The Risky Self

How people perceive and respond to being at risk for diabetes and cardiovascular disease;

the role of genetic risk information and self-concept
The study project presented in this thesis was conducted within the EMGO+ Institute for Health and Care Research, Department of Public and Occupational Health of the VU Medical Center in Amsterdam. The EMGO+ Institute participates in the Netherlands School of Primary Care Research which was reacknowledged in 2005 by the Royal Netherlands Academy of Arts and Sciences. In 2010 the EMGO+ Institute received an excellent review by the international external evaluation committee of all Dutch university research, as organized by the universities in the Netherlands.

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The Risky Self

How people perceive and respond to being at risk for diabetes and cardiovascular disease; the role of genetic risk information and self-concept

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ter verkrijging van de graad Doctor aan de Vrije Universiteit Amsterdam, op gezag van de rector magnificus prof. dr. L.M. Bouter, in het openbaar te verdedigen ten overstaan van de promotiecommissie van de faculteit der Geneeskunde op woensdag 1 juni 2011 om 15.45 uur in de aula van de universiteit, De Boelelaan 1105

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Chapter 1

Introduction
General Introduction

Many different risks threaten our health. Some risks are inborn, originating from people’s genetic make-up. Variations or mutations that occur in the DNA sequence of one more genes can cause or predispose for diseases. Unhealthy behaviour such as smoking and eating high-fat food can also increase susceptibility to diseases. Genetic and behavioural factors often act together to cause the expression of a disease. In the expression of a monogenic condition, caused by a mutation in one single gene, behavioural factors often play a minor role. More often diseases such as diabetes and cardiovascular disease are multifactorial in nature, resulting from variations in multiple genes interacting with behavioural and environmental factors, none of which on its own would be likely to cause the disease (Holtzman, 2000). Because members of families share the same variations of genes that can occur, have common behaviour and are exposed to the same environmental influences, multifactorial diseases often show a clustering pattern within families (Scheuner et al., 1997). A family history of disease is therefore associated with an increased susceptibility to multifactorial diseases (Valdez et al., 2010).

With the rapid developments in the field of health screening technology, people are increasingly confronted with new information about their susceptibility to a range of diseases, including genetic risk information based on DNA testing or family history. Whether or not people benefit from these developments may not only depend on the changeability of risks (i.e. preventive options) but also on how people perceive themselves in relation to genetic and behavioural risks (i.e. the “Risky Self”). Little is known about people’s ideas and theories on genetic and behavioural factors that increase their susceptibility to diseases and how these interact with beliefs about who they are as a person. The central objective of this thesis was to gain understanding on how people perceive and respond to being at risk for two common multifactorial diseases, type 2 diabetes and cardiovascular disease.

This introductory chapter first gives a brief overview of the prevalence, prevention and risk assessment of type 2 diabetes and cardiovascular disease. This is followed by an outline of the key dimensions of lay representations (or mental models) of being at risk. Subsequently, the mental model will be discussed in relation to the self-concept, i.e. people’s ideas about who they are as a person and in relation to preventive behaviour. The chapter will conclude with a description of the research questions and methods, and an outline of this thesis.

Type 2 diabetes and cardiovascular diseases

The incidence of type 2 diabetes (T2D) and cardiovascular diseases (CVD) is a major health problem throughout the world (WHO, 2005). T2D is a chronic metabolic disorder that is characterized by high blood glucose in the context of insulin resistance and insulin deficiency leading to micro- and macro
vascular damage. Severe complications can result from improperly managed T2D or late diagnosis, including slow healing wounds, renal failure, amputations and blindness. It is also a major risk factor for developing CVD (Smith, 2007). In the Netherlands, it is estimated that approximately 900,000 people have T2D, including 30% of undiagnosed cases (RIVM, 2010). CVD are a class of diseases that involve arteries or veins (e.g. heart attack and stroke) usually caused by arteriosclerosis. In the Netherlands, over a million people suffer from CVD and 40,000 people die each year of CVD (RIVM, 2010).

