield Hospital

Dear Mr Stribling

Health Perceptions of People who have a sibling who has experienced a heart attack. Our ref. no. 6154

Further to your application dated 11 December 2000 you will be pleased to know that the Leicestershire Research Ethics Committee at its meeting on 5 January 2001 approved your application to undertake the above mentioned research. The Committee approved the amended information sheet.

The Committee felt that this should be considered a pilot study and which will lead to the development of a questionnaire.

Your attention is drawn to the attached paper which reminds the researcher of information that needs to be observed when ethics committee approval is given.

Yours sincerely

R F Bing
Chairman
Leicestershire Research Ethics Committee

(NB All communications relating to Leicestershire Research Ethics Committee must be sent to Leicestershire Health)

Appendix 3

Qualitative phase

Trust indemnity letter of approval

University Hospitals of Leicester NHS
NHS Trust

DIRECTORATE OF RESEARCH AND DEVELOPMENT

Leicester General Hospital
Gwendolen Road
Leicester
LE5 4PW

Direct Dial: (0116) 258 4109
Fax No: (0116) 258 4226
e-mail: aimee.geary@uhl-tr.nhs.uk

Tel: 0116 2490490
Fax: 0116 2584666
Minicom: 0116 2588188

24 January 2001

Mr J Stribling
Research Nurse
Cardiology
Glenfield Hospital

Dear Mr Stribling

RE: Project Number: 6685 *[Please quote this number in all correspondence]*
Health perceptions of people who have a sibling who has experienced a heart attack.

We have now been notified by the Ethical Committee that this project has been given ethical approval (please see the attached letter from the Ethical Committee).

Since all other aspects of your Glenfield R&D notification are complete, I now have pleasure in confirming full approval of the project on behalf of the University Hospitals of Leicester NHS Trust.

This approval means that you are fully authorised to proceed with the project, using all the resources which you have declared in your notification form.

The project is also now covered by Trust Indemnity, except for those aspects already covered by external indemnity (e.g. ABPI in the case of most drug studies).

We will be requesting annual and final reports on the progress of this project, both on behalf of the Trust and on behalf of the Ethical Committee.

In the meantime, in order to keep our records up to date, could you please notify the Research Office if there are any significant changes to the start or end dates, protocol, funding or costs of the project.

I look forward to the opportunity of reading the published results of your study in due course.

Yours sincerely

Dr Nichola Seare
Research and Development Business Manager

Appendix 4

Qualitative phase

Letter of introduction

[Date]
[Participants name]
[Participants address]

Dear [Name of potential participant]

We are writing to you as your brother / sister recently took part in a heart study at Glenfield Hospital Leicester, and indicated that you may also be willing to help with a related study.

We are carrying out a study which aims to investigate the views and lifestyles of people who have never had any heart problems themselves, but have a brother / sister who has experienced a heart attack under the age of 50. We are writing to invite you to take part in this study. Enclosed is an information leaflet about what would be involved if you agreed to take part in the study. The study will **not** involve any medical examinations, tests or procedures.

We would be most grateful if you are able to help us with this study. When you have thought about it, please return the reply slip in the prepaid envelope enclosed, indicating whether or not you agree to take part. Please feel welcome to speak to any of the investigators listed on the leaflet before you decide whether to participate

With many thanks.
Yours sincerely,

Julian Stribling
Research Nurse

Appendix 5

Qualitative phase
Information sheet

University Hospitals of Leicester NHS
NHS Trust

Glenfield Hospital
Groby Road
Leicester
LE3 9QP

Tel: 0116 2871471
Fax: 0116 2583950
Minicom: 0116 2879852

Health perceptions of people who have a sibling who has experienced a heart attack

Information leaflet for participants
Version 2, December, 2000

We would like to invite you to participate in our study, which is looking at the health perceptions and lifestyles of people who have a brother and / or sister who has had a heart attack.

1. What is the purpose of the study?

We are keen to learn more about the views and lifestyles of people who have never had any heart trouble themselves, but who have a brother and / or a sister who has had a heart attack. In order to understand how families are affected by heart disease, we feel it is very important to seek the views of family members.

2. What will be involved if I take part in the study?

If you agree to take part we would like to interview you. Julian Stribling will carry out the interviews. Julian is a nurse who is working on a number of different studies looking at heart disease. He will arrange the interviews to take place at a time and place convenient for you. The interviews will probably last about 1 hour. With your permission Julian will tape-record the interviews. This is to avoid Julian having to take notes, but if you would prefer not to be recorded just let him know.

The study will **not** involve you in any medical examinations, procedures, tests or treatments.

3. Will information obtained in the study be confidential?

All information you give us will be confidential. Any tapes used during interview will be destroyed afterwards. Your name will not appear anywhere on documents about the study.

With your permission, we will let your GP know that you have participated in our study, but he / she will not be told anything about what you have said.

4. What if I am harmed by the study?

This study, like all medical research is covered for mishaps in the same way, as for patients undergoing treatment in the NHS i.e. compensation is only available if negligence occurs.

5. What happens if I do not wish to participate in this study or wish to withdraw from the study?

If you do not wish to participate in this study, or if you wish to withdraw from the study at any time you may do so without justifying your decision. Your future treatment will not be affected.

6. Where can I get further information about the study?

For further information, please contact Julian Stribling by telephoning: 0116 256 3791. Listed below are the names of everyone else involved in running this study. Any of them would also be delighted to talk to you if there is anything you wish to discuss.

Investigators:

Mr. Julian Stribling,
Research Nurse,
University of Leicester,
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Professor NJ Samani
Professor of Cardiology,
University of Leicester,
Division of Cardiology,
Clinical Sciences Wing,
Glenfield Hospital,
Leicester, LE3 9QP

Appendix 6

Qualitative phase

Reply slip

REPLY SLIP

Health perceptions of people who have a sibling who has experienced a heart attack

Please return this slip even if you do not wish to take part in the study

To: Julian Stribling
Research Nurse
University of Leicester
Division of Cardiology
Glenfield Hospital
Leicester, LE3 9QP

I agree to being contacted regarding possible participation in the above study ☐

If you are happy for us to contact you by phone, please write your phone

number here:..... Best time to contact me:... ... ...

I prefer not to take part in this study and I understand that I will not be contacted again about it ☐

Signed:..... Date:.....

[Name]

[Address]

Appendix 7

Qualitative phase

Participant consent form

University Hospitals of Leicester NHS
NHS Trust

Glenfield Hospital
Groby Road
Leicester
LE3 9QP

Tel: 0116 2871471
Fax: 0116 2583950
Minicom: 0116 2879852

Health perceptions and of people who have a sibling who has experienced a heart attack

Consent form for participants in phase1

Investigators: Mr. Julian Stribling, Dr. Bridget Young, Dr. Paul Lambert, and Professor NJ Samani

This consent should be read in conjunction with the Participant Information Leaflet (Version 2, dated December, 2000).

- I agree to take part in the above study as described in the information leaflet.
- I understand that I may withdraw from the study at any time without justifying my decision and without affecting my normal care and medical management.
- I agree to an interview being audio-taped. I understand that all information will remain confidential, and that tapes will be destroyed at the end of the study.
- I understand that this research is covered for mishaps in the same way, as for patients undergoing treatment in the NHS i.e. compensation is only available if negligence occurs.
- I have read the Information Leaflet for Participants on the above study and have had the opportunity to discuss the details with **Julian Stribling** and ask any questions. The nature and the purpose of the study has been explained to me and I understand what will be required if I take part.

Signature of participant .....

Date .....

(Name in BLOCK LETTERS) .....

I confirm I have explained the nature of the study, as detailed in the Participant Information Leaflet, in terms, which in my judgment are suited to the understanding of the person.

Signature of investigator .....

Date .....

(Name in BLOCK LETTERS) .....

Appendix 8

Qualitative phase

Participant characteristics form

CHD Perceptions Study

Unique identifier _____

Date of interview _____

Gender Male ☐ Female ☐

Age _____

Occupation _____

Marital status Single ☐ ; Married / Co-habiting ☐ ;
Separated / Divorced ☐ ; Widowed ☐

Area of residence _____

First part of postcode _____

Housing type _____

Gender of affected sibling Male ☐ Female ☐

Age of affected sibling _____

Appendix 9

Qualitative phase

Prompt guide

Interview prompt guide: version 3

Experiences of brother / sister's heart problems

General beliefs about the causes of heart problems

Beliefs about causes of brother / sister's heart problems including the role of family history / genetics in causing heart problems

Regularity of contact / perceptions of closeness (including perceived genetic closeness) to brother / sister – if you look and behave like your brother / sister, are you more likely to experience the same illnesses? Why, for example are some family members more likely to have certain conditions?

Beliefs about vulnerability to health / heart problems / relevance of brother / sisters experience to self

The significance of the age of onset of heart problems, and whether this indicates a possible role of genetic factors

Perceptions of control in relation to health / heart problems

Beliefs about the importance of lifestyle in moderating personal vulnerability to heart problems

Impact of brother / sister's heart problems on lifestyle (exercise, smoking, diet etc.)

Relative importance of hereditary / genetics v's behavioural factors in heart disease

Emotional impact of brother / sister's heart problems (worry / anxiety / mood etc).

Access to health information, and its utilisation. Information seeking as a consequence of brother / sister's event

Beliefs / feelings about sibling taking part in a genetic study – positive and negative aspects? Would you like to have had a say about whether they took part? Would you like to know more about the research and its findings?

The role of hereditary / genetics in diseases, specifically heart disease

Any ideas about genetic "risk" verses genetic "resilience"

How possible is it to "insure", or make –up for genetic risk factors by adopting a healthier lifestyle?

What degree of certainty is attached to genetic factors? Is there any degree to which genetic factors are amenable to change?

What benefits does genetic research have for your brother / sister, your family, society in general? What are the drawbacks?

How are genetic tests different from other sorts of tests, e.g. having your blood pressure checked?

Appendix 10

Qualitative phase

Letter to GP

Date

Dear Dr. [GP. Name]
[GP Address 1]
[GP Address 2]
[GP Address 3]
[GP Address 4]

Re: Health perceptions of people who have a sibling who has experienced a heart attack

Study participant: [Title], [Name], [Address]

The above person, who is registered with your practice, has taken part in the above-mentioned study. This is a qualitative study examining the views and lifestyles of unaffected people who have a sibling who has experienced a heart attack. Participation in this study involved an interview. No individual reports or actions will be based on the study's results, and the study will have no effect on the patient's routine management.

If you require more information, please do not hesitate to telephone me on 0116 256 3791.

Kind regards.
Yours sincerely,

Julian Stribling
Research Nurse

Appendix 11

Qualitative phase

Open codes

CHD Perception Study

Points From Transcript 5: 3, 4, 2: 1: 9: 7

1) Shock of heart attack / unexpected / Devastated / Not the typical person you would think would have a... / It came out of the blue / He is the last person that I thought would....
2) Heart attack unexpected in a fit / young person
3) Risk factors:
    a) Smoking
    b) Diet (high fat) Rich foods
    c) Drinking / "Drinking a hell of a lot"
    d) Overweight /obese
    e) Stress
        i. Occupational
        ii. Financial
        iii. Getting irate
    f) Exercise / a slow lifestyle / Not keeping fit / A big part to do with it (T9, p2)
    g) Hereditary
    h) Increasing age
    i) How you eat (time)
    j) Over-indulgence
    k) Cumulative effects of risk factors
    l) "Peri-natal" nutrition

4) Cultural habits re: diet / drinking / Smoking ("It was big to smoke") / Binge drinking / Eating late at night / Take aways.
5) Healthy appearance
6) Age risky lifestyle started
7) "Not been affected so far"
8) Stress in the time leading up-to heart attack / Always gets worked up / stressed easily / Depends how you handle it.
9) Hereditary factors
10) Hereditary factors dwelling on mind / crossed my mind.
11) Influences of media coverage on health and heart problems
12) Family clustering of heart disease / cancers
13) Unaffected individuals in family clusters
14) Views about the idea:
    Closeness = increased likeliness of heart disease
15) "The whole thing goes a bit deeper"

16) Number of relatives affected

17) Ideas about moderation of risk. Motivation:

i) Family history: CHD / Cancer

ii) Socially

iii) Immediate health benefits

iv) Not motivated / not associating risk factors

v) Tests might frighten me to give up smoking / change lifestyle

vi) Going to the gym

vii) Anti-smoking

viii) Parental role model

18) Difficulty sustaining a healthy diet / healthy lifestyle / lack of will-power

19) Bad lifestyle hadn't had a chance to do damage

20) Stoical / stoicism.

21) "Never think it's going to happen to you or close friends"

"It wont bother me, it's the bloke down the road".

"I'm alright Jack"

22) "Reassurance" (linked to 17), "Excuses".

23) Scared / Vulnerability for personal health

24) Avoidance of consulting / Not going for a check-up

i) Other priorities

ii) Inertia

25) Little you can do about hereditary problems

26) Best to look on the bright side – hope that you haven't got hereditary problems

27) Predisposition may be altered, eg.

High blood pressure

High cholesterol

28) Peoples reactions to genetic tests:

"Reassurance

"Not the best thing for them"

Psychological consequences (depression if result +ve)

Financial problems

Pressure on other members of family

How do you live with that information?

29) Choice to moderate predisposed risk. "I think they can reduce the risk if they change their lifestyle dramatically. "You can't use that as an excuse" "If I keep myself fit, I've done my bit"

30) What genetic tests can tell you:

- Predisposition / susceptibility
- Likelihood

- Nasty diseases
- A disease you can't do anything about

31) Genetic tests are qualitatively different from other tests eg. Blood pressure. OR, the opposite. More appropriate, tell you a hell of a lot more

32) Unalterable nature of genes.

33) Genetic problem is a unique problem – sets you apart

Ideas about uniqueness.

34) Fundamental

- Serious
- Building blocks of life
- Root problem
- "It's your being"

35) Consequences of genetic tests

- Insurance (genetic underclass)
- Employers
- Cultural changes? eg. Of insurance companies
- Frightening.
- Ability to exploit knowledge
- Do I change my life now?

36) Sources of health information:

- Internet
- GP
- Library (Public / Professional)
- BHF literature and advert
- TV
- Chemist

37) Trust yourself in the hands of people doing the test

38) Faith / acceptance in science

39) Cross cultural differences in attitudes to medical profession.

40) Too trusting?

41) Finality / certainty of genetic tests.

42) Ideas about: public interest in genetics / genetics research.

43) Complicated (?genetics?)

44) No implications for individual

Just a one-off

Unfortunate

"It didn't make me stop and think"

No effect on my life. (T7, p6)

45) No previous family history

46) Luck of the draw

a) Sibling chosen
b) Randomness
c) Fate
d) Just one of those things
e) "It's down to fate if you're going to have you're going to have one"
f) If it happens it happens

47) Paradox that you cannot change predisposed risk
48) Heart attack changed siblings lifestyle / outlook (enhanced his life) / Gets very depressed / short-tempered / not able to work/ Frightening event. Psychological impact of MI / Heart attack messes your brain / Taught him a lesson.
49) "You can try and prevent it but I don't think you can actually stop it."
50) Balance risks / compensation
51) If you've survived a heart attack, you must have a strong heart.
52) Strength / weakness might be passed on
53) Searching for an explanation (Blame)
54) Ideas about causes of heart attack (linked to point 3)
    1) "It's pressure on your heart"
    2) "Your heart 's giving way under it"
    3) "Your hearts saying that's it for the minute"
    4) "The way we live"
    5) "Over indulgence – that's the biggest problem"
    6) "Sit about about and eat fatty foods, it's just going to rest on your heart" (T9, p2)
55) Ideas about outlook / mental attitude:
    1) "It's how you feel
    2) "If you worry about things too much then things happen." (page 11 tran. 3)
56) Never asked by member of medical profession about family history.
57) Not informing GP of sibling's heart attack. "It didn't occur to me to mention it."
58) Initial grave concern for sibling following heart attack. "It really upset me... I couldn't believe how it did upset me." "Try and see him through it" (T9, p3)
59) Positive views about the prospect of genetic tests.
60) Views about genetic tests being similar to other screening tests.
61) Ideas about health problems of younger siblings in large families – "the runt of the litter" (Mother didn't give her body time to recover)
62) "I associated smoking more with cancer than the heart".
63) Fear of cancer, but not heart disease, wasting diseases
    "Cancer frightens me" "It weighs on my mind"
64) Negative images of health related behaviour – "all I'd seen were me mam with a fag in her mouth". (T9, p2)

65) Suggestions for information:

E.g. TV. Adverts "What to do if someone has a heart attack" (T9, p5)

66) Ideas about quantifying potential benefits ton health in terms of life years.

67) Coronary candidacy – inevitable

68) Implying that males had a greater consequence of their heart attack. "Dad had a major one" (T7, p1)

69) Indirect referral identified. E.g. (T7, p2) – one of the doctors at the general suggested we should go for a check up.

70) Negative experiences of visiting GP after sibs MI. "To be truthful it was almost like he didn't give a toss, and I was asking him something totally pointless" T7, p3

"Made me feel quite small

"Like a hypochondriac"

71) Realisation of ↑ risk after 2 first degree relatives affected with CHD

72) Smoking "blamed" for almost everything by medical profession. "I think they're using smoking as an excuse"

73) Associating smoking with the lungs, not the heart.

74) People scared / frightened of genetics

75) Feelings of vulnerability

76) Ideas about:

a) With heart disease / heart attack you either die or survive, but with cancer death seems the inevitable outcome.

b) No acknowledgement of chronic morbidity associated with CHD.

77) Preference of heart attack as a mode of death (instant) rather than a long lingering death.

Appendix 12

Qualitative phase

Categories

From 30/11/2001

CHD Perception Study: Categories- 2nd Draft

1. Identified Risks

1.1.1 Risk Factors

1.1.2 Smoking (3a)

1.1.2 Diet (high fat) Rich foods (3b)

1.1.3 Drinking / "Drinking a hell of a lot" (3c)

1.1.4 Overweight /obese (3d)

1.1.5 **Stress** (3e)

1.1.5.1 Occupational (i)

1.1.5.2 Financial (ii)

1.1.5.3 Getting irate (iii)

1.1.5.4 Stress in the time leading up-to heart attack (8)

1.1.5.5 Always gets worked up (8)

1.1.5.6 Stressed easily (8)

1.1.5.7 Depends how you handle it (8)

1.1.6 Exercise (3f) a slow lifestyle / Not keeping fit / A big part to do with it (T9,p2)

1.1.7 Hereditary (3g)

1.1.8 Increasing age (3h)

1.1.9 How you eat (time) (3i)

1.1.10 Over-indulgence (3j)

1.1.11 Cumulative effects of risk factors (3k)

1.1.12 "Peri-natal" nutrition (3l)

1.2 **Cultural habits**

1.2.1 Diet

1.2.2 Drinking / Binge drinking (4)

1.2.3 Smoking ("It was big to smoke") (4)

1.2.4 Eating late at night / Take- aways' (4)

1.3 Age risky lifestyle started (6)

1.4 Bad lifestyle hadn't had a chance to do damage (19)

1.5 Ideas about causes of heart attack (54)

1.6 "It's pressure on your heart" (54i)

1.7 "Your heart 's giving way under it" (54ii)

1.8 "Your hearts saying that's it for the minute" (54iii)

1.9 "The way we live" (54iv)

1.10 "Over indulgence – that's the biggest problem" (54v)

1.11 "Sit about about and eat fatty foods, it's just going to rest on your heart" (T9, p2) (54vi)

2. Moderation of Risk

2.1 Ideas about moderation of risk

2.1.1 Motivation: (17)

2.1.2 Family history: CHD / Cancer (17i)

2.1.3 Socially (17ii)

2.1.4 Immediate health benefits (17iii)

2.1.5 Not motivated / not associating risk factors (17iv)

2.1.6 Tests might frighten me to give up smoking / change lifestyle (17v)

2.1.7 Going to the gym (17vi)

2.1.8 Anti-smoking (17vii)

2.1.9 Parental role model (17viii)

2.2 **Difficulty sustaining a healthy lifestyle** (18)

2.2.1 Diet (18)

2.2.2 Lack of will-power (18)

2.3 Scared / Vulnerability for personal health (23)

2.4 Little you can do about hereditary problems (25)

2.5 **Predisposition may be altered**, eg. (27)

2.5.1 High blood pressure

2.5.2 High cholesterol

2.6 Choice to moderate predisposed risk.

2.6.1 "I think they can reduce the risk if they change their lifestyle dramatically.

2.6.2 "You can't use that as an excuse"

2.6.3 "If I keep myself fit, I've done my bit" (29)

2.7 **No implications for individual** (44)

2.7.1 Just a one-off

2.7.2 Unfortunate

2.7.3 "It didn't make me stop and think"

2.7.4 No effect on my life. (T7, p6)

2.8 **Ideas about fate**

2.8.1 Luck of the draw (46)

2.8.2 Sibling chosen (46a)

2.8.3 Randomness (46b)

2.8.4 Fate (46c)

2.8.5 Just one of those things (46d)

2.8.6 "It's down to fate if you're going to have you're going to have one" (46e)

2.8.7 If it happens it happens (46f)

2.9 Paradox that you cannot change predisposed risk (47)

2.10 "You can try and prevent it but I don't think you can actually stop it." (49)

2.11 Balance risks / compensation (50)

2.12 Negative images of health related behaviour – "all I'd seen were me mam with a fag in her mouth". (T9, p2) (64)

2.13 Ideas about quantifying potential benefits to health in terms of life years. (66)

2.14 Coronary candidacy – inevitility (67)

2.15 Realisation of ↑ risk after 2 first degree relatives affected with CHD (71)

3. Hereditary Aspects

3.1 Hereditary factors (9)

3.2 Hereditary factors dwelling on mind / crossed my mind (10)

3.3 Influences of media coverage on health and heart problems (11)

3.4 Family clustering of heart disease / cancers (12)

3.5 Unaffected individuals in family clusters (13)

3.6 Views about the idea: (14)

Closeness = increased likeliness of heart disease

3.7 **Ideas about the fundamental nature of genetics** (34)

3.7.1 "The whole thing goes a bit deeper" (15)

3.7.2 Fundamental (34)

3.7.3 Serious

3.7.4 Building blocks of life

3.7.5 Root problem

3.7.6 "It's your being"

3.7.7 Number of relatives affected (16)

3.7.8 Little you can do about hereditary problems (25) [Also in moderation of risk]

3.7.9 Best to look on the bright side – hope that you haven't got hereditary problems (26)

3.8 **Peoples' reactions to genetic tests**: (28)

3.8.1 "Reassurance

3.8.2 "Not the best thing for them"

3.8.3 Psychological consequences (depression if result +ve)

3.8.4 Financial problems

3.8.5 Pressure on other members of family

3.8.6 How do you live with that information?

3.9. **What genetic tests can tell you**: (30)

3.9.1 Predisposition / susceptibility

3.9.2 Likelihood

3.9.3 Nasty diseases

3.9.4 A disease you can't do anything about

3.10 **Genetic tests are qualitatively different from other tests**

3.10.1 eg. Blood pressure. OR,

3.10.2 the opposite.

3.10.3 More appropriate,

3.10.4 tell you a hell of a lot more (31)

3.11 **Unalterable nature of genes**. (32)

3.11.1 Genetic problem is a unique problem – sets you apart (33)

3.11.2 Ideas about uniqueness.

3.12. **Consequences of genetic tests** (35)

3.12.1 Insurance (genetic underclass)

3.12.2 Employers

3.12.3 Cultural changes? eg. of insurance companies

3.12.4 Frightening.

3.12.5 Ability to exploit knowledge

3.12.6 Do I change my life now?

3.13 Trust yourself in the hands of people doing the test (37)

3.14 Faith / acceptance in science (38)

3.15 Too trusting? (40)

3.16 Finality / certainty of genetic tests (41)

3.17 Ideas about: public interest in genetics / genetics research (42)

3.18 Complicated (?genetics?) (43)

3.19 No previous family history (45)

3.20 Strength / weakness might be passed on (52)

3.21 Positive views about the prospect of genetic tests (59)

3.22 Views about genetic tests being similar to other screening tests (60)

3.23 Realisation of ↑ risk after 2 first-degree relatives affected with CHD (71) [Also MOR]

3.24 People scared / frightened of genetics (74)

3.25 Lack of knowledge / not informed about genetic aspects (78)

3.26 Younger age of onset = ↑ role of hereditary factors (79)

4. The Nature of the disease (CHD)

4.1 **Heart attack changed siblings lifestyle**

4.1.1 Outlook (enhanced his life)

4.1.2 Gets very depressed

4.1.3 Short-tempered

4.1.4 Not able to work

4.1.5 Frightening event.

4.2 **Psychological impact of MI**

4.2.1 Heart attack messes your brain / Taught him a lesson (48)

4.3 If you've survived a heart attack, you must have a strong heart (51)

4.4 "I associated smoking more with cancer than the heart" (62)

4.5 **Fear of cancer, but not heart disease**, wasting diseases (63)

4.5.1 "Cancer frightens me" "It weighs on my mind"

4.6 Implying that males had a greater consequence of their heart attack

4.6.1 "Dad had a major one" (T7, p1) (68)

4.7 Associating smoking with the lungs, not the heart (73)

4.8 Ideas about: (76)

4.8.1 With heart disease / heart attack you either die or survive, but with cancer death seems the inevitable outcome. (76a)

4.9 No acknowledgement of chronic morbidity associated with CHD (76b)

4.10 Preference of heart attack as a mode of death (instant) rather than a long lingering death (77)

4.11 Warning signs (80)

4.12 Suddenness of a heart attack (81)

5. Resources

5.1 **Sources of health information**: (36)

5.1.1 Internet

5.1.2 GP

5.1.3 Library (Public / Professional)

5.1.4 BHF literature and advert

5.1.5 TV

5.1.6 Chemist

5.2 Cross cultural differences in attitudes to medical profession (39)

5.3 Never asked by member of medical profession about family history (56)

5.4 Not informing GP of sibling's heart attack. "It didn't occur to me to mention it." (57)

5.5 Suggestions for information: (65)

E.g. TV. Adverts "What to do if someone has a heart attack" (T9, p5)

5.6 Indirect referral identified. E.g. (T7, p2) – one of the doctors at the general suggested we should go for a check up (69)

5.7 **Negative experiences of visiting GP after sibs MI.**

5.7.1 "To be truthful it was almost like he didn't give a toss, and I was asking him something totally pointless" T7, p3

5.7.2 "Made me feel quite small"

5.7.3 "Like a hypochondriac" (70)

5.7.4 Smoking "blamed" for almost everything by medical profession. "I think they're using smoking as an excuse" (72)

6. Reactions to relatives MI

6.1 Shock of heart attack

6.1.1 Unexpected

6.1.2 Devastated

6.1.3 Not the typical person you would think would have a...

6.1.4 It came out of the blue

6.1.5 He is the last person that I thought would.... (1)

6.1.6 Heart attack unexpected in a fit / young person (2)

6.1.7 Healthy appearance (5)

6.2 "Never think it's going to happen to you or close friends" (21)

6.2.1 "It won't bother me, it's the bloke down the road".

6.2.2 "I'm alright Jack"

6.3 "Reassurance" (linked to 17), "Excuses" (22)

6.4 **Avoidance of consulting** / Not going for a check-up (24)

6.4.1 Other priorities (24i)

6.4.2 Inertia (24ii)

6.5 Searching for an explanation (Blame) (53)

6.6 **Ideas about outlook / mental attitude** (55)

6.6.1 "It's how you feel (55i)

6.6.2 "If you worry about things too much then things happen." (page 11 tran. 3) (55ii)

6.7 **Initial grave concern for sibling following heart attack.**

6.7.1 "It really upset me...I couldn't believe how it did upset me."

6.7.2 "Try and see him through it" (T9, p3) (58)

6.8 Ideas about health problems of younger siblings in large families – "the runt of the litter" (Mother didn't give her body time to recover) (61)

7.Miscellaneous

7.1 "Not been affected so far" (7)

7.2 Stoical / stoicism (20)

Appendix 13

Qualitative phase

An example of a report from NUD*IST

++
+++ ON-LINE DOCUMENT: A14
+++ Retrieval for this document: 42 units out of 551, = 7.6%
++ Text units 93-100:
JS. So with your dad having a heart attack suddenly and dying from it, do you think, 93
exactly with your brother at a young age, how much do you feel family history 94
plays in heart problems. 95
96
A14. I don't know really, I have never thought about it. I have never thought an awful 97
lot about it. Just sort of put it to the back of my mind. ??????that many chances 98
with other problems, you know, heart attack, never thought about it. I don't think 99
you do though, you always think it will never happen to me don't you. 100
++ Text units 169-172:
A14. I don't know really, I just pushed it to the back of my mind I suppose. I mean I 169
do have regular blood tests but I don't suppose they would pick that, high 170
cholesterol level up unless I was really tested for that would they? No I never 171
thought about it. 172
++ Text units 366-369:
366
A14. I think people are afraid of cancer because you suffer such a lot of pain its, it 367
reduces you to nothing over a long time, 368
369
++ Text units 372-374:
A14. And at the end there's not a lot of, well then can do surgery on certain cancers if 372
they're caught early, but on heart disease a lot of peoples views are well it's quick 373
I've gone. 374
++ Text units 377-379:
A14. Mm. I mean if you have one good heart attack you've gone, that's it the end of it, 377
but you know if you've got a bad heart you've really got to slow down I suppose, 378
I, I don't know. 379
++ Text units 381-388:
JS. Right. You say one good heart attack I've – I never heard it described as that 381
382
A14. Well (laughs), that's it isn't it! (laughs). You know, like my dad had one, but I 383
mean there's more an' more things now they can do to sort of bring you 'round if 384
you've help there, then there was what, how long's my dad been dead, about 385
thirty, thirty three years, thirty four years, I mean there wan't a lot in then was 386
there? People just had a heart attack and that was it. 387
388
++ Text units 391-392:
A14. That's how I think a lot of people look at it. 391
392

++ Text units 414-417:

414

A14. But to be quite honest I tend to put it to the back of me mind. I suppose if I had a 415
check up and I had got a dodgy heart I'd erm have to slow down a bit or 416
whatever. I'd have to do what they told me (laughs). 417

++ Text units 451-454:

A14. Not an awful lot really, I've not got paranoid about it and sat and thought oh well 451
me brother had an heart attack and me dad did, I'm, I'm going to have one, you 452
know I just, I never thought about it. It was just a shock, but I've not doted on it 453
or anything. 454

++ Text units 513-514:

JS. Have you ever considered yourself at high risk, of heart attacks? 513
A14. Never thought about it.

+++ ON-LINE DOCUMENT: A20
+++ Retrieval for this document: 51 units out of 642, = 7.9%
++ Text units 123-128:

JS. Well, you say he was very young to have a heart attack at 48, do you think its more 123
significant than somebody having a heart attack at say 60? 124

125

A20. Well yes, obviously the younger you are the less you expect it. The older you get it's 126
not quite so shocking, its more usual I suppose, to expect someone to have a heart attack 127
when they are older 128

++ Text units 133-136:

A20. I was advised to have a cholesterol check, which I did and I passed as it were. I was 133
okay, but err yes you do tend to think does it run in the family, still do obviously but err I'm 134
not ??? deep water, but then again you don't always get warnings do you. Yes I suppose it's 135
in the back of mind that I could always follow suit, but touch wood I've not had any twinges 136

++ Text units 230-242:

JS. That's ????. Another thing that people have said and has been going on doing 230
interviews like this is that, they are much more frightened of cancer than heart disease. I 231
don't know if that's 232

233

A20. Mmm, yes I think I would say that yes, I think I would be more worried about cancer 234
than heart disease. If someone said to me you've got to have one or the other I think I'd 235
sooner have heart disease. I suppose you could always chance the ultimate is to have a heart 236
transplant, you could be cured, but you ???? so I suppose if you think you would cop for one 237
or the other you might as well cop for the one with an outside chance, you know. I think I'd 238
sooner have heart disease than lung cancer. Obviously preferably neither but given the choice 239
of one or the other. But heart disease is the biggest killer isn't it, interesting that one. You 240

know I would say I'd be more afraid of cancer than heart disease 241
242
++ Text units 411-418:
A20. Yes, yes I would tend to bury my head in the sand I think, you know
if they ever told 411
me you've got, its like I suppose if you ever have cancer. If you got
err if you could have 412
mmm perhaps 2 or 3 years of quality life, you know, say it was a slow
cancer, then you could 413
have 2 or 3 years not knowing about it and carrying on as normal, I think
I'd sooner do that 414
rather then somebody say, you've got cancer, its incurable, you've got a
couple of years. I 415
think those last 2 years would be probably miserable, because you know
your gonna, I'd 416
sooner you know if that was a scenario I think I'd sooner not know. 417
418
++ Text units 540-544:
A20. I just put it down to the fact that he'd had a heart attack, he's
survived, he was okay, 540
had he watched his lifestyle a little bit, but I didn't really put it
down, I didn't really relate it 541
to my own lifestyle too, it was his problem, not mine. Even though I
suppose, with him being 542
my brother I suppose it could have. 543
544