Guidelines for the prevention of CVD and T2D recommend the appropriate use of medication and adoption of a healthy lifestyle such as sufficient levels of physical activity in individuals with increased risk (EASD, 2007). Clinical trial evidence shows that lifestyle modifications (e.g. eating more healthily and increased physical activity) can effectively reduce the risk of T2D (Knowler et al., 2002; Lindstrom et al., 2006) and CVD (Holme et al., 2006). However, it has become clear that general health education programs aimed at the whole population have limited effect (Kreuter, Strecher & Glassman, 1999; Noar, Benac & Harris, 2007). One way of increasing the effectiveness of these programs is to target interventions at individuals who are at increased risk of developing these diseases.

The identification of individuals at increased risk for T2D and CVD risk is traditionally based on the presence of multiple risk factors such as advanced age, raised cholesterol levels, high blood pressure, (central) obesity and being a smoker (Hippisley-Cox et al., 2009; D’Agostino et al., 2004; Conroy et al., 2003). The growing knowledge of the genetic component of multifactorial diseases and advances in genetic testing have opened the door for new possibilities for risk assessment. At present, several genetic tests for monogenic disorders associated with CVD are available. Because the presence of such a monogenic disorder often confers a significant disease risk, predictive testing for these mutations may be very informative, not only for the index person but for relatives as well. The most prevalent monogenic disorder associated with a high risk of CVD is Familial Hypercholesterolemia (FH). FH is an autosomal dominant inherited monogenic disease characterized by an impaired metabolism of low-density lipoprotein cholesterol (caused by a mutation of the LDL-receptor gene) and an increased risk of developing premature CVD. In Western countries, the estimated frequency of FH is 1 in 400 - 500 people (Austin et al., 2004). People diagnosed with FH can reduce their risk of developing CVD through the use of cholesterol-lowering medication and the adoption of a healthy lifestyle (not smoking, healthy diet and sufficient physical activity) (Civeira, 2004). There are also some rare forms of diabetes that are caused by a single gene mutation, of which maturity-onset diabetes of the young (MODY) is the most common. However, these forms are not associated with risk factors for the more common T2D that predominantly starts at middle age (Dora, Patti & Kahn, 2008; Vaxillaire & Froguel, 2008). Most forms of T2D and CVD result from
variations in multiple genes interacting with other risk factors such as overweight and smoking. New low-risk gene variants for T2D (Voight et al, 2010) and CVD (Arking & Chakravarti, 2009) are discovered rapidly and screening technology makes it possible to screen large populations for multiple low-risk gene variants (Hirshhorn et al., 2005). There are several commercial companies offering genetic susceptibility testing for these variants associated with T2D and CVD through the Internet (Hogarth, Javitt & Meltzer, 2008). However, these tests currently have limited value in predicting disease risk (Janssens et al., 2008), and as such can not (yet) be used in clinical practice.

Another important source of genetic risk information to which most people have access is the health status of their close relatives (family history). Compared to genetic risk profiling, family history information has the advantage that it not only reflects the consequences of multiple genetic factors, but also captures the complex interactions between genetic and behavioural factors, and may therefore be a better determinant of disease risk than genetic profiling (Hall, Matthews & Morley, 2010).

**Lay representations or mental model of being at risk**

How people perceive and respond to their risk may depend on how they process and evaluate information about their risk. When identified as being at risk for a disease such as T2D and CVD, people will try to make sense of their health status by integrating the information concerning their health into a mental model that already exists in their minds. In such a model, new information is integrated with prior beliefs and knowledge about health, illnesses, preventive behaviour and risks.