++
+++ ON-LINE DOCUMENT: A3
+++ Retrieval for this document: 152 units out of 386, = 39%
++ Text units 61-71:
A3. Er well until er his wife phoned me, Rene phoned me to say he'd had 61
one I hadn't got any inkling at all, that there was anything wrong and 62
er the family as regards heart diseases, he was er, played cricket, 63
football, well all the boys did. We all played football, we all kept 64
ourselves reasonably fit. Erm we all smoked, consequently I think he's 65
stopped now, which I'd like to do, I could never do that, I think at the 66
time so I don't think it really bothered me much. It was six, seven 67
years later before I got the ..... It didn't seem nothing, it didn't 68
sort of thing Ah Len's had a heart attack, I've got to completely 69
change my life. It never entered my head that 70
71
++ Text units 74-79:
A3. No it didn't change my life because I didn't, looking back I thought 74
well I think it's a one off, because the simple reason is that I don't 75
recall anything of any sign, I can't remember I can't recall any part of 76
the family having heart problems, so it didn't come into my aspect that 77
well, sorry Len you're just a one off and that's unfortunate but you're 78
the one that was chosen and that's the way I've probably thought, erm 79
++ Text units 81-96:
JS. So why do you think Len was chosen? 81
82
A3. I don't know, it's er er difficult to say. It's er hard to put into
words I 83
suppose what you try, what you think. It's why he was chosen, why is 84
anybody chosen? Er it's just sometimes the luck of the draw. You 85
just, you are or you aren't, erm I' m not a great, well I think you can, 86
I've known people fit as a fiddle, don't do anything wrong, just as 87
susceptible to have any disease as a person who's totally the opposite. 88
I think sometimes a lot of probably could be.....and you die 89

because of the way you are but I think he was a reasonably fit lad at his 90
age. I mean if he'd have been 30 years older then you're thinking well 91
he's probably had a stressful life, but at that age, I think, heart
attacks 92
at that age is quite a young age, I'd have thought. That's the only
thing 93
that bothered me about it, he's a bit young. Erm, but as you say there's 94
no rules, no rule to say you can have it at 5 or 10 or 75, 85. 95
96
++ Text units 115-131:
115
A3. Well I'd been planning for, I mean I don't know if you've ever 116
smoked, it's easier to see people say Yes you don't want to do this, it's 117
er , it's er easier said than done, er I'd started from an early age 118
probably 14, 15 and you've nearly half your life, er I can understand 119
people now not being able to break it. I mean I've tried for uff I think 120
everyone's gone in through January 1st right, New Year's resolution, 121
we'll pack the fags in this year and it lasts for two or three weeks, er
I 122
don't know, I suppose at the back of your mind you thinking of 123
something that's erm, my Mum died of cancer so you think to yourself 124
if I'm going to end up dying of cancer, so I didn't put smoking and 125
heart together, because smoking there's obviously cancer of the lungs, 126
it's a disease, erm, I don't know if people still now, I didn't go if I 127
pack the fags in, I'll have a good heart. I went if I pack the fags in,
I'm 128
going to breathe easier, probably less stress on the heart, it probably 129
will come back that way but I didn't come out and say well if I do that, 130
I don't do that. Erm, 131
++ Text units 206-224:
206
A3. Um, no, no I don't urm, I think I've got to check the rhythm. I
think 207
the last time I had one I was er so the likeliest thing is to have a
check- 208
up then. They wanted to find out then, I tend to come, when I came 209
out the nurse where I was, she gave me er a onceover, this that and the 210
other, I mean I must admit I've not had a MOT now for probably seven 211
or eight years so I don't know, but I'm in myself I feel reasonably fit.
I 212
go swimming two or three times a week, I say I play golf, I walk the 213
dog, I walk her about twenty five, thirty miles per week, er I bike ride 214
now and again if I can help it, I don't, it's, I eat, I'm probably about 215
half a stone overweight, I'd like to weigh about twelve and a half and 216
I'm about 13 2 which, well hold back I'm 53, I'm not going to think 217
that too much, because I'm going to put a bit of weight back on but 218
I've not put much on since I've stopped playing football, probably a 219
stone and a bit which in thirty years I'm not too bothered about. 220
That's why I think I don't feel vulnerable. I could drop dead from a 221
heart attack tomorrow so that's just one in , that's in my mind, there's 222
not a lot I can do about that, I don't think anybody can do anything 223
about it. You can't stop it happening, you can try and prevent it but I 224
++ Text units 227-235:
JS. So how could you try and prevent it do you think? 227
228
A3. Well as I said you diet and you things, and try and keep yourself 229
reasonably, and exercise I mean you have to walk what you say is a lot 230
but it isn't a lot to do over the course of seven days, you're only
talking 231
about three or four mile a day and you've got a big dog who wants to 232

walk four or five miles anyway, it's doing me just as good as it is him, 233
so it's er, I like to think I'm reasonably fit put it put it that way. 234
235
++ Text units 272-286:
JS. What about things like erm hereditary aspects and genetics? Do you 272
think it might play a big part or ? 273
274
A3. Erm well, at the time I said before when Len had it, it was trying to 275
find someone in the family and as near as I could damned find it I er, 276
we couldn't find anything where they said well yes er we might have 277
found one who had a heart attack, it could be a third generation away 278
in a brother or a cousin as I said. It wasn't a direct line from my Mum, 279
her Mum, it wasn't a line down, it was zigzagging all over you know it 280
wasn't the direct line so as I said well that's when I think to myself 281
well Len's just been the unfortunate one out of us six, either one of is 282
was six to one to have it and Len's the unfortunate one to have it, 283
which why it weren't worrying Christine, Ranshie myself or Jacqueline 284
because it had to be, or what anybody would do you know, I don't 285
know um. 286

Appendix 14

Qualitative phase

An example of a memo

Memos

1 30/11/01

Following discussions with BY and PL, it was felt that there was a possible link between code 71 (the realisation of an increased risk of CHD after 2 first degree relatives affected), which seems to fit into the theme of Moderation of risk and Hereditary aspects. With reference to transcript A4 page 4, it seems that rationalising the positive aspects of health-related behaviour compensates for the idea of vulnerability.

2 30/11/01 Vulnerability

Related to the above to some extent, it seems that vulnerability should find a place into the themes Moderation of Risk and Hereditary Aspects.

3 30/11/01 Never think it's going to happen

As part of the of the above discussions, it was decided to move Code 21 from Reaction to relatives MI to Moderation of Risk – at least for the time being!

4 6/12/01 Cathartic Counselling

Following interview a17, I wonder if what some of these people are telling us is that the experience of being a research participant and exploring some of the issues that they feel / think about has been beneficial to them. Furthermore, A17 said that she feels a service should be available, or perhaps a self-help group to be there in a supportive capacity – e.g. could this be undertaken by the BHF?

NB: I need to look at Dave Thompson's work with spouses of coronary patients!

5 15/12/01 Guardianship of Genetic Information

Reading the transcript of interview A17 and I think that similar ideas have cropped up elsewhere, but I cannot remember where! The idea that there ought to be some sort of guardianship for genetic information seems common.

6 8/1/02 Support network

Looking at the transcript of interview A17, it really does seem that what this participant needed was some kind of support network / structure to be available.

7 22/01/02 Support network / Gender differences

I think that there may be gender differences in the out-look of some of these people, particularly with reference to the need / request for support networks.

8 29/01/02 Trigger for consultation

Looking at transcript A18, there seems to be a necessity for a trigger to prompt people to seek medical advice. Although people my have a realisation of increased risk, this participant needed to start experiencing chest pains herself before seeking advice!

9 12/02/02 Detachment

While preparing the abstract for the BPS, it became apparent that detachment is / was perhaps a bigger issue than previously realised. BY suggested that we needed to look into this when interviews were coded in NUD*IST.

10 8/05/02 "Ostriches"

While preparing theme framework, one of the key things that appear to crop up in relation to genetic testing is the notion of "Ostriches". The idea that people would rather "bury their head in the sand" or "rather not know" about their individual risk. Certainly transcript A20 seems to refer to this quite forcefully.

11 02/05/03

See Memo 8 – trigger for consulting. This in interview A20 too. I've coded under 6.2 I the theme / node NUDIST system. I wonder if this needs a separate code, or to split the node?

12 12/07/03

Interview A18, line 162-167 "...something on the hereditary side of heart attacks. I do think there is something there..." This should fit into 2.1, but ideally fists somewhere in 3.1 – perhaps need a sub-category of 3.1?

Appendix 15

Qualitative phase
Characteristics of participants

Summary characteristics of participants

ID	Age	Gender	Occupation	Marital status	Smoking status	Ages smoked	Number of cigarettes		Gender of affected sibling	Age of affected sibling	Age of affected sibling at MI
A1	49	Female	Printer	Divorced					Female	48	47
A2	51	Female	Kitchen assistant	Married					Female	50	49
A3	53	Male	Installation engineer	Married					Male	50	30
A4	41	Female	Carer (nurse)	Married					Male	42	40
A5	43	Male	Business analyst	Married	Never		N/A		Female	50	47
A6	39	Female	Housewife	Married	Never		N/A		Male	43	41
A7	32	Male	Sanitation officer	Married	Current		20		Female	36	36
A8	41	Male	Driver	Single	Previous	14-18	20-40		Male	48	43
A9	30	Male	Sheet metal worker	Married	Never		N/A		Male	44	41
A10	34	Male	Own taxi business	Married	Previous	14-29	20		Male	40	36
A11	47	Male	Plasterer / welder	Separated	Never		N/A		Male	50	48
A12	46	Female	Book keeper	Married	Current		10-20		Male	51	32
A13	54	Male	Lagger / builder	Married	Previous	20-39	20		Male	50	43
A14	55	Female	Housewife / carer	Married	Previous	24-27	16		Male	51	46
A15	50	Female	Seamer (knitwear)	Married	Previous	12-41	>30		Male	58	51
A16	58	Female	Own business	Married	Previous	18-52	5-15		Male	50	44
A17	58	Female	Research assistant	Widowed	Previous	20-33	40		Male	53	53
A18	53	Female	Machine minder	Divorced	Previous	18-34	20		Female	57	45
A19	57	Male	Printer	Married	Previous	14-52	>40		Male	59	49
A20	47	Male	Printer	Married	Previous	15-33	20		Male	53	48

Thus data were collected prior to the interviews using the form described in the previous chapter. The data on the left of the shadowed vertical is all regarding the interview, while the data to the right is concerning their affected sibling. Of the affected siblings of the interviewees, the majority (15) were male, with current ages ranging from 36-59 years and their age at MI being 30-49 years.

Appendix 16

Qualitative phase
Final thematic framework

Final thematic framework

Theme 1: The experiences of premature heart attack in a sibling

1.1 Surprise / Shock

1.2 Vulnerability

1.3 Detachment

1.4 Complacency / indifference

1.5 Beliefs about moderating risk

1.6 Barriers to moderating risk

1.7 Information and support

1.8 Lifestyle changes following siblings' heart attack

Theme 2: Explanations for siblings' heart attack

2.1 Life event

2.2 Causal attributions

Theme 3: Inheritance and genetics

3.1 Nature and genetics

3.2 The nature of genetic testing

3.3 Views about future susceptibility testing

3.4 Public interest in genetic research

Theme 4: Experiences with heath care professionals

Appendix 17

Qualitative phase

Glossary of symbols used in quotes

Glossary of symbols used in quotes

The following symbols were used when presenting the quotes from participants in the qualitative phase of the study:

Symbol	Explanation
(A7)	This refers to the seventh participant in phase one (qualitative phase).
[text]	These signify that text was inserted into the quote to contextualise the meaning, or maintain confidentiality. In all cases, the names of participants' sibling or other person (e.g. GP) have been replaced to ensure anonymity.
...	This signifies that some of the quote has not been presented because it was felt that it did not enhance the meaning.

Appendix 18

Quantitative phase Protocol

Health perceptions of people who have a sibling who has experienced a heart attack (LREC 6685) (Phase 2)

Study Protocol: Written December 2000, amended April 2002

Investigators: Stribling J, Young B, Lambert PC, Samani NJ.

Objectives

1. To quantify the findings of an earlier qualitative study to further investigate the risk perceptions of individuals currently unaffected by CHD, who have a sibling who has experienced a premature heart attack.
2. To compare the risk perception and health beliefs of unaffected individuals, whose sibling has experienced a premature heart attack, with individuals who have no family history of early onset heart disease.

Background

Coronary Heart Disease (CHD) is a leading cause of morbidity and mortality in the western world. In England alone, some 110,000 annual deaths are attributable to CHD, and this is one of the key areas of Government health policy (Department of Health, 2000). CHD has a complicated and multi-factorial aetiology (Tunstall-Pedoe et al., 1997), and whilst conventional risk factors (smoking, hypercholesteraemia, hypertension and diabetes mellitus) can certainly account for a large proportion of events, it has been estimated that approximately 50% of "risk" remains unexplained (Nora, 1983).

Genetic predisposition is rapidly emerging to account for much of the unknown risk of CHD (Marian, 1999; Broeckel et al., 2002). Family history (or familial aggregation) of CHD is an "independent" risk factor for CHD (Hopkins et al., 1988), and an important predictor for premature disease (Eaton et al., 1996). Much of the criticism of early studies into familial clustering of CHD argued that a "shared" adverse environment between family members could be responsible, but adoption studies (Sorenson et al., 1988) and twin studies (Marenberg et al., 1994), have seriously undermined these criticisms. Having a first-degree relative affected by CHD under the age of 65 increases the individual's risk by at least twofold, and a maternal history of MI (myocardial infarction) under the age of 50 increases the risk tenfold (Samani & Singh, 2001). The rapidly advancing area of molecular biology and the Human

Genome Project may revolutionise medicine (Collins, 1999) by providing clear indications of how individuals can modify their inherited risks through behavioural change, but whether the health-related behaviour of individuals will be affected positively is unclear (Marteau & Senior, 1997).

Lay beliefs or perceptions about the aetiology and causation of CHD (Davison, et al., 1991), may emphasise notions of "coronary candidacy" and "fatalism", which individuals may feel absolve them from personal responsibility for their own health and encourage them to believe that their own behaviour is not relevant to their risks of developing CHD. Defensive biases in health-related judgements, particularly in relation to information about risk factor status have also been observed (Croyle, et al., 1997). The extent to which such beliefs and judgements are shared by those with a family history of CHD is unknown. There is some evidence that individuals who currently do not have CHD seem to have a poor understanding of their own relative risk and appear resistant to behavioural change even when their siblings (Becker & Levine, 1987), or mother is affected (Allen & Blumenthal, 1998). For example, individuals may persist in smoking even when they perceive that they have a family history of CHD (Hunt, et al., 2000), and often do not perceive themselves to be at increased risk (Hunt et al., 2001). A much better understanding is therefore needed of perceptions of risk among those with a higher risk of CHD by virtue of family history, and how these perceptions affect behaviour.

Setting

Among the current genetic studies based at Glenfield Hospital and the University Division of Cardiology is PRAMIS (**P**latelet **R**eactivity **A**nd polymorphisms in **M**yocardial **I**nfarction **S**tudy), a case-control association study of people (under the age of 50) with premature myocardial infarctions. Having access to the participants of this study, and their unaffected siblings, places the Division of Cardiology in an ideal position, in collaboration with the Department of Epidemiology and Public Health, to investigate the perception of risk of CHD and smoking behaviour of unaffected individuals whose siblings have experienced a heart attack.

Design

This is a two-phase study consisting of a qualitative phase involving semi-structured interviews and a quantitative phase involving a postal survey. Phase 1 of the study

received Ethics Committee approval (Ref. No. 6685) in January 2001 and data collection has recently been completed.

Report on Phase 1

A qualitative approach was adopted, involving semi-structured interviews, within a grounded theory framework. Semi-structured interviews were completed with 20 unaffected individuals (aged 30-58), whose siblings had experienced a premature MI (<55 years) and had participated in the PRAMIS study. Quota sampling was used to represent the diversity of social class of the PRAMIS cohort. Interviews were tape-recorded, transcribed verbatim, and data analysis adopted the constant comparison method (Green, 1998).

Participants in this study expressed a sense of detachment from their siblings' MI, indicating that their siblings' heart problems had little or no significance or relevance for their judgements of own risk status. Participants highlighted their siblings' lifestyle (e.g. smoking, dietary habits, stress) or chance, rather than inheritance as an explanation of their siblings' heart problems. Amongst those participants who had consulted their GP for advice following their siblings' MI, some had been investigated, but many felt that their concerns had been dismissed or trivialised, often reinforcing their existing feelings of detachment. Participants viewed genetic tests as fundamentally different from other screening procedures, and some were highly resistant to the future prospect of screening for genetic risk factors for heart disease. This work has been submitted as a paper to the national conference of the *British Psychological Society* conference *(Health Psychology Division)* (Stribling et al. 2002), and locally for the Leicestershire Research Day. Further publications are planned and will follow.