**Key dimensions of the mental model**

Several attempts have been made to outline the important features of lay representations of being at risk. Although many common chronic multifactorial diseases share several behavioural and physiological risk factors as well as preventive recommendations, representations of being at risk are likely to be disease specific. Leventhal’s Common Sense Model of the self-regulation of health and illness (CSM) suggests that lay representations of a specific illness threat generally consist of a cognitive part, with five interlinked key dimensions (beliefs and knowledge about identity, time-line, causes, consequences and controllability), and an interacting affective part (Leventhal et al., 1997). Identity beliefs include the abstract labels associated with a condition, such as ‘malignant’, as well as concrete symptoms, such as pain or fever. Causal beliefs concern factors that place one at risk. Time-line concerns beliefs about when an illness will strike as well as the speed and nature of progression. Beliefs about consequences include the anticipation and evaluation of possible outcomes of an illness, like a shortened life expectancy, disabilities and social consequences. Controllability beliefs involve beliefs about whether the illness can be prevented, cured or controlled. These include beliefs
about the own capability to change the health status and the efficacy of preventive options (e.g. diet changes). The CSM suggests a relationship between control and causal beliefs. For example, diet changes will not be seen as effective in reducing risk if people do not consider an unhealthy diet an important cause of disease. The affective part of the mental model mostly consists of emotional responses to being at risk such as fear and worry. Emotional responses are likely associated with beliefs about the nature and seriousness of the consequences.

Although research in which the CSM-framework is used has mainly focused on individuals affected by an illness (Hagger & Orbell, 2003), it has also been applied to understand responses of unaffected individuals at risk of developing an illness (e.g. Decruyenaere et al., 2000; van Oostrom et al., 2007; Kaptein et al., 2007; Cameron, 2008). When describing mental models of being at risk such as for diabetes\(^1\) and CVD the same dimensions can be distinguished. However, some dimensions of the CSM may require a modified specification. With regard to the identity dimension, people at risk do not present disease symptoms, but they might possess certain characteristics that they believe to be indicative of increased disease risk, for example high blood pressure can be indicative for CVD-risk. Also, the anticipation and evaluation of possible outcomes of an illness risk may not be (completely) covered by describing the consequences of the associated illness. In addition, representations may also include specific beliefs about the susceptibility to diseases. Three dimensions primarily serve as the basis for the generation of susceptibility beliefs: identity, causal beliefs and time line (Leventhal, 1999; Weinstein 1999; Cameron, 2003). For example, having high blood pressure and believing that high blood pressure can be cause for CVD is likely to raise perceived susceptibility to CVD within the next 10 years.

**How awareness of risk factors may shape the mental model**

Mental models of being at risk may differ depending on people’s awareness of risk factors. Receiving positive DNA-test results, establishing an increased genetic susceptibility to disease, as well as a positive family history specifically refer to a potential genetic origin of the risk. It has been argued that once people consider an underlying genetic foundation for a condition, a particular set of associated beliefs or genetic essentialist biases is triggered (Dar-Nimrod & Heine, 2010). Specifically, these biases are likely to affect causal beliefs. In particular, they may lead people to perceive genes as the sole cause of the condition or at least strengthen genetic attributions of risk thereby potentially weakening attributions to lifestyle factors (Marteau & Weinman, 2006). In addition, as genes are mostly immutable, being aware of a genetic predisposition or an increased genetic susceptibility to a disease may result in genetic fatalism; the belief that the risk cannot be changed

\(^1\) As most lay persons do not distinguish between different forms of diabetes we use the generic term diabetes when referring to people’s illness beliefs.
(Alper & Beckwith, 1993). Alternatively, people may believe that the biological processes invoked by gene variations can be controlled through biologically based actions such as taking medication (Marteau & Weinman, 2006; Smerenck et al., 2009). Genetic risk factors may also affect beliefs about consequences and emotional responses. People may perceive a genetic predisposition or an increased genetic susceptibility to a disease as more severe and more threatening than increased susceptibility due to behavioural factors (Senior, Marteau & Peters, 1999; Shiloh, Rashuk-Rosenthal, Benyamini, 2002). Awareness of disease specific risk factors can also be part of identity beliefs. It can also be expected that receiving positive DNA-test results, experiencing an illness in the family, and/or awareness of other risk factors will raise perceived susceptibility to disease when people believe these factors to be causes of disease.

The mental model in relation to the self-concept

How people perceived their risk may also depend on how people perceive themselves in relation to these risks. The processing of self-relevant information, including risk information, may be affected by people’s ideas about who they are as a person, i.e. the self-concept. The self-concept can be described as a highly interconnected knowledge and beliefs structure in which different self-beliefs, concerning physical and psychological attributes, are loosely organized (Markus & Sentis, 1982; Linville, 1987; Campbell et al., 1996; Baumeister, 2010).

Each person has a multiplicity of self-beliefs. However, only a few of them are active in focal awareness. Different situations (e.g. being at work versus being at home) can activate different self-beliefs. Furthermore, beliefs that are seen as self-defining are rich and well articulated while beliefs that are less important to one self-definition will be less elaborated. For example, a person who considers “being healthy” a defining aspect of the self will have a richer set of beliefs (e.g. about typical examples, food preferences and expected behaviour) than someone who does not consider “being healthy” an important aspect of the self.

Besides differences in content, as expressed in self-beliefs, the organization of the self-concept may also reflect fundamentally different perspectives on human nature (Chiu, Hong, & Dweck, 1997; Levy, Plaks, & Dweck, 1999; Dweck, 2000; Dweck & Molden, 2005). People with a deterministic, static perspective on human nature see their physical and psychological attributes as fixed over time and situations. Others have a more dynamic perspective and analyze and understand themselves in terms of goals, needs and states of mind and put more emphasis on situational influences. These differences in perspective can result in different patterns of self-relevant information processing. Those with a static perspective selectively attend to self-relevant information that facilitates trait-based judgments about the self and is consistent with a deterministic perspective, while those with a dynamic perspective are more attentive to information
that is consistent with a dynamic perspective (Levy et al., 1999). Processing information about potential health problems may follow the same pattern. Those with a static perspective may be more likely than those with a dynamic perspective to regard a susceptibility to a disease such as diabetes and CVD as a physical attribute that is fixed and unchangeable.

**The mental model in relation to preventive behaviour**

People at risk for T2D and CVD are often encouraged to engage in preventive behaviour (i.e. adopt a healthy lifestyle, and adhere to medication) in order to reduce their risk. The CSM proposes that mental models of being at risk will guide the identification and use of appropriate means to reduce risk (Leventhal et al., 1997). That is, people will only adhere to preventive recommendations if the recommendation corresponds with their representations of risk. For example, it is unlikely that a person diagnosed with FH will adopt a healthy lifestyle when he or she thinks that their susceptibility to CVD-risk is predominantly caused by genetic factors and as a result does not consider the adoption of health lifestyle as an appropriate means to reduce risk.

With some health threats, the nature of the threat and the appropriate course of action may be easily understood by the individual (Marteau & Weinman, 2006). For example, most people are aware that smoking causes damage to the lungs and that smoking cessation will reduce this damage. However, the nature of other health threats may be more difficult to grasp. For example, people who are diagnosed with FH by DNA-testing may not understand what this means in terms of their susceptibility to CVD nor what they could do to reduce risk.

It should also be noted that there is a dynamic relationship between the adoption of preventive behaviour and perceived and objective susceptibility to disease risk. That is, if people adopt preventive behaviour this will reduce their risk. For example, taking cholesterol lowering medications will reduce the risk of CVD by lowering cholesterol levels. This reduction of risk is likely to result in a decreased perceived susceptibility to CVD.

Besides the CSM, several other theories such as the Health Belief Model, Protection Motivation Theory and Theory of Planned behaviour have been proposed to explain why individuals engage (or fail to engage) in preventive behaviours (for a description see for example: Noar, 2005). These theories share concepts that can be linked to the key dimensions of the CSM, including beliefs about potential health threats (e.g. perceived susceptibility) and about control over this threat (e.g. response efficacy) (Weinstein, 1993). A person’s decision to engage in preventive behaviour is suggested to arise from a perceived susceptibility to health threats and the expectation that something can be done to reduce this risk. Other elements of health behavioural theories referring to specific health behaviours such as social norms and self-efficacy, are not incorporated in the CSM. While these construct certainly contribute to explaining health behaviour they do not specifically
refer to or influence representations of being at risk and therefore fall outside the scope of this study project.