Phase 2

A postal survey questionnaire will be utilised to quantify key aspects of the findings from the interviews of phase 1, and in particular, to estimate the proportion of unaffected individuals who report their siblings' health problems to be of little consequence for their own health. Our study will also involve a comparative element: a questionnaire will be sent to a control group of individuals who have no family history of early onset heart disease (the same control group as for PRAMIS) to compare the risk perceptions and beliefs about preventative behaviour of the two groups.

The issues to be addressed in the second phase of our study have been partly identified from the qualitative phase of our study and partly from existing published work. A near final draft of our questionnaire is attached. This has been piloted locally with colleagues, friends and relatives of the researchers to ensure that the questions are sufficiently clear and easy to interpret. Further piloting will be undertaken to finalise question wording and sequencing, and the format and layout of the questionnaire. Where possible we have derived questions from previously published work, which is relevant to the issues that we aim to investigate. However, to fully address the aims of our study, it has been necessary to devise some completely new questions, or to slightly modify questions from previously published work. The questionnaire is divided into two sections, the first for both cases (unaffected siblings of PRAMIS participants) and controls, and the second (i.e. last page) for cases only.

Prior to undertaking the interviews, we had intended to measure health-related behaviour as part of the postal questionnaire, however after studying the literature in some detail, it has become apparent that it would not be feasible to undertake this in a manner that is valid and reliable (e.g. Huston et al, 2000) with the limited resources available for our study. Furthermore, the interviews have indicated that health-related behaviour was of only secondary importance to the participants, in relation to causal attributions, personal risk and genetic testing. We have included questions on smoking, however, as it is relatively straightforward to measure, and gives an indication of one important aspect of health behaviour.

The main themes from the interviews, therefore, that are to be followed-up and measured in this postal survey, include causal attributions for heart disease, perceptions of personal risk, attitudes to developments in genetic research and health technology, and the views about the prospect of future genetic screening and testing. These were identified by the interviewees of Phase 1 of the study as issues of considerable importance, and are issues for which it was possible to derive questions from previously published work: causal attributions (Weinman et al. 2000, adopted from De Valle & Norman, 1992), perceptions of personal risk (Wilcox & Stefanick, 1999), and genetic developments (Human Genetics Commission, 2001).

Particular issues identified in the interviews for which it was necessary for us to modify questions from previously published work are:

- Beliefs about heart disease relative to other classes of major illnesses (cancer and stroke) using questions modified from Wilcox & Stefanick (1999), who explored the beliefs of women about their fear, knowledge and perceptions of developing specific cancers (breast, colon and lung) in relation to heart disease. For our study these questions have been modified to facilitate comparison of individuals' views about these illnesses as groups or classes of disease. In other words, we refer to beliefs about cancer in general, rather than to specific types of cancer, and to heart disease rather then to specific types of heart disease.
- Perceptions of the importance of health or health as a value using questions which we have slightly modified from work by Lau et al. (1986), following our pilot which indicated that some of the original questions were ambiguous.

For certain issues we had to devise our own questions. This includes:

- Attitudes to the prospect of future susceptibility testing for common multi-factorial conditions. The Human Genetics Commission investigated attitudes to genetic testing, but this was mostly in the context of parental decisions about genetic testing (HGC, 2001). Therefore, we have developed questions to measure attitudes about the prospect of future genetic testing, in relation to the disease groups of cancer, heart disease and stroke.
- The reactions of individuals to their siblings' MI. These can be found on the final page of the questionnaire and will be distributed solely to the unaffected siblings of the PRAMIS participants. These questions build upon more of the themes identified from the interviews, particularly unaffected siblings' experiences of GP consultations, and their desire for support and information following their siblings' MI.

Additionally, smoking is again used as an easily measurable aspect of behaviour change (modified from Office for National Statistics, 1997) in the section for unaffected siblings.

Source of Subjects:

- Unaffected individuals who have 1 sibling who has experienced a heart attack (i.e. siblings of PRAMIS cases which, comprises approximately 200 people).

- Unaffected individuals, where there is no family history of heart disease (i.e. the PRAMIS control group, which comprises approximately 200 people).

Recruitment (further to question 14a)i) on ethics application form)

Letters of invitation to this study will come from Professor Samani (Lead Clinician and Principal Investigator for the PRAMIS study). However because of the family nature of the study, the process of invitation will be different for each group.

Comparison Group (PRAMIS Controls)

A letter of invitation will be sent directly to the PRAMIS controls. Included with this will be an information sheet, the questionnaire and postage paid return envelope. The letter will include contact details for Julian Stribling (Research Nurse) so that potential participants can discuss, by telephone, any concerns that they might have. They will indicate whether or not they consent by completing and returning the questionnaire, or by ticking a box on the questionnaire to indicate that they prefer not to participate and returning the uncompleted questionnaire to us. Individuals who decline to participate will not be contacted further about the study. Individuals who do not reply will be sent up to two reminders.

Unaffected Sibling Group (Sibling of PRAMIS Cases)

A letter of invitation will be sent directly to the PRAMIS cases. Included with this, will be an information sheet, reply-slip (with postage paid return envelope) and a stamped (but un-addressed) envelope for their sibling. [NB. The envelope for the sibling will contain a letter, information sheet, the questionnaire and postage paid return envelope].

We will be asking PRAMIS participants to forward the envelope to **one** of their siblings (after discussion with their sibling if they wish), and to return the reply-slip stating whether they have either forwarded the envelope, or whether they have opted to decline and wish no further contact. Therefore, we will not be contacting unaffected siblings directly at this stage, and will not be requesting access to their personal details. The envelope to the sibling will include a letter, information sheet, questionnaire and postage paid return envelope. The letter will also include contact details for Julian Stribling (Research Nurse) so that potential participants can discuss, by telephone, any concerns that they might have. Unaffected siblings will indicate whether or not they consent by completing and returning the questionnaire, or by ticking a box on the questionnaire to indicate that they prefer not to participate

and returning the uncompleted questionnaire to us. Siblings who decline to participate will not be contacted further about the study. Individuals who do not reply will be sent up to two reminders.

Statistical power of the study (further to question 12e) on ethics application form)

In relation to objective one of our study: assuming a response rate of 60% (from unaffected siblings of PRAMIS cases), and a percentage of unaffected siblings having a particular risk perception or health belief of 60%, this percentage will be estimated to within approximately 9% of its true value. In relation to objective two: there is 80% power to detect a difference between the unaffected sibling group and the affected sibling group of 18% (at the 5% significance level).

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Appendix 19

Quantitative phase

Research ethics letter of approval

Leicestershire, Northamptonshire and Rutland Health Authority

Melanie Sursham
Direct Dial 0116 258 8610

10 May 2002

Gwendolen Road
Leicester
LE5 4QF

Tel: 0116 273 1173
Fax: 0116 258 8577
Mini Com: 0116 258 8640
DX 709470 Leicester 12

Mr J M Stribling
Research Nurse
Division of Cardiology
Clinical Sciences Wing
Glenfield Hospital

Dear Mr Stribling

Health perceptions of people who have a sibling who has experienced a heart attack (Phase 2) Phase 1 being Project No 6685 – our ref. no. 6697

Further to your application dated 15 April, you will be pleased to know that the Leicestershire Research Ethics Committee at its meeting held on the 3 May 2002 approved your application to undertake the above mentioned research.

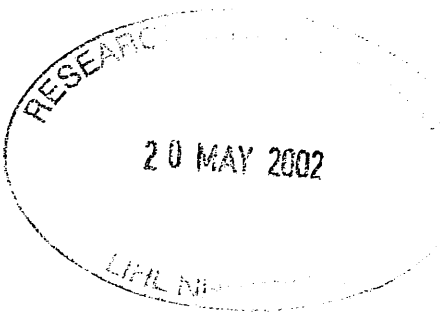
The Committee felt that the questions being asked had been phrased in such a way that the answer was implied and suggested that more open phrasing of the questions was required.

Your attention is drawn to the attached paper which reminds the researcher of information that needs to be observed when Ethics Committee approval is given.

Yours sincerely

P G Rabey
Chairman
Leicestershire Research Ethics Committee

(NB All communications relating to Leicestershire Research Ethics Committee must be sent to the Committee Secretariat at Leicestershire, Northamptonshire and Rutland Health Authority. If, however, your original application was submitted through a Trust Research & Development Office, then any response or further correspondence must be submitted in the same way.)



Appendix 20

Quantitative phase

Trust indemnity letter of approval

University Hospitals of Leicester NHS
NHS Trust

DIRECTORATE OF RESEARCH AND DEVELOPMENT
Director: Professor J Feehally
Business Manager: Dr N J Seare
Co-ordinators: Glennis Jarvis and Aimee Geary

Direct Dial: (0116) 258 4109
Fax No: (0116) 258 4226

Leicester General Hospital
Gwendolen Road
Leicester
LE5 4PW

Tel: 0116 2490490
Fax: 0116 2584666
Minicom: 0116 2588188

20 May 2002

Mr J Stribling
Research Nurse
Division of Cardiology
Glenfield Hospital

Dear Mr Stribling

RE: Project Number: 7825 *[Please quote this number in all correspondence]*
Health perceptions of people who have a sibling who has experienced a heart attack Phase 2 (Phase 1 being Project No. UHL 6685)

We have now been notified by the Ethical Committee that this project has been given ethical approval (please see the attached letter from the Ethical Committee).

Since all other aspects of your UHL R+D notification are complete, I now have pleasure in confirming full approval of the project on behalf of the University Hospitals of Leicester NHS Trust.

This approval means that you are fully authorised to proceed with the project, using all the resources which you have declared in your notification form.

The project is also now covered by Trust Indemnity, except for those aspects already covered by external indemnity (e.g. ABPI in the case of most drug studies).

We will be requesting annual and final reports on the progress of this project, both on behalf of the Trust and on behalf of the Ethical Committee.

In the meantime, in order to keep our records up to date, could you please notify the Research Office if there are any significant changes to the start or end dates, protocol, funding or costs of the project.

I look forward to the opportunity of reading the published results of your study in due course.

Yours sincerely

Dr Nichola Seare
Research and Development Business Manager

Appendix 21

Quantitative phase

Letter to PRAMIS case

University Hospitals of Leicester NHS
NHS Trust

Glenfield Hospital
Groby Road
Leicester
LE3 9QP

Tel: 0116 2871471
Fax: 0116 2583950
Minicom: 0116 2879852

Date:

To:

Dear

Re: Health perceptions of people who have a sibling who has experienced a heart attack (Phase 2)

You kindly participated in a research study at Glenfield Hospital some time ago (the PRAMIS study) undertaken by Dr Ravi Singh under my supervision. I am very grateful for your help.

We are now conducting a further study to examine the views **of brothers or sisters** of people like yourself who have had a heart attack at a young age. We are contacting you again because we would like to send your brother or sister a short questionnaire to find out their views about their health. We believe that this is an important area and one that has had little research.

We would be very grateful if you could read the attached information about our study, and send the enclosed envelope (containing details of the study) to any **one** of your brothers or sisters with whom you are in contact. You may wish to discuss our study with your brother or sister first before sending the envelope on to them.

If your brother or sister agrees to help us with our study, it will only involve them completing a simple postal questionnaire, which should take no more than fifteen minutes. There are **no** visits or blood samples required. Further details of the study are given on the enclosed information leaflet. To forward the details to your brother / sister, please address and post the white stamped envelope, containing a letter and the questionnaire. There is no obligation for them to take part if they do not wish to do so. Then, please complete the reply slip enclosed with this letter and return it to me using the brown pre-paid envelope. If you prefer not to pass on details of our study to your brother or sister please tell us by ticking the appropriate box on the reply slip, and we will not trouble you again.

I would like to thank you for taking time to read this letter and hope to hear from you soon. If you have any queries, please feel free to contact Julian Stribling (Research Nurse) on 0116 256 3791.

Yours sincerely

Professor NJ Samani
Professor of Cardiology
University of Leicester

Appendix 22

Quantitative phase

Reply slip from PRAMIS case

REPLY SLIP

Health perceptions of people who have a sibling who has experienced a heart attack (Phase 2)

Please return this slip, even if you do not wish to take part in the study, using the brown postage paid envelope.

To: Julian Stribling
Division of Cardiology
Clinical Sciences Wing
Glenfield Hospital,
LEICESTER, LE3 9QP

I have forwarded the letter to my brother / sister (please delete as appropriate) as requested ☐

If you are happy for us to contact you by phone, please write your phone

number here:..... Best time to contact me:... ... ...

I prefer not to forward this information to my brother / sister

☐ **and do not wish to be contacted again about this study**

Signed:..... Date:.....

Name:_____

Address:_____

_____

_____

Appendix 23

Quantitative phase

Letter to sibling of PRAMIS case

University Hospitals of Leicester NHS
NHS Trust

Glenfield Hospital
Groby Road
Leicester
LE3 9QP

Tel: 0116 2871471
Fax: 0116 2583950
Minicom: 0116 2879852

INVITATION TO PARTICIPATE IN A RESEARCH STUDY

Re: Health perceptions of people who have a sibling who has experienced a heart attack (Phase 2)

As you may know your brother / sister, participated in a research study at Glenfield Hospital some time ago (the PRAMIS study). We are contacting you via him / her to invite you to help us with our further study, the aim of which is to find out more about the views of people (like yourself) who have a brother or sister who has had a heart attack at a young age. This letter has been posted to you by your bother / sister at our request and he / she has not released details of your name or address to us.

We believe that our study is investigating some very important issues that have been little researched in the past. If you would like to take part, it will involve you completing the enclosed questionnaire, which should take no more than fifteen minutes. There are **no** visits or blood samples required. To help you decide about whether to participate, we have enclosed an information leaflet, which gives more details of our study.

If you would like to take part in this study, please complete the questionnaire and the participant details form, and return these to us in the pre-paid envelope. If you prefer not to participate, please indicate this on the back of the questionnaire and then return it to us, leaving the remaining questions blank.

I would like to thank you for taking time to read this letter and hope to hear from you soon. If you have any queries, please feel free to contact Julian Stribling (Research Nurse) on 0116 256 3791.

Yours sincerely

Professor NJ Samani
Professor of Cardiology
University of Leicester

Appendix 24

Quantitative phase

Information sheet - unaffected sibling version

University Hospitals of Leicester NHS
NHS Trust

Glenfield Hospital
Groby Road
Leicester
LE3 9QP

Tel: 0116 2871471
Fax: 0116 2583950
Minicom: 0116 2879852

Health perceptions of people who have a sibling who has experienced a heart attack

Information leaflet for participants in Phase 2
Version 1, April 2002

Participant version

We would like to invite you to participate in a study, which is looking at the health perceptions and lifestyles of people who have a brother or sister who has had a heart attack.

1. What is the purpose of the study?

We are keen to learn more about the views of people who do not have any heart trouble themselves, but who have a brother or a sister who has had a heart attack. In order to understand how families are affected by heart disease, we feel it is very important to seek the views of family members.

2. What will be involved if I take part in the study?

If you agree to take part we would like you to complete the enclosed questionnaire and return it in the pre-paid envelope provided. We anticipate that it will only take you about 15 minutes to complete the questionnaire. There are no right or wrong answers: we are merely interested in **your own views and opinions**.

The study will **not** involve you in any medical examinations, procedures, tests or treatments.

3. Will information obtained in the study be confidential?

This study will be undertaken in association with the University of Leicester, and all information you give us will be treated confidentially, in accordance with the data protection act. Your name will not appear anywhere on documents about the study.

4. What if I am harmed by the study?

There are no blood tests, visits or treatments involved and we do not anticipate any harm. However, this study, like all medical research is covered for mishaps in the same way, as for patients undergoing treatment in the NHS i.e. compensation is only available if negligence occurs.

5. What happens if I do not wish to participate in this study or wish to withdraw from the study?

If you do not wish to participate in this study please tell us by ticking the appropriate box on the front of the questionnaire and return it in the prepaid envelope. This will tell us that you do not wish to be contacted further. Your future treatment will not be affected.

6. Where can I get further information about the study?

For further information, please contact Julian Stribling by telephoning: 0116 256 3791. Listed below are the names of everyone else involved in running this study. Any of them would also be delighted to talk to you if there is anything you wish to discuss.