The study project
This study project was performed to gain a better understanding of people’s mental model of being at risk for diabetes and CVD and how these mental models relate to awareness of genetic and other risk factors and to people’s self-concept.

The conceptual framework
Based on the literature described above, a simplified conceptual framework can be constructed (see Figure 1). This framework describes the key dimensions of the mental model of being at risk (1), how awareness of genetic and other risk factors shape the mental model (2a), the mental model in relation to the self-concept (2b), and the mental model in relation to preventive behaviour (2c). The conceptual framework provides the base for the present study project. Numbers correspond with the research questions.

Figure 1: The conceptual framework; the mental model of being at risk and the relationships with risk factors, self-concept and preventive behaviour

Research questions
In this thesis the following questions are addressed.

1. How do people perceive being at risk for diabetes and CVD?
   a. What are the key dimensions of people’s mental models of being at risk for diabetes and CVD (i.e. identity, time line, causal beliefs, consequences and control, and affective responses and perceived susceptibility)?
   b. What are the relationships between the key dimensions of people’s mental models of being at risk for diabetes and CVD (i.e. identity, time line, causal beliefs, consequences and control, and affective responses and perceived susceptibility)?
2. How is awareness of risk factors and people’s self-concept related to mental models of being at risk for diabetes and CVD and to preventive behaviour?
   a. How are different types of risk (positive DNA test results, positive family disease history and other risk factors) related to differences in mental models of being at risk for diabetes and CVD?
   b. What is the relationship between people’s self-concept and their mental models of being at risk for diabetes and CVD?
   c. What is the relationship between people’s mental models of being at risk for diabetes and CVD and preventive behaviour?

Study design
To be able to answer the research questions, data were collected from individuals who were previously identified as being ‘at risk’ for T2D or CVD based on different types of risk (see Table 1). All participants were at risk for either T2D or CVD (or both), but did not have any symptoms of the disease.

Table 1: Different types of risk for cardiovascular disease and type 2 diabetes

<table>
<thead>
<tr>
<th>Risk type</th>
<th>Cardiovascular disease</th>
<th>Type 2 diabetes</th>
</tr>
</thead>
<tbody>
<tr>
<td>Based on positive DNA tests results</td>
<td>A</td>
<td>–</td>
</tr>
<tr>
<td>Based on positive family disease history</td>
<td>B</td>
<td>D</td>
</tr>
<tr>
<td>Based on other risk factors (e.g. being overweight, high cholesterol levels)</td>
<td>C</td>
<td>E</td>
</tr>
</tbody>
</table>

First, to get a better understanding of the different aspects of people’s mental model of and responses to being at risk, semi-structured interviews were held with a small subset of people at risk for T2D (Pijl et al., 2009a) or CVD (unpublished data). In addition, an analogue study was performed to validate a self-concept measure assessing the extent to which people hold a dynamic versus static view on the self (the results are described in chapter 5 of this thesis). The results of the interviews and analogue study were subsequently used to develop a descriptive questionnaire assessing risk factors, self-concept, lay representations of diabetes and CVD and preventive behaviour. The study protocol was approved by the Medical Ethical Committee of the VU University Medical Center, Amsterdam in 2005.
Descriptive questionnaire study

To represent the different risk types (see Table 1: A - E) data were collected from three separate samples:

1. The first sample (n=81; representing A) consisted of individuals recently diagnosed with FH by DNA testing (aged 20 – 80 years) contacted with the collaboration of the StOEH (Foundation for Tracing Hereditary Hypercholesterolemia - Dutch acronym). In the Netherlands, family members of clinically diagnosed FH patients are traced in a nationwide family cascade screening program by the StOEH. Located family members are invited to participate in the screening program. If a person decides to participate, a genetic field worker visits him or her at home, provides information about the condition and the treatment options and takes a blood sample for cholesterol measurement and DNA analyses. The blood sample is subsequently tested for the mutation in the LDL receptor gene found in the index patient. Results are communicated by letter. Screened positives are advised to consult their general practitioner and/or a vascular specialist.