Investigators:

Mr. Julian Stribling
Research Nurse
University of Leicester
Division of Cardiology
University of Leicester
Clinical Sciences Wing
Glenfield Hospital
Leicester, LE3 9QP
Tel. 0116 256 3791

Dr. Bridget Young
Honorary Visiting Fellow
Department of Epidemiology
and Public Health
22-28 Princess Road West
Leicester
LE1 6TP

Dr. Paul Lambert
Lecturer in Medical Statistics
Department of Epidemiology and Public Health
University of Leicester
22-28 Princess Road West
Leicester, LE1 6TP
Tel. 0116 252 5407

Professor NJ Samani
Professor of Cardiology
University of Leicester
Division of Cardiology
Clinical Sciences Wing
Glenfield Hospital
Leicester, LE3 9QP

Appendix 25

Quantitative phase

Information sheet - comparison group version

University Hospitals of Leicester NHS
NHS Trust

Glenfield Hospital
Groby Road
Leicester
LE3 9QP

Tel: 0116 2871471
Fax: 0116 2583950
Minicom: 0116 2879852

Date:

To:

Dear

Re: Health perceptions of people who have a sibling who has experienced a heart attack (Phase 2)

Comparison Group Version

You kindly participated in a research study at Glenfield Hospital some time ago (the PRAMIS study) carried out by Dr Ravi Singh under my supervision, for which we are very grateful.

We would now like to invite you to help us again with a further smaller study. This is a **postal questionnaire** study to find out about the views and beliefs of people **who have no personal or family history of heart problems (like yourself).** We will use the information you give us to compare your views with those of other people who have a family history of heart problems. We believe that this is an important area, and one that has had little research.

If you agree to participate, it will involve you completing the enclosed questionnaire, which should take no more than fifteen minutes. There are **no** visits or blood samples required. To help you decide, we have enclosed an information leaflet, which gives more details of our study.

If you would like to take part in this study, please complete the questionnaire and return it to us in the pre-paid envelope. If you prefer not to participate, please indicate this by ticking the appropriate box on the front of the questionnaire. This will tell us that you do not wish us to contact you further about this study.

If you have any queries, please do not hesitate to contact Julian Stribling (Research Nurse) on 0116 256 3791.

I would like to thank you for taking time to read this letter and hope to hear from you soon.

Yours sincerely

Professor NJ Samani
Professor of Cardiology
University of Leicester

University Hospitals of Leicester NHS
NHS Trust

Glenfield Hospital
Groby Road
Leicester
LE3 9QP

Tel: 0116 2871471
Fax: 0116 2583950
Minicom: 0116 2879852

Health perceptions of people who have a sibling who has experienced a heart attack

Information leaflet for participants in Phase 2
Version 1, April 2002

Version for comparison group

We would like to invite you to participate in our study, which is looking at the health perceptions and beliefs of people **who have no personal or family history of heart problems** (like yourself). We will use this information to compare your views with those of people who have a brother or sister who has had a heart attack.

1. What is the purpose of the study?

We are keen to learn more about the views of people who have never had any heart trouble themselves, but who have a brother or a sister who has had a heart attack. In order to understand how families are affected by heart disease, we feel it is very important to seek the views of family members, and to be able to compare them with the views of people where there are no heart problems in the family.

2. What will be involved if I take part in the study?

If you agree to take part we would like you to complete the enclosed questionnaire and return it in the pre-paid envelope provided. We anticipate that it will only take you about 15 minutes to complete the questionnaire. There are no right or wrong answers: we are merely interested in **your own views and opinions**.

The study will **not** involve you in any medical examinations, procedures, tests or treatments.

3. Will information obtained in the study be confidential?

This study is being undertaken in association with the University of Leicester, and all information you give us will be treated confidentially. Your name will not appear anywhere on documents about the study.

4. What if I am harmed by the study?

There are no blood tests, visits or treatments involved and we do not anticipate any harm. However, this study, like all medical research is covered for mishaps in the same way, as for patients undergoing treatment in the NHS i.e. compensation is only available if negligence occurs.

5. What happens if I do not wish to participate in this study or wish to withdraw from the study?

If you do not wish to participate in this study please tell us by ticking the appropriate box on the front of the questionnaire and return it in the prepaid envelope. This will tell us that you do not wish to be contacted further. Your future treatment will not be affected.

6. Where can I get further information about the study?

For further information, please contact Julian Stribling by telephoning: 0116 256 3791. Listed below are the names of everyone else involved in running this study. Any of them would also be delighted to talk to you if there is anything you wish to discuss.

Investigators:

Mr. Julian Stribling
Research Nurse
University of Leicester
Division of Cardiology
Clinical Sciences Wing
Glenfield Hospital
Leicester, LE3 9QP
Tel. 0116 256 3791

Dr. Bridget Young
Honorary Visiting Fellow
Department of Epidemiology and Public Health
University of Leicester
22-28 Princess Road West
Leicester LE1 6TP
Tel: 0116 252 3214

Dr. Paul Lambert
Lecturer in Medical Statistics
Department of Epidemiology and Public Health
University of Leicester
22-28 Princess Road West
Leicester, LE1 6TP
Tel. 0116 252 5407

Professor NJ Samani
Professor of Cardiology
University of Leicester
Division of Cardiology
Clinical Sciences Wing
Glenfield Hospital
Leicester, LE3 9QP

Appendix 26

Quantitative phase

Letter to GP

Dr. GP Name
GP Address 1
GP Address 2
Leicester

12th May 2004

Dear Dr. GP Name

Re: PRAMIS Case
Address:
Date of birth:

The above person, who is registered with your practice, kindly participated in a research study investigating inherited aspects of coronary heart disease some time back.

We are hoping to contact him again for a further brief study, which will involve a postal questionnaire. However, before we undertake this, we are anxious to ensure that this person is still alive. We would be very grateful if you could confirm that this person is still registered with your practice, and at the address above by signing below and returning in the stamped, addressed envelope.

We have enclosed a copy of the original consent form for your information. If you require any further information, please do not hesitate to contact Julian Stribling on 0116 256 3791.

GP Name:
I can confirm above patient registered with us
Signature:
Date:

With thanks and best wishes,

Yours sincerely,

NJ Samani
Professor of Cardiology

Julian Stribling
Research Nurse

Appendix 27

Quantitative phase

Questionnaire

University of Leicester

University Hospitals of Leicester NHS

NHS Trust
Glenfield Hospital

ID ☐ ☐ ☐ ☐

Please answer the following questions

1) What is your sex? Male ☐ Female ☐

2) What is your date of birth? Day ☐☐ Month ☐☐ Year ☐☐☐☐

3) What is your postcode? _____

4) What is (was) the full title of your current (or last) job? _____

5) How would you describe your work? (please tick one box only)

Professional ☐ Managerial ☐ Skilled non-manual ☐

Skilled manual ☐ Partly skilled ☐ Unskilled ☐

6) How old were you when you finished full-time education?
(School or college, whichever you last attended full-time) (please tick one box only)

16 or under ☐ 17 ☐ 18 ☐ 19 or over ☐

7) a) Do you smoke cigarettes at all nowadays? Yes ☐ No ☐

b) If yes, about how many cigarettes a **day** do you usually smoke now?

1-5 ☐ 6-10 ☐ 11-15 ☐ 16-20 ☐ 20 or more ☐

8) Please indicate how many times you have visited your GP in the last **2 years**

None ☐ 1-3 Times ☐ 4-6 Times ☐ 7 Times, or more ☐

9) Over the last twelve months would you say your health has on the whole been:

Good? ☐ Fairly good? ☐ Not good? ☐

10) Please indicate how much you agree or disagree with each of the following statements by ticking the box that best represents your views

	Strongly disagree	Tend to disagree	Neither	Tend to agree	Strongly agree
a) Diabetes **increases** the risk of heart disease	☐	☐	☐	☐	☐
b) Genetic factors **increase** the risk of heart disease	☐	☐	☐	☐	☐
c) Stress or worry **increases** the risk of heart disease	☐	☐	☐	☐	☐
d) Eating fatty foods **increases** the risk of heart disease	☐	☐	☐	☐	☐
e) Smoking **increases** the risk of heart disease	☐	☐	☐	☐	☐
f) Lack of exercise **increases** the risk of heart disease	☐	☐	☐	☐	☐
g) High blood pressure **increases** the risk of heart disease	☐	☐	☐	☐	☐
h) Chance or fate **is involved** in the development of heart disease	☐	☐	☐	☐	☐
i) Being overweight **increases** the risk of heart disease	☐	☐	☐	☐	☐
j) High cholesterol **increases** the risk of heart disease	☐	☐	☐	☐	☐

11) Of the following 10 factors, please pick **the 5** that **you** think are the most important in increasing the risk of heart disease. Please list them in the order that best represents how important you think they are (with 1 being the most important)

- Diabetes
- Eating fatty foods
- Lack of exercise
- Genetic factors
- Smoking
- High blood pressure
- High cholesterol
- Stress or worry
- Being overweight
- Chance / fate

1 _____

2 _____

3 _____

4 _____

5 _____

12) Please list any other things that you think are important risk factors for heart disease:

_____

_____

Please indicate how much you agree or disagree with each of the following statements by ticking the box that best represents your views

		Strongly disagree	Tend to disagree	Neither	Tend to agree	Strongly agree
13 a)	More people die from cancer than heart disease	☐	☐	☐	☐	☐
b)	A man over age 65 is much more likely to die from heart disease than a woman over age 65	☐	☐	☐	☐	☐
c)	Having a parent with heart disease increases my risk of developing the disease	☐	☐	☐	☐	☐
d)	Having a brother or sister with heart disease increases my risk of developing the disease	☐	☐	☐	☐	☐

14) **Compared with other people of *your* age and sex**, what do you think are the chances of ***you*** getting the following diseases in the next 15 years?

		Very low	Low	Average	High	Very high
a)	Heart disease	☐	☐	☐	☐	☐
b)	Stroke	☐	☐	☐	☐	☐
c)	Cancer	☐	☐	☐	☐	☐

15) Do you think it is possible for ***you*** to do anything to reduce your risk of the following diseases?

		Yes, definitely	Yes, maybe	No, probably not	No, definitely not
a)	Heart disease	☐	☐	☐	☐
b)	Stroke	☐	☐	☐	☐
c)	Cancer	☐	☐	☐	☐

16) Which **one** of the following illnesses do you fear most? (please tick one box only)

Heart disease ☐

Stroke ☐

Cancer ☐

Please indicate how much you agree or disagree with each of the following statements by ticking the box that best represents your views

		Strongly disagree	Tend to disagree	Neither	Tend to agree	Strongly agree
17a)	If you don't have your health, you don't have anything	☐	☐	☐	☐	☐
b)	Good health is only of minor importance in a happy life	☐	☐	☐	☐	☐
c)	There are few things more important than good health	☐	☐	☐	☐	☐
18a)	New genetic developments will bring cures for many diseases	☐	☐	☐	☐	☐
b)	Research on human genetics is tampering with nature and is therefore unethical	☐	☐	☐	☐	☐
c)	Genetic research has the potential to improve ***my*** health	☐	☐	☐	☐	☐
19a)	Genetic research will lead to improvements in the health of future generations	☐	☐	☐	☐	☐
b)	If a genetic test were available, that could indicate that I was at increased risk of developing heart disease, I would want to have such a test	☐	☐	☐	☐	☐
c)	If a genetic test were available, that could indicate that I was at increased risk of having a stroke, I would want to have such a test	☐	☐	☐	☐	☐
d)	If a genetic test were available, that could indicate that I was at increased risk of developing cancer, I would want to have such a test	☐	☐	☐	☐	☐

Thank you for completing this questionnaire. Please return in the stamped addressed envelope provided

For the following questions, please indicate how you felt following your brother or sister's heart attack

20) In the months following your brother or sister's heart attack, did you visit your family doctor or a health profession specifically to discuss any concerns you might have had as a consequence of this?

a) Yes ☐ No ☐

	Strongly disagree	Tend to disagree	Neither	Tend to agree	Strongly agree
b) If yes: I found the consultation with my family doctor very helpful	☐	☐	☐	☐	☐

21) Did you seek ***information*** about inherited aspects of heart disease from any of the following sources? (please tick all that apply)

The Internet ☐ Television ☐ Public Library ☐ Books ☐

Newspapers ☐ Magazines ☐ British Heart Foundation Literature ☐

Didn't know where to go ☐

Other health care professionals ☐ Specify_____

	Strongly disagree	Tend to disagree	Neither	Tend to agree	Strongly agree
24) I needed to speak to a health professional in the months after my brother or sister's heart attack	☐	☐	☐	☐	☐
25) Adequate services are available for understanding inherited aspects of heart disease	☐	☐	☐	☐	☐

26) **If you smoke, please answer the following:**

Compared to **before** your brother or sister's heart attack, would you say you smoke more, less or about the same nowadays?

I smoke **much more** nowadays ☐

I smoke **more** nowadays ☐

I smoke **about the same** nowadays ☐

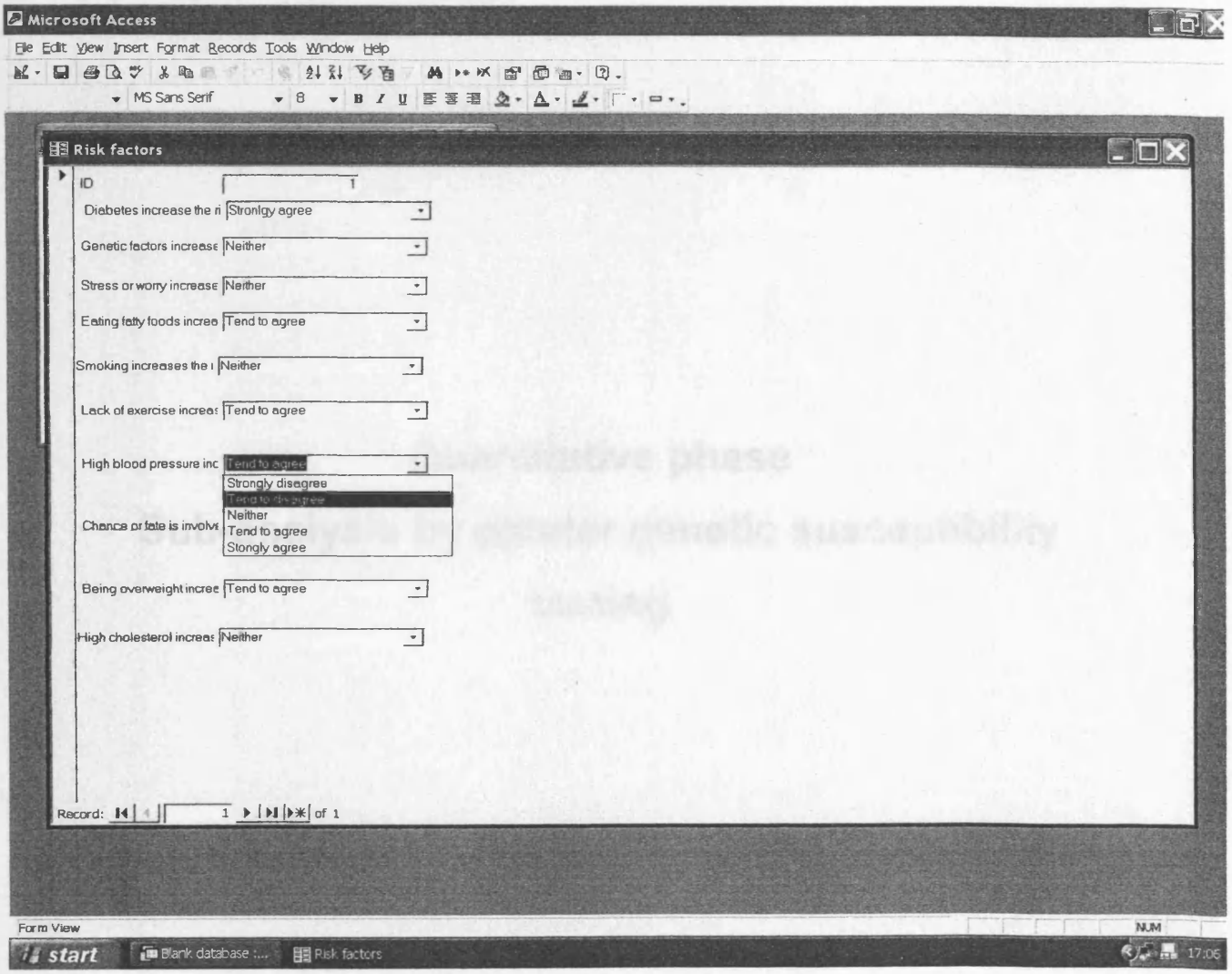
I smoke **less** nowadays ☐

I smoke **much less** nowadays ☐

Thank you for completing this supplementary questionnaire. Please return BOTH questionnaires in the stamped addressed envelope provided

Appendix 28

Quantitative phase
Example of database entry form


Microsoft Access
File Edit View Insert Format Records Tools Window Help
MS Sans Serif
Risk factors
ID
Diabetes increase the ri
Stronlgy agree
Genetic factors increase
Neither
Stress or worry increase
Neither
Eating fatty foods increa
Tend to agree
Smoking increases the r
Neither
Lack of exercise increas
Tend to agree
High blood pressure inc
Tend to agree
Strongly disagree
Neither
Tend to agree
Stongly agree
Chance or fate is involv
Being overweight increa
Tend to agree
High cholesterol increas
Neither
Record:
of 1
Form View
NUM
start
Blank database :...
Risk factors
17:05

Appendix 29

Quantitative phase
Sub-analysis by gender genetic susceptibility testing

Sub-analysis by gender: Views on the possibility of genetic susceptibility testing for common diseases

Heart disease

Males		Strongly Disagree	Tend to disagree	Neither	Tend to agree	Strongly agree	Mean	Difference in mean (95% CI)	p- value*
If a genetic test were available, that could indicate that I was at increased risk of developing heart disease, I would want to have such a test	Unaffected Sibling	0 *(0%)*	1 *(4.3%)*	1 *(4.3%)*	7 *(30.4%)*	14 *(60.9%)*	4.48	0.399 (-0.037 – 0.835)	0.072
	Comparison group	4 *(3.2%)*	9 *(7.1%)*	7 *(5.6%)*	59 *(46.8%)*	47 *(37.3%)*	4.08		

Female		Strongly Disagree	Tend to disagree	Neither	Tend to agree	Strongly agree	Mean	Difference in mean (95% CI)	p- value*
If a genetic test were available, that could indicate that I was at increased risk of developing heart disease, I would want to have such a test	Unaffected Sibling	0 (0%)	5 (13.9%)	1 (2.8%)	15 (41.7%)	15 (41.7%)	4.11	0.066 (-0.534 – 0.665)	0.827
	Comparison group	2 (9.1%)	1 (4.5%)	1 (4.5%)	8 (36.4%)	10 (45.5%)	4.05		

Footnote to tables:

* Numerical values were assigned to the responses, for example, Strongly Disagree being 1 → Strongly Agree being 5

** Therefore, the Mean figure is the mean score on the scale described above

*** Calculated using a T-test

Stroke

Males		Strongly Disagree	Tend to disagree	Neither	Tend to agree	Strongly agree	Mean	Difference in mean (95% CI)	p- value*
If a genetic test were available, that could indicate that I was at increased risk of having a stroke, I would want to have such a test	Unaffected Sibling	0 (0%)	1 (4.3%)	1 (4.3%)	1 (4.3%)	13 (34.8%)	4.43	0.363 (-0.092 – 0.819)	0.117
	Comparison group	5 (4.0%)	9 (7.1%)	8 (6.3%)	54 (42.9%)	50 (39.7%)	4.07		

Females		Strongly Disagree	Tend to disagree	Neither	Tend to agree	Strongly agree	Mean	Difference in mean (95% CI)	p- value*
If a genetic test were available, that could indicate that I was at increased risk of having a stroke, I would want to have such a test	Unaffected Sibling	0 (0%)	4 (11.1%)	1 (2.8%)	17 (47.2%)	14 (38.9%)	4.14	0.048 (-0.532 – 0.628	0.869
	Comparison group	2 (9.1%)	1 (4.5%)	1 (4.5%)	7 (31.8%)	11 (50%)	4.09		

Cancer

Males		Strongly Disagree	Tend to disagree	Neither	Tend to agree	Strongly agree	Mean	Difference in mean (95% CI)	p- value*
If a genetic test were available, that could indicate that I was at increased risk of developing cancer, I would want to have such a test	Unaffected Sibling	0 (0%)	1 (4.3%)	1 (4.3%)	7 (30.4%)	14 (60.9%)	4.48	0.415 (-0.055 – 0.884)	0.083
	Comparison group	6 (4.8%)	9 (7.1%)	7 (5.6%)	53 (42.1%)	51 (40.5%)	4.06		

Females		Strongly Disagree	Tend to disagree	Neither	Tend to agree	Strongly agree	Mean	Difference in mean (95% CI)	p- value*
If a genetic test were available, that could indicate that I was at increased risk of developing cancer, I would want to have such a test	Unaffected Sibling	0 (0%)	4 (11.1%)	0 (0%)	14 (38.9%)	18 (50.0%)	4.28	0.187 (-0.415 – 0.789)	0.537
	Comparison group	2 (9.1%)	1 (4.5%)	3 (13.6%)	3 (13.6%)	13 (59.1%)	4.09		

Appendix 30

Abstract, programme and presentation, Leicestershire Research Day, 2002

Abstract for submission to Leicestershire Research Day

For consideration in the category of: **Cardiovascular Disease & Stroke**

Title

Having a sibling who has experienced a premature myocardial infarction (MI): what are the risk perceptions of unaffected siblings?

Authors

Stribling J, Young B, Lambert PC, Samani NJ

Background

Advances in molecular technology offer the possibility of susceptibility testing in the future for common multi-factorial conditions. This qualitative study investigated the accounts of people whose siblings had experienced a premature MI, to explore their concepts of familial health risks, their beliefs about attenuation of these risks and their views about future genetic testing.

Methods

Semi-structured interviews with 20 unaffected individuals (aged 30-58), whose siblings had experienced an MI (<50 years), and had participated in a previous study investigating genetic and thrombotic aspects of CHD. Quota sampling was used to reflect the social class of the previous study population. Interviews were tape-recorded and transcribed verbatim. Data analysis adopted the constant comparison approach within a grounded theory framework.

Results

Participants in this study expressed a sense of detachment from their siblings' MI, indicating that their siblings' heart problems had little or no significance or relevance for their judgements of own risk status. Participants highlighted their siblings' lifestyle (e.g. smoking, dietary habits, stress) or chance, rather than inheritance as an explanation of their siblings' heart problems. Amongst those participants who had consulted their GP for advice following their siblings' MI, some had been investigated, but many felt that their concerns had been dismissed or trivialised, often reinforcing their existing feelings of detachment. Participants viewed genetic tests as fundamentally different from other screening procedures, and some were highly resistant to the future prospect of screening for genetic risk factors for heart disease.

Discussion

Identifying individuals at increased risk of developing CHD is a key part of the current health policy and targeted prevention strategies have the potential to greatly improve preventative health care as molecular research in this field advances. Our study suggests that individuals who have a greater potential risk of inherited heart disease do not perceive their risk status as elevated, and have important reservations about the future of predictive genetic testing that may limit the acceptability of these new technologies.

Leicestershire Research Day 2002
Friday 14th June
Leicester City Football Club

The following are the titles and authors of the research projects that will be presented at the Leicestershire Research Day 2002.

Cardiovascular Disease and Stroke category

13.40 **Dr A James P Clover, Clinical Research Fellow** – 'Subcontractile electrical stimulation causes increased capillary density in stable claudicants – a potential new model for inducing therapeutic angiogenesis'

14.05 **Dr Kathryn Gill, Clinical Research Fellow** – 'New approaches to neovascularisation using endothelial progenitor cells'

14.30 **Dr David A Payne, Research Fellow** – 'Clopidogrel reduces thromboembolism after carotid endarterectomy: a randomised trial'

14.55 **Mr Julian Stribling, Research Nurse** – 'Having a sibling who has experienced a premature myocardial infarction (MI): what are the risk perceptions of unaffected siblings?'

Cancer and Open category

13.40 **Dr Ruth H Green, SpR/Research Fellow** – 'Reduced asthma exacerbations with a management strategy directed at normalising the sputum eosinophil count. A randomised comparison with traditional management'

14.05 **Mr C Arun, Clinical Research Fellow** – 'Prognostic significance of elevated endothelin -1 levels in patients with colorectal cancer'

14.30 **Dr Surinder S Birring, Clinical Research Fellow** – 'Development of a new health related quality of life measure for patients with chronic cough'

14.55 **Dr Sundar Santhanam, Specialist Registrar** – 'Tumours of the Testis, a 15 year experience'

Mental Health category

13.40 **Dr Mohammed Al-Uzri, Clinical Lecturer** – 'Memory impairment in schizophrenia'

14.05 **Dr Michael Dennis, Senior Lecturer/Honorary Consultant Psychiatrist** – 'Suicide and deliberate self-harm in the elderly: an examination of risk factors with implications for prevention'

14.30 **Dr Heather Dipple, Consultant in Rehabilitation Psychiatry** – 'The experience of motherhood in women with severe and enduring mental illness'

14.55 **Mr Chris Stowers, Consultant Clinical Psychologist/Head of Rehabilitation Psychology** – 'The use of seclusion in mental health settings: implications for nurse/patient relationships'

Having a sibling who has experienced a premature myocardial infarction (MI) : what are the risk perceptions of unaffected siblings?

Stribling J[1]
Young B[2]
Lambert PC[2]
Samani NJ[1]
14/06/2002

Division of Cardiology[1] & Department of Epidemiology & Public Health[2], University of Leicester

Context of study

1 **CHD is a key part of Government Health Policy**

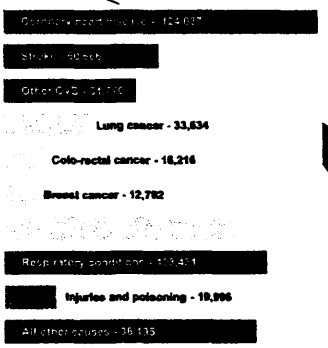
"*General Practitioners and primary care teams should*

Identify all people at significant risk of cardiovascular disease but who have not developed symptoms and offer them appropriate advice and treatment to reduce their risks" *(Standard 4, NSF, 2000)*

2 **Rapid advances in molecular technology**

The Human Genome Project, and other research offer the possibility of susceptibility testing for common multi-factorial conditions in the future

Deaths by cause in the UK, 2000



Source: BHF website (ONS, 200

Premature MI and Family History

Risk Factor	Risk Ratio
CHD – 1° relative <55yrs	9.4
Cholesterol > 6.5 mmol/L	4
Smoking >1/2 pack per day	4.0
Diabetes	2.7
Hypertension (BP >160 100)	1.8
Triglycerides >5.0 mmol/L	1.51

(Nora, 1983)

Objectives of CHD Perception Study

To characterise the risk perceptions and beliefs of individuals currently unaffected who have a sibling who has experienced an before the age of 50

Methods

- Semi- structured interviews
- Unaffected individuals whose siblings experienced a premature MI
- Affected sibling had participated in previous (PRAMIS)
- Recruited via the PRAMIS cases

PRAMIS

	Cases	Controls
Number	200	200
Male: Female Ratio	87:13	87:13
Mean age	47.3	47.2
Mean event age	42.7	-

(Singh et al., 2001)

Current Study Methods

Semi-structured interviews, using a prompt list of topics:

- Beliefs about causes of siblings' MI
- Beliefs about personal vulnerability
- Views about genetic research and the possibility of future genetic testing

All interviews audio-taped and transcribed verbatim

Analysis based on the constant comparison method, assisted by QSR NUD.IST software

Participants

20 out of 63 approached lived within travelling distance or agreed to participate

White

10 male, 10 female

Aged 30 – 58, mean age = 47

From a range of different occupational groups

Findings: Main Themes Identified

1. Causal attributions for siblings' MI
2. Ideas about personal risk
3. Hereditary aspects
4. The nature of CHD
5. Resources
6. Reactions

1 Causal attributions for siblings' MI

Risk Factors	Most participants identified risk factors for CHD, but often required prompting to consider family history
Stress	Participants frequently talked about the perceived role of stress in CHD
Cultural habits	Some talked about the role of cultural habits, especially in relation to diet
Fate	The role of fate / fatalism and being "chosen" was often discussed

2 Ideas about personal risk

Characterisation of self	Feelings of personal vulnerability and acknowledging individual risk
Moderating perceived risk	Motivation for moderating personal risk
Barriers to moderating perceived risk	Expressions of difficulty sustaining "healthy" lifestyle, concern about conflicting information
Detachment	The idea that their siblings' MI is of little or no consequence to their health

Detachment

"I've never thought about it. Just sort of put it to the back of your mind...you always think it will never happen to me, don't you?"

(Female, 55, brother had an MI at 51, father died a[t] 68)

"Whatever happened to him was totally different to me because I'm a different person"

(Male 53, brother had MI at 3[1])

3 Hereditary aspects (a)

Inheritance / Genetics

Fundamental nature of genetics	"Having a genetic problem you tend to think that's a root problem with you" *(Male 43, sister had an MI at 47)*
Seriousness	Genetic problems, or conditions with genetic component were often di[s]cussed in terms of being a serious or "nas[t]y" conditions
Public interest in genetic research	A range of views were expressed about general interest and anxiety about genetics

3 Hereditary aspects (b)

Genetic Testing

The nature of genetic tests	"If somebody mentions some genetic test ... in my perception it jumps into a different league" *(Male 43, [s]ister had an MI at 47)*
Consequences	Participants seemed aware of consequences of genetic testing: insurance / pressure on family
Views about susceptibility testing	Some strong views about the future of genetic testing were expressed

Views about susceptibility testing

" No, I would not want to know that!... If somebody told me I'd got a high risk of having this I'd say "Oh ****- thank you very much – now how do I live my life?", I'm 53 and now you've told me I'm 90% gonna die from heart problems"

(Male 53, brother had [MI] at [age] 50)

"If I can't do anything about it, I'd sooner not know... bury my head in the sand. What you don't know, can't hurt you"

(Male, 47, brother had MI at 4[8])

"Yes, I would have a test...I don't think it's a bad thing. You know, you can sort of do something about it, or avoid it, or at least hold it back."

(Male, 47, brother had [an] MI at 48)

4 The nature of CHD

Life event	Having an MI was seen as a major life event (for their sibling), and many participants talked about the psychological impact that it had had on their sibling
Complacency / indifference	MI was often discussed in terms of being something you recover from, as a favourable mode of death (particularly in relation to cancer)

"One good heart attack and you've gone, that's the end of it"

(Female 55, brother MI at 51)

5 Resources

Suggestions for information — Some novel ideas for health information were expressed

Experience of research — The process of being i... seemed "therapeutic" for

Experiences with HCP following siblings' event

Support networks

Experiences with health care professionals following siblings' MI

"...my doctor ... at the time didn't seem to be all that interested...to be truthful it was almost li... I was asking him something totally pointless"

(Male 32, sister had an MI

"The practice nurse asked "what are you having thi... done for?" [cholesterol], so I thought I'm all right, I'll b... all right... so why bother?"

(Male, 34, brother had an MI at 3

Support networks

"It would be nice to be able to go somewhere or pick the 'phone up and talk to somebody...you need input from people who know, from professional people...[but] your GP hasn't got time, nor has your pr...e nurse"

(Female, 58, brother ha... t 50)

"I wanted to sit down and talk to someone about that, you know, but I can't go to my GP."

(Male, 47, brother had an MI at 48)

---

6 Reactions

Surprise / shock — Most participants expressed shock and surprise at a seemingly young healthy person having an MI

Consulting behaviour — Barriers to consulting the GP include... too busy and the perception that it would ... of time

Lifestyle changes — Some, had however, made positive change... their lifestyle as a consequence of siblings'

Consulting Behaviour

"[If I said to my GP] "now look, my brothers had a heart attack, I'd like to be checked over" "Why?" would be what I think I would get...it seems as if the doct... thinks that I'm trying to convince myself that I'm going t... e one – which I'm not! I would be saying, you know, I ju... na make sure I'm OK"

(Female 51, brother had an MI

"Something more important always used to crop up..."

(Male 43, sister had an MI at 47)

Lifestyle changes

"Yes, I mean ... with my brother, you know ...you've just got to look after yourself"

(Male, 30, brother had ... at 41)

"I was quite keen on starting to keep fit and [my siste... MI] has become an added motivation to keep fit"

(Male 43, sister had an MI at 47)

4

Conclusions

- **Barriers to consulting: siblings' MI did not warrant a visit to GP**
- **Those who did consult felt their GP was un[illegible] or uninterested**
- **Beliefs about unalterable nature of genetic factor[illegible]**
- **Varying beliefs about personal vulnerability**

Clinical Implications

Unaffected siblings of MI patients are an important and easily identifiable group. However, they are often over looked in primary prevention *(Hengstenberg et al, 200[illegible])*

Desire for a support programme among some unaf[illegible] siblings– could this be linked with cardiac rehabilitati[illegible]

Modern technologies need to be acceptable to consumers health care – how acceptable will future genetic tests for C[illegible] be?