2. The second sample (n=49; representing B and C) consisted of individuals (aged 30 -75 years) recruited among patients (registered with general practices) with a suspected high risk for CVD, participating in an ongoing intervention study aimed at improving patient adherence to lifestyle advice (IMPALA - Koelewijn et al., 2008). In this intervention, CVD-risk and preventive options were discussed with the patient by a practice nurse.

3. The third sample (n=255; representing B, C, D and E) consisted of individuals (aged 57–79 years) who previously participated in a population based screening program to identify people with undiagnosed T2D. The program was carried out from 1998 to 2000 among inhabitants of a semi-rural region of the Netherlands (Spijkerman et al., 2002a). Participants in this program first filled out a Symptom Risk Questionnaire (SRQ), which included questions about age, family history of T2D, Body Mass Index (Ruige at al., 1997). High scores not only indicate a high risk of developing T2D but also a high risk for CVD (Spijkerman et al., 2002b). Screen positives (SRQ score ≥ 6) were subsequently biochemically tested for T2D. For the present study project, a random selection (n=319) from all screen positives who did not have T2D at the time of testing were approached by mail. These individuals had been informed of the negative test results by letter but did not receive further information about their disease risk or preventive options.
Potential participants were sent information about the purpose of the study and an accompanying informed consent form. After receiving informed consent, participants were sent a postal questionnaire.

Outline of this thesis
Chapter 2 examines the extent to which self-reported risk factors of diabetes and CVD are translated into perceived disease risk (research question 2a). In addition, the moderating role of causal beliefs on perceived disease risk is explored (research question 1b). In the third chapter, representations of CVD and risk reducing behaviour of people diagnosed with FH by DNA testing are described (research questions 1a and 2a). In addition, the relationships between representations (research question 1b), and between representations and risk reducing behaviour are examined (research question 2c). In chapter 4, representations of CVD risk and risk reducing behaviour are compared across two samples of individuals at risk for CVD; with and without a known genetic predisposition for CVD. Differences between the samples are further explored to assess the role of DNA test results compared to family history and non-genetic risk factors (research question 2a and 2c). Chapter 5 describes the results of the analogue study that was performed to validate a self-concept questionnaire intended to assess the perceived ability to change self-attributes (Self-Malleability). Predictive validity is tested by asking participants to imagine themselves in different health scenarios (representing different types of CVD risk) and assessing perceived control over risk and preferences for risk reducing actions (research question 2b). In chapter 6, a hypothesized model representing the relationships between family history of diabetes, Self-Malleability, perceived control over risk and perceived efficacy and adoption of preventive behaviour is tested (research questions 2a, 2b and 2c). The purpose of chapter 7 is to discuss the evidence for the use of family history as a tool for primary prevention of common chronic diseases, in particular for tailored interventions aimed at promoting healthy lifestyles (research questions 2a and 2c). In addition, recommendations for further studies are addressed. In the final chapter the findings of the previous studies are discussed. This chapter also addresses the implications for general and public health practice and for further research.
Chapter 2

Being at risk for diabetes and cardiovascular diseases: causal beliefs and perceptions of disease risk

Abstract
Understanding people’s perceptions of disease risk and how they compare to epidemiological models might improve the effectiveness of risk communication. This study examined perceived disease risk and causal beliefs of type 2 diabetes (diabetes) and cardiovascular diseases (CVD), the relationship between self-reported risk factors and perceived disease risk and the influence of causal beliefs on perceived disease risk in people at increased risk.

The sample (n=255) consisted of individuals at increased risk for diabetes and CVD (aged 57-79 years). Participants completed a postal questionnaire assessing risk factors, perceived risk and causal beliefs (ideas about diseases causation and risk factors) of diabetes and CVD. We employed regression analyses to examine the relationship between risk factors and perceived disease risk and to explore to what extent causal beliefs strengthened the relationship between risk factors and perceived disease risk.