Next Phase of the study

A more systematic analysis of this data is required

Phase 2 of the study, a postal questionnaire to qu[illegible]ome of the findings from the interviews. Additionally, thi[illegible] have a comparative element, by including the control[illegible] from PRAMIS

Acknowledgements

Dr. Ravi Singh – PRAMIS study

Link-Up, patient support charity at Glenfield Hospi[illegible]

Appendix 31

Abstract, programme and presentation, British Psychological Society, 2002

Abstract British Psychological Society

Stribling J, Young B, Lambert P, Samani NJ

Title

Health perceptions of unaffected individuals whose siblings have experienced a premature myocardial infarction (MI).

Purpose

To explore the health perceptions of people whose siblings have experienced a premature MI, to ascertain whether this affects health-related behaviour, and to assess these individuals readiness to accept the future of predictive genetic testing.

Methods

Semi-structured interviews with 20 unaffected individuals (aged 30-58), whose siblings had experienced an MI (<55 years), and had participated in a previous study investigating genetic and thrombotic aspects of CHD. Quota sampling was used to reflect the social class mix from where the population was drawn. Interviews were tape-recorded and transcribed verbatim. Data analysis adopted the constant comparison approach within a grounded theory framework.

Findings

The main themes emerging from the interviews were: attributions believed to cause their siblings heart attacks; ideas about moderating their individual risk; an array of ideas about hereditary aspects of MI and the prospect of future genetic testing; ideas about the nature of CHD (particularly in relation to fears of cancer); ideas about resources (experiences with health care professionals, suggestions for support and health information); and reactions to siblings MI. Some gender differences were noted, particularly in relation to suggestions for support networks.

Conclusions

Identifying individuals at increased risk of developing CHD is a key part of the NSF, and targeting prevention strategies will become a reality as molecular research in this field advances. However, there seemed to be little acknowledgement by these people of their own risk, and a very mixed attitude to the future of predictive testing. This work will influence the design of a postal questionnaire.

For Abstract book email:
H.L.Bekker@leeds.ac.uk

The British Psychological Society

Division of Health Psychology Annual Conference

Sheffield 4th-6th September 2002

"FACILITATING HEALTHIER SOCIETIES"

ACADEMIC PROGRAMME

Co-Hosted by Sheffield Hallam University & the University of Sheffield.

Location Academic Programme: City Campus, Sheffield Hallam University.

Website: www.ardenarmitage.supanet.com/DHP2002.htm

Friday 6th September: morning.

	Pennine LT (follow BLUE) ***Themed papers - Illness perceptions.*** Chair - *Dick Eiser*	**Peak LT (follow GREEN)** ***Symposium: Paediatric Cancer.*** Convenor & Discussant – *Christine Eiser.*	**Norfolk 310 (follow ORANGE)** ***Symposium: Interpretative phenomenological analysis (IPA) and pain research.*** Convenor – *Jonathan Smith* Discussant – *Sandra Horn*
09:00-09:20	Evidence for the discriminant validity of illness representation dimensions across a number of studies *Martin Hagger, S Orbell*	Improving survival rates in children with cancer: the role of physical exercise. *David Johnson, A Robertson*	Pain and the self *Mike Osborn & J ASmith*
09:20-09:40	The roles of illness perceptions, coping and social supporting the functioning of people suffering from Rheumatoid Arthritis *S Tomlinson, D Stewart, J Howard & Suresh*	The effect of parenting on quality of life of children with cancer. *Veronica Greco, C Eiser & J R Eiser*	Pain and age *Vanessa Trowell & M Levine*
09:40-10:00	The roles of illness beliefs, treatment beliefs and perceived severity of symptoms in explaining distress of cancer patients during chemotherapy treatment *Ingela Thune-Boyle, L B Myers & S Newman*	Distress in children undergoing stem cell transplantation and their parents: impact of complimentary health promotions interventions. *Sean Phipps, M Dunavant, S Lensing, S Rai,*	Pain and relationships *Barry Mason*
10:00-10:20	Adjustment, illness representations and coping in men with lower urinary tract symptoms *L Glover, K Gannon, J McLoughlin & M Emberton*	Parenting in a crisis: conceptualising mothers of children with cancer. *Bridget Young, M Dixon-Woods, M Findlay, D Heney*	Pain and professionals *Sarah Dean, S Payne & J Weinman*
10:20-10:40	Illness representations and well-being in newly diagnosed & long-standing patients with diabetes *Alison Weardon, C Paschalides, R Dunkerley, C Dickens, C Bundy & R Davies*	Discussion	Discussion
Coffee 10:40-11:00 Atrium			
	Pennine LT (follow BLUE) ***Individual papers*** Chair - *Sarah Grogan*	**Peak LT (follow GREEN)** ***Individual papers*** Chair - *Paschal Sheeran*	**Norfolk 310 (follow ORANGE)** ***Themed Papers - IPA*** Chair - *Jonathan Smith*
11:00-11:20	Size matters: fallacies about men presenting atypical genito-sexual development *Paul Chadwick, K L M Liao, M Boyle & G Conway*	Depression following stroke: a cognitive enquiry *Ellen Townend, M Sharpe, D Tinson & J Kwan*	*Men and women's experience of Chlamydia testing in a sexual health clinic.* *Jane Darroch, L B Myers & J Cassell*
11:20-11:40	"It's hard being a man": patterns of self-referral in men with symptoms of prostate disease *Susan Hale, S Willott & S Grogan*	Predicting activity limitations: cognition and emotion are independent predictors of exercise and fitness in patients with coronary artery disease. *Derek Johnston & Marie Johnston.*	Having a sibling who has experienced a premature myocardial infarction (MI): what are their health and risk perceptions? *Julian Stribling, B Young, P Lambert & N Samani*
KEYNOTE Professor Robert Plomin 11:45-12:30 Pennine LT (BLUE)			
Closing Ceremony & Trailer for Next Year's Conference at Staffordshire University 12:30 – 13:00 Pennine LT (BLUE)			
Lunch 13:00-14:00 Atrium			

Having a sibling who has experienced a premature myocardial infarction (MI) : what are the risk perceptions of unaffected siblings?

Stribling J[1]
Young B [3]
Lambert PC[2]
Samani NJ[1]
06/09/2002

Division of Cardiology[1] & Department of Epidemiology & Public Health[2], University of Leicest
Department of Psychology, University of Hull[3]

Context of study

1 **CHD is a key part of Government Health Policy**

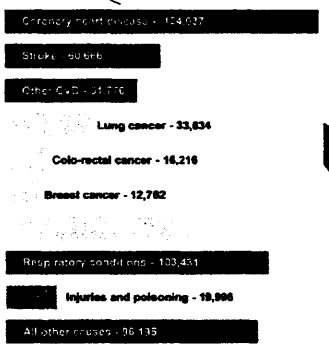
"*General Practitioners and primary care teams should*

Identify all people at significant risk of cardiovascular disease but who have not developed symptoms and offer them appropriate advice and treatment to reduce their risks" *(Standard 4, NSF, 2000)*

2 **Rapid advances in molecular technology**

The Human Genome Project, and other research offer the possibility of susceptibility testing for common multi-factorial conditions in the future

Deaths by cause in the UK, 2000



Source: BHF website (ONS, 200

Premature MI and Family History

Risk Factor	Risk Ratio
CHD – 1° relative <55yrs	4
Cholesterol > 6.5 mmol/L	4.
Smoking >1/2 pack per day	4.0
Diabetes	2.7
Hypertension (BP >160 / 100)	1.8
Triglycerides >5.0 mmol/L	1.51

(Nora, 1983)

Objectives of CHD Perception Study

To characterise the risk perceptions and beliefs of individuals currently unaffected who have a sibling who has experienced an before the age of 50

Methods

- Semi- structured interviews
- Unaffected individuals whose siblings experienced a premature MI
- Affected sibling had participated in previous (PRAMIS)
- Recruited via the PRAMIS cases

PRAMIS

	Cases	Controls
Number	200	200
Male: Female Ratio	87:13	87:13
Mean age	47.3	47.2
Mean event age	42.7	-

(Singh et al., 2001)

Current Study Methods

Semi-structured interviews, using a prompt list of topics:

- Beliefs about causes of siblings' MI
- Beliefs about personal vulnerability
- Views about genetic research and the possibility of future genetic testing

All interviews audio-taped and transcribed verbatim

Analysis based on the constant comparison method, assisted by QSR NUD.IST software

Participants

20 out of 63 approached lived within travelling distance or agreed to participate

White

10 male, 10 female

Aged 30 – 58, mean age = 47

From a range of different occupational groups

Findings: Main Themes Identified

1. **Causal attributions for siblings' MI**
2. **Ideas about personal risk**
3. **Hereditary aspects**
4. **The nature of CHD**
5. **Resources**
6. **Reactions**

1 Causal attributions for siblings' MI

Risk Factors	Most participants required prompting to consider family history
Stress	Participants frequently talked about the perceived role of stress in CHD
Fate	The role of fate / fatalism and being "chosen" was often discussed

2 Ideas about personal risk

Detachment	The idea that the siblings' MI was of little or no consequence to their health

Detachment

"I've never thought about it. Just sort of put it to the back of your mind...you always think it will never happen to me, don't you?"

(Female, 55, brother had an MI at 51, father died [illegible])

"Whatever happened to him was totally different to me because I'm a different person"

(Male 53, brother had MI at 30)

3 Hereditary aspects (a)

Inheritance / Genetics

Fundamental nature of genetics	"Having a genetic problem you tend to think that's a root problem with you" *(Male 43, sister had an MI at 47)*
Seriousness	Genetic problems, or conditions with genetic component were often discussed in terms of being a serious or "nasty" conditions

3 Hereditary aspects (b)

Genetic Testing

The nature of genetic tests	"If somebody mentions some genetic test ... in my perception it jumps into a different league" *(Male 43, [illegible] had an MI at 47)*
Consequences	Participants seemed aware of consequences of genetic testing: insurance / pressure on family
Views about susceptibility testing	Some strong views about the future of genetic testing were expressed

Views about susceptibility testing

" No, I would not want to know that!... If somebody told me I'd got a high risk of having this I'd say "Oh ****- thank you very much – now how do I live my life?", I'm [illegible]3 and now you've told me I'm 90% gonna die from heart problems"

(Male 53, brother had [illegible])

"If I can't do anything about it, I'd sooner not know... but my head in the sand. What you don't know, can't hurt you"

(Male, 47, brother had MI at 4[illegible])

"Yes, I would have a test...I don't think it's a bad thing. You know, you can sort of do something about it, or avoid it, or at least hold it back."

(Male, 47, brother had [illegible]MI at 48)

4 The nature of CHD

Life event	Having an MI was seen as a major life event (for their sibling), and many participants talked about the psychological impact that it had had on their sibling
Complacency / indifference	MI was often discussed in terms of being something you recover from, as a favourable mode of death (compared with cancer)

"One good heart attack and you've gone, that's the end of it"

(Female 55, brother MI at 51)

5 Resources

Suggestions for information	Some novel ideas for health information were expressed
Experience of research	The process of being interviewed seemed "therapeutic" for
Experiences with HCP following siblings' event	
Support networks	

Experiences with health care professionals following siblings' MI

"...my doctor ...at the time didn't seem to be all that interested...to be truthful it was almost like I was asking him something totally pointless"

(Male 32, sister had an MI

"The practice nurse asked "what are you having this done for?" [cholesterol], so I thought I'm all right, I'll b all right... so why bother?"

(Male, 34, brother had an MI at 36)

Support networks

"It would be nice to be able to go somewhere or pick the 'phone up and talk to somebody...you need input from people who know, from professional people...[but] your GP hasn't got time, nor has your practice nurse"

(Female, 58, brother had t 50)

"I wanted to sit down and talk to someone about that, you know, but I can't go to my GP."

(Male, 47, brother had an MI at 48)

6 Reactions

Surprise / shock	Most participants expressed shock and surprise at a seemingly young healthy person having an MI
Consulting behaviour	Barriers to consulting the GP include busy and the perception that it would b of time
Lifestyle changes	Some had, however, made positive change their lifestyle as a consequence of siblings'

Consulting Behaviour

"[If I said to my GP] "now look, my brother's had a heart attack, I'd like to be checked over" "Why?" would be what I think I would get...it seems as if the doctor thinks that I'm trying to convince myself that I'm going to one – which I'm not! I would be saying, you know, I ju na make sure I'm OK"

(Female 51, brother had an MI

"Something more important always used to crop up..."

(Male 43, sister had an MI at 47)

Lifestyle changes

"Yes, I mean ... with my brother, you know ...you've just got to look after yourself"

(Male, 30, brother had a t 41)

"I was quite keen on starting to keep fit and [my siste MI] has become an added motivation to keep fit"

(Male 43, sister had an MI at 47)

Conclusions

- **Barriers to consulting: siblings' MI did not warrant a visit to GP**
- **Those who did consult felt their GP was un[illegible] or uninterested**
- **Beliefs about unalterable nature of genetic facto[illegible]**
- **Varying beliefs about personal vulnerability**

Clinical Implications

Unaffected siblings of MI patients are an important and easily identifiable group. However, they are often over looked in primary prevention *(Hengstenberg et al, 200[illegible])*

Desire for a support programme among some unaff[illegible] siblings– could this be linked with cardiac rehabilitatio[illegible]

Modern technologies need to be acceptable to consumers[illegible] health care – how acceptable will future genetic tests for C[illegible] be?

Acknowledgements

Dr. Ravi Singh – PRAMIS study

Link-Up, patient support charity at Glenfield Hosp[illegible]

Appendix 32

Qualitative abstract submitted for Leicestershire Research Day, 2005

Delivering the Best: A conference to celebrate research across the health communities of LNR
14th July 2005 - Imago Centre - 14th July 2005

Abstract Submission

Title: Missed opportunities in primary prevention: a qualitative study of the experiences of people seeking medical help following their siblings' heart attack

Short Summary of Research (Max 250 words)

Background: Coronary Heart Disease (CHD) is a major health problem. Identifying people at high risk of developing CHD and offering them appropriate advice and treatment to reduce their risk should be undertaken by primary care staff (National Service Framework, Standard 4). Family history of CHD is known to be an independent risk factor, and having a sibling affected with CHD at a young age increases an individual's risk significantly. The experiences of people seeking help from primary care staff with health concerns following their siblings' heart attack were investigated as part of a qualitative study.

Method: Semi-structured interviews with 20 unaffected individuals (aged 30-58), whose siblings had experienced a heart attack (<55 years), and had participated in a previous study investigating genetic aspects of CHD. All interviews were audio taped, transcribed verbatim and analysed using the constant comparison method.

Findings: Some participants expressed no need or desire to visit their GP following their siblings' MI. Others felt inhibited from seeking help because of previous negative experiences. Of those who did visit their GP, many felt that their concerns were dismissed or trivialised, or that they were made to feel as if they wasting the staff time.

Conclusion: Identifying people at high risk of developing CHD, and offering them appropriate advice and treatment to reduce their risk is an important part of primary prevention. This study would suggest that there is considerable room for improvement in this aspect of service provision.

Outcome/ Impact of research

Family history of CHD could be utilised as a method of identifying people at risk of high of developing the disease, rather than being viewed as a non-modifiable risk factor (Yoon et al., 2002).

Yoon PW, Scheuner MT, Peterson-Oehlke KL, Gwinn M, Faucett A, Khoury MJ. Can family history be used as a tool for public health and preventive medicine? *Genetics in Medicine* 2002;**4**:304-10.

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Abstracts should be submitted by 13th May 2005

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Appendix 33

Quantitative abstract submitted for Leicestershire Research Day, 2005

Delivering the Best: A conference to celebrate research across the health communities of LNR
14th July 2005 - Imago Centre - 14th July 2005

Abstract Submission

Title: Risk perceptions and views of genetic research: a comparison between people whose siblings' have had a heart attack, and people where there is no family history of CHD

Short Summary of Research (Max 250 words)

Background: Coronary Heart Disease (CHD) is a leading cause of premature death, and a family history of CHD is an important risk factor. Based on the findings of an earlier qualitative study, a postal questionnaire was developed and sent to participants of a previous case-control study investigating genetic aspects of CHD.

Method: The questionnaire was sent to unaffected siblings of people who had experienced a heart attack under the age of 50 years, and to a comparison group where there was no family history of CHD. The questionnaire explored knowledge about CHD, perceived risk and control, fear of CHD (in relation to other conditions), and views about genetic developments.

Results: Response rate from unaffected siblings was 32.6% (n= 59), and 70.8% (n=148) from the comparison group. Unaffected siblings were more likely to believe that they had a higher chance of developing CHD than the comparison group (p=0.001), and also were more likely to fear CHD than the comparison group (p=0.001). Both groups were equally likely to feel that they could reduce their risk of CHD or cancer. Broadly similar beliefs about genetic technology and the possibility of genetic susceptibility testing were observed in both groups. Only 30% of unaffected siblings sought medical help or reassurance following their siblings' heart attack.

Conclusion: Unaffected siblings feared CHD, and felt that they had a high risk of developing it, but also felt that they could reduce their personal risks of developing CHD

Outcome/ Impact of research

Family history of CHD is an important risk factor and could be utilised as a means of identifying people at risk. How people interpret their risk, will have implications for their health–related behaviour.

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Appendix 34

Letter of acceptance, programme and slides for Leicestershire Research Day, 2005

NHS

Leicestershire Primary Care Research Alliance

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LE1 7PE

Telephone: 0116 295 4080
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1 June 2005

Letter Ref: RA2627

Julian Stribling
Academic Co-ordinator
Eastern Leicester PCT
Prince Philip House
Malabar Road
Leicedster
LE1 2NZ

Dear Julian

Missed opportunities in primary prevention: a qualitative study of the experiences of peple seeking medical help following their siblings' heart attack

Risk perceptions and views of genetic research: a comparison between people whose siblings' have had a heart attack, and people where there is no family history of CHD

Thank you for submitting the above abstracts for the LNR *'Delivering the Best'* Research Conference to take place on the 14th July 2005. The panel has now reviewed all the applications and we are delighted to inform you that both your abstracts have been considered suitable for the conference, but due to the high numbers of quality presentations submitted we are unable to accept both for presentation.

We would like to invite you to either consider combining them into one oral presentation, or to present one orally and submit the other as a poster presentation. In addition it was felt by the review panel that the title of the abstract *'Missed Opportunities in Primary Prevention...'* was subjective and could be misleading. It was therefore suggested that this title be revised.

Oral presentations are to last for 20 minutes, with a short period of 5 minutes for questions at the end. Additionally, we would like you to submit a short biography of the main presenters and any PowerPoint presentations to the LPCRA by Friday 8th July so that we can include these details in the delegate packs.

Posters may be brought on the day, but we would request that someone remain with the poster during intervals to answer any questions raised by the delegates. Accommodation of a range of poster sizes is possible, but we will assume that your poster will be standard A1 size unless otherwise notified. Standard fixation materials will be provided.

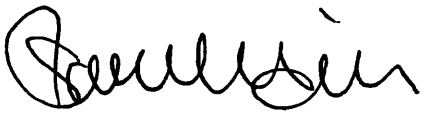
The panel would like to draw presenters attention to the fact that the theme of the conference is 'Dissemination of the outcomes and impacts of research ***on practice'*** and also that the audience will be multi-professional, from across all sectors of healthcare. We would therefore ask that

presentations/posters not only consider the methodology and results of the research, but also discuss the work in context, with the relevance of the research to the health care explained.

I would be grateful if you could write and confirm that you are still willing to present your work and confirm in what format this will take.

Congratulations and we look forward to seeing you on the 14th July 2005.

Yours sincerely
On behalf of the LNR' Delivering the Best' Conference Organising Committee



Sue Palmer Hill, RGN, MSc
R & D Manager
Leicestershire Primary Care Research Alliance

'Delivering the Best' Agenda

TIME	EVENT	TITLE	PRESENTER
0900		REGISTRATION & COFFEE	POSTER VIEWING
0930		Introduction	
0940	Opening Address	**Translational Medicine: At The Heart of Integrated Healthcare**	Prof. Anthony Woodman Chair, Translational Medicine Cranfield University
1010	Presentation 1	**Dual ErbB1/ErbB2 tyrosine kinase inhibition - a potential adjunct to systemic chemotherapy in bladder cancer**	Miss Lynsey A McHugh Clinical Research Fellow Urology Group University of Leicester
1040	COFFEE		POSTER VIEWING
1100	Presentation 2	**Urotensin II in the Prediction of Acute Myocardial Infarction (AMI) and Risk of Major Adverse Cardiac Event (MACE) in Humans**	Dr S Q Khan Clinical Research Fellow Dept of Cardiovascular Sciences University of Leicester
1130	Presentation 3	**To establish whether a pharmacist can improve quality and cost effectiveness of prescribing by reviewing patient medication in care homes**	Mrs Susanna Taylor Prescribing Advisor Leicester City West PCT
1200	Presentation 4	**Choosing and using an NHS Walk-in Centre**	Ms Clare Jackson Research Associate Department of Health Sciences, University of Leicester
1230	Presentation 5	**People whose siblings have had a premature heart attack: their views and experiences in the era of human genomics**	Julian Stribling Academic Co-ordinator Eastern Leicester PCT
1300	LUNCH		POSTER VIEWING
1400	Presentation 6	**Psychological morbidity in patients presenting with non-cardiac chest pain to a Rapid Access Chest pain Clinic**	Dr Noelle Robertson Snr Lecturer in Clinical Psychology University of Leicester
1430	Presentation 7	**Physical aggression towards others in adults with learning disabilities: Prevalence and associated factors**	Ms Freya Tyrer Research Associate University of Leicester
1500	Presentation 8	**Goal setting as a tool for focused dietary management in UK children with Type 1 Diabetes**	Dr Sheridan Waldron Dietetic Manager Leics Nutrition & Dietetic Service
1530	TEA		POSTER VIEWING
1600	Keynote Speaker	**Supporting clinical academic research in the modern NHS**	Dr Jonathan Barratt MRC/DH Clinician Scientist Honorary Consultant Nephrologist Leicester University/LGH
1630		**Summary and Close** **Presentation of the Awards**	

University of Leicester

Eastern Leicester NHS
Primary Care Trust

People whose siblings have had a premature heart attack: their views and experiences in the era of human genomics

Stribling J[1]

Young B[2]

Lambert PC[3]

Samani NJ[4]

14/07/2005

Department of Health Sciences, University of Leicester & Eastern Leicester PCT[1]

Division of Clinical Psychology, University of Liverpool[2]

Department of Health Sciences, University of Leicester[3]

Department of Cardiovascular Sciences, University of Leicester[4]

Context of the study

CHD is a key part of Government Health Policy

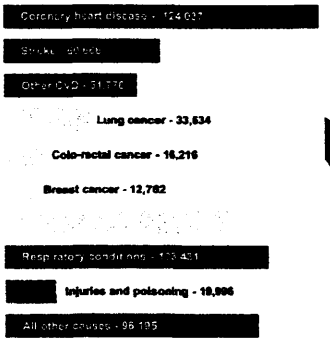
"*General Practitioners and primary care teams should*

Identify all people at significant risk of cardiovascular disease but who have not developed symptoms and offer them appropriate advice and treatment to reduce their risks" *(Standard 4, NSF, 2000)*

Rapid advances in molecular technology

The Human Genome Project, and other research may offer the possibility of susceptibility testing for common multi-factorial conditions in the future

Deaths by cause in the UK, 2000



Source: BHF website (ONS, 200

Premature MI and family history

Risk Factor	Risk Ratio
CHD 1° relative <55yrs	10.4
Cholesterol > 6.5 mmol/L	4.
Smoking >1/2 pack per day	4.0
Diabetes	2.7
Hypertension (BP >160 100)	1.8
Triglycerides >5.0 mmol/L	1.51

(Nora, 1983)

Aims of the study

To examine the health beliefs, risk perceptions, experiences, and attitudes to possible genetic susceptibility testing among individuals currently unaffected by CHD, who have a sibling who has had a heart attack under the age of 50

To utilise the results of this pilot study to develop a postal questionnaire to send to a larger group of unaffected siblings and a comparison group where there is no family history of CHD

Qualitative methods

- 20 semi- structured interviews using a prompt guide:
  - Experiences of, and beliefs about causes of siblings' heart attack
  - Beliefs about personal vulnerability
  - Views about genetic research and the possibility of future genetic susceptibility testing
- Unaffected individuals whose siblings had experienced premature heart attack and had participated in previous study (PRAMIS)
- All interviews audio-taped and transcribed verbatim
- Analysis based on the constant comparison method

Sample source: PRAMIS

	Cases	Controls
Number	209	2
Male: Female Ratio	87:13	87:
Mean age	47.3	47.2
Mean event age	42.7	-

(Singh et al., 2001)

Participants

- 20 unaffected siblings of PRAMIS cases
- White
- 10 male, 10 female
- Aged 30 – 58, mean age = 47
- From a limited range of occupational groups
- Of their affected siblings, 15 were male, and the age had a heart attack ranged from 30-51 years

Findings: main themes identified

1. **The experiences of premature heart attack in a sibling**
2. **Explanations for siblings' heart attac**
3. **Inheritance and genetics**
4. **Experiences with health care professionals**

The experiences of premature heart attack in a sibling

Surprise / shock
Vulnerability
Detachment
Complacency indifference
Beliefs about moderating risk
Barriers to moderating risk
Information and support
Lifestyle changes following sibling's heart attack

Detachment

"I've never thought about it. Just sort of put it to the back of your mind...you always think it will never hap to me, don't you?"

(Female, 55, brother had an MI at 51, father died o 58)

"Whatever happened to him was totally different to me because I'm a different person"

(Male 53, brother had MI at 3

Explanations for siblings' heart attack

Causal attributions
Life event

Inheritance and genetics

Nature of genetics

The nature of genetic testing

Views about susceptibility testing

Public interest in genetic research

Views about susceptibility testing

" No, I would not want to know that!... If somebody told me I'd got a high risk of having this I'd say "Oh ****- thank you very much – now how do I live my life?", I'm 53 and now you've told me I'm 90% gonna die from heart problems"

(Male 53, brother had [illegible])

"If I can't do anything about it, I'd sooner not know... bu my head in the sand. What you don't know, can't hurt yo

(Male, 47, brother had MI at 4

Experiences with health care professionals

A particularly worrying finding of the qualitative phase was the experiences of participants when they visited their GP for an assessment of their personal risk or reassurance regarding their own health – many felt that their concerns were dismissed or trivialised.

"...my doctor ...at the time didn't seem to be all that interested...to be truthful it was almost like... I was asking him something totally pointless"

(Male 32, sister had an MI at 36)

"The practice nurse asked "what are you having the test done for?" [cholesterol], so I thought I'm all right, I'll be all right... so why bother?"

(Male, 34, brother had an MI at 3

Consulting behaviour

"[If I said to my GP] "now look, my brothers had a heart attack, I'd like to be checked over" "Why?" would be what I think I would get...it seems as if the doctor thinks that I'm trying to convince myself that I'm going to have one – which I'm not! I would be saying, you know, I just wanna make sure I'm OK"

(Female 51, brother had an MI

Quantitative methods

- The quantitative phase of the study aimed to quantify some of the findings of the earlier qualitative phase
- A postal questionnaire was developed utilising questions / statements from previously published research and a small number of questions developed specifically for this study
- Sample source: PRAMIS cases and controls, which allowed comparison with a group where there was no family history of CHD

Main questionnaire: sent to all participants

Demographic — Current health status

Risk factors — Knowledge of heart disease

Perception of risk — Disease feared

Attitudes to genetic research

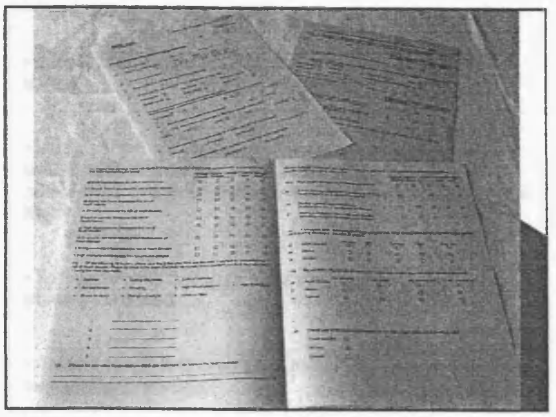
Attitudes to genetic susceptibility testing

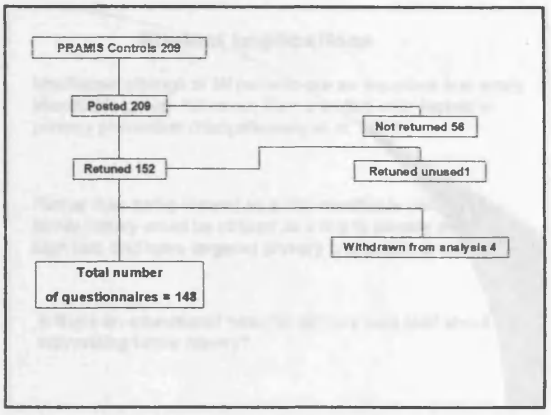
Supplementary questionnaire: unaffected siblings on

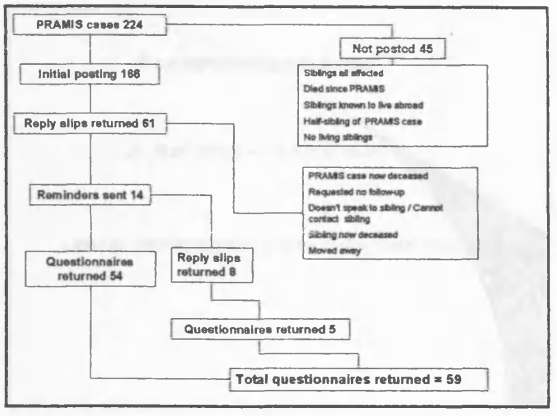
Consultation — Sources of information

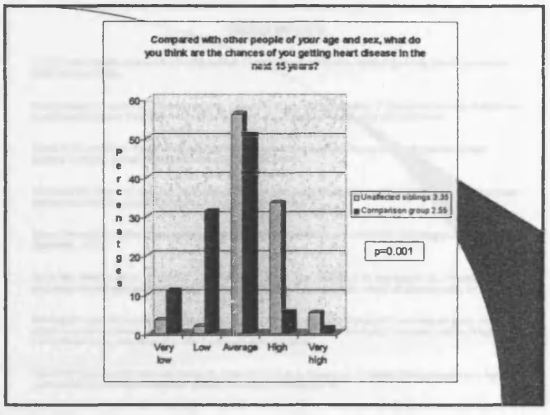
Access to health care professionals / adequacy of service

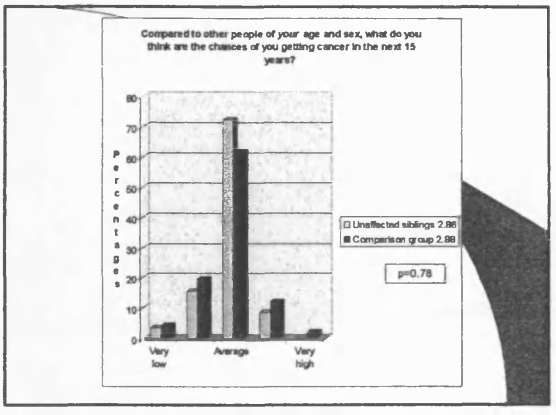
Current smoking behaviour

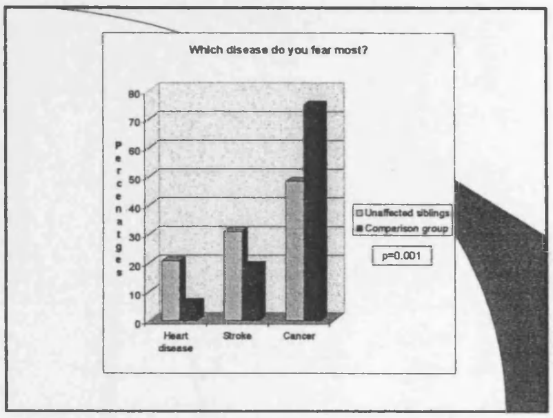












Conclusions

- Integrating qualitative and quantitative methods can be useful
- Participants had varying beliefs about personal risk, but unaffected siblings were significantly more likely than the comparison group to view their risk of developing CHD as high
- Some worrying findings regarding consultation with primary care staff
- A small number of men were highly resistant to genetic susceptibility testing

Clinical Implications

Unaffected siblings of MI patients are an important and easily identifiable group. However, they are often over looked in primary prevention *(Hengstenberg et al, 2001)*

Rather than being viewed as a non-modifiable risk family history could be utilised as a tool to identify pe high risk, and have targeted primary prevention strate

Is there an educational need for primary care staff about interpreting family history?

Acknowledgements

Dr. Ravi Singh – PRAMIS study

Link-Up, patient support charity at Glenfield Hosp

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Appendix 35

Best Conference Presentation Award for Leicestershire Research Day, 14/07/2005

The Healthcare Communities of the Leicestershire, Northamptonshire & Rutland SHA

Delivering the Best
imago Conference Centre
14 July 2005

Best Conference Presentation Award

This award is sponsored by Trent RDSU
for research education including courses, books and materials

Awarded to

.....Julian Stribling.....

A one-day conference celebrating the contribution research plays in delivering excellence in care

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