Associations between risk factors and perceived diabetes and CVD risk were weak. Perceived risk, causal beliefs and explained variance of risk factors on perceived risk were lower for diabetes than for CVD. Stronger beliefs concerning overweight as a cause of diabetes and smoking as a cause of CVD strengthened the association between risk factor and perceived disease risk.

Although people in general seem to have some understanding of disease causation, they only partially translate self-reported risk factor information into perceptions of risk. To improve understanding of risk information, it may be relevant to specifically address how personal risk factors can contribute to the development of diabetes and CVD.
Introduction

Prevention programs for common chronic diseases, such as Type 2 diabetes and cardiovascular disease (CVD) often incorporate disease risk information to improve people’s understanding of their disease risk. An improved understanding is assumed to help people to make better decisions regarding their health, including treatment choices and intentions to adopt a healthier lifestyle. Studies, however, show that informing people about disease risk is often of limited success (Edwards et al., 2000). One possible explanation is that people’s understanding of disease risk may conflict with the epidemiological perspective and therefore the information is subject to misinterpretation. To improve the effectiveness of risk communication, it is important to have a better understanding of how people perceive their risk of developing a disease.

Several factors contribute to the development of diabetes and CVD. Many patients who develop diabetes or CVD share a background of physiological characteristics associated with a disordered metabolism such as insulin resistance, cholesterol abnormalities, hypertension, and central obesity (D’Agostino et al., 2004; Smith, 2007). These metabolic abnormalities increase with age and are mostly caused by a combination of genetic factors and behavioural factors (i.e. unhealthy eating habits, physical inactivity and smoking). In addition, having diabetes increases the risk of CVD by 2 to 4 times (Smith, 2007). Also, a family disease history, reflecting the consequences of interactions among multiple genetic and behavioural factors, is a strong and independent risk factor for diabetes (Harrison et al., 2003; Valdez et al., 2007) and CVD (Kardia et al., 2003; Nasir et al. 2007; Murabito et al, 2005).

Some researchers argued that perceptions of disease risk are based on matching personal risk factor information with beliefs about risk factors and causes of disease (Leventhal, Kelly & Leventhal, 1999; Weinstein, 1999; Cameron, 2003; Gerend et al., 2004). The extent to which people believe a certain factor could be cause of disease (or will increase disease risk) is likely to strengthen the relationship between personal risk factor information and perceived disease risk. If a factor does not fit with beliefs about disease causation, it is not likely to raise perceived risk. For example, smoking may only be associated with higher disease risk if it is considered an important cause of disease.

In the present study, we examined how people perceive their risk of Type 2 diabetes (diabetes) and cardiovascular disease (CVD) in relation to epidemiological models of disease risk. The few studies on causal beliefs for diabetes and CVD show that most people believe that both lifestyle factors and genetic factors could be a cause of these diseases (Sanderson et al, 2009: Walter et al., Pijl et al., 2009a). However, these studies did not examine the role of causal beliefs in perceived disease risk nor made comparisons across diseases. The objective of this study was to examine to
what extent causal beliefs of diabetes and CVD strengthened the relationship between self-reported risk factors and perceived disease risk.

**Methods**

**Design, sample and procedure**

We used data from a cross-sectional study performed in 2007. The sample was recruited from a database of a population-based screening program to identify people with undiagnosed diabetes among inhabitants of a semi-rural region of the Netherlands, carried out from 1998 to 2000 (Spijkerman et al., 2002a). Participants in this program first filled out a Symptom Risk Questionnaire, which included questions about age, family history of diabetes and BMI. Screen positives (scores ≥ 6) were subsequently biochemically tested for diabetes. High scores not only indicate a high risk of developing diabetes but also a high risk for CVD (Ruige et al., 1997). For the present study, we approached a random selection (n=319) from all screen positives who did not have diabetes at the time of testing by mail. The letters included study information and informed consent forms. Participants who stated not to have diabetes, and signed and returned the informed consent form were sent a postal questionnaire (participation rate was 80%). The VU University Medical Center Ethical Committee in Amsterdam approved the study protocol.

**Measures**

**Sample characteristics**

Participants provided data on age, sex, marital status, and educational level.

**Coding of self-reported risk factors**

We used four measures to assess physiological risk factors associated with a disordered metabolism. Participants were asked whether they were told that the cholesterol level in their blood was too high and whether their blood pressure was too high. For both items, we coded “yes” responses as 1 and “no” responses as 0. With self-reported weight (in kg) and height (in m) we calculated Body Mass Index (BMI = kg/ m²); a normal weight (BMI below 25) was coded as 0, overweight (a BMI between 25 and 29.9) as 1, and obese (a BMI of 30 and above), as 2.

In addition, we collected data on three behavioural risk factors: unhealthy diet and eating habits, lack of physical activity, and smoking behaviour. The first two behaviours were assessed by the extent of agreement with the statements “I attend to my diet and see to it that I eat healthy everyday” and “I see to it that I am sufficiently physical active everyday” (explained in the instruction to participants as
eating food with little saturated fat and at least two pieces of fruit and 200 grams of vegetables every
day and at least half an hour of moderate intense physical activity, such as walking, biking, swimming
and gardening, five days a week) (response categories: completely disagree (1) – completely agree
(5). Response categories 1, 2 and 3 were recoded as 1 and categories 4 and 5 were coded as 0.

Participants also indicated which category best described their smoking behaviour (response
categories: “I am a smoker”, “I stopped smoking less than 2 years ago”, “I stopped smoking more
than 2 years ago”, and “I have never smoked (with any regularity)”. Non-, and ex-smokers were
assigned a score of 0 and current smokers a score of 1.

Participants also provided information on how many (0, 1, 2 and more) and which of their 1st degree
family members (parents and siblings) were affected by diabetes and CVD respectively.

**Perceived disease risk**
With two items for diabetes and two items for CVD, we assessed perceived disease risk: “How likely
do you think it is that you will get diabetes (CVD) within the next 10 years” (very unlikely (1) - very
likely (7)), “Based on your feelings what is the chance that you will develop diabetes (CVD) within the
next 10 years” (very low (1) - very high (7)). Reliability (Spearman-Brown coefficient) of this measure
was $r_\text{sh} = .93$ for diabetes and $r_\text{sh} = .93$ for CVD.

**Causal beliefs**
In a list of possible causes of diabetes (CVD), taken from the revised Illness Perception Questionnaire
(Moss-Morris et al., 2002) participants indicated for each cause the extent to which they believed it
could be a cause of diabetes (CVD). For the present study, we selected the causes associated with
established risk factors: “advancing age”, “unhealthy diet or eating habits”, “lack of physical activity”,
“smoking” and “hereditary, diabetes (CVD) runs in the family”. In addition, participants indicated to
which extent they believed “being overweight” could be a cause of diabetes and to which extent they
believed “raised cholesterol levels”, “raised blood pressure” and “having diabetes” would increase
the risk of developing CVD. Responses for all items were assessed on five-point rating scales (range:
definitely not (1)–definitely (5)).

**Statistical analyses**
To describe the characteristics of the sample and perceived disease risk and causal beliefs of diabetes
and CVD, we generated means, standard deviations and frequencies. Cases with missing values were
pairwise deleted from the subsequent analyses. Simple and multiple regression analyses were
employed to examine the predictive value of the nine risk factors on perceived risk of diabetes and CVD. To explore the moderating role of causal beliefs on perceptions of disease risk, we performed a series of separate hierarchical multiple regression analyses for each risk factor. In the first step, we entered the risk factor and the associated causal belief in the regression, in the second step, we added an interaction term (product of risk factor and causal belief) to the regression. A moderator effect is present if the interaction term explains a significant amount of variance in perceived risk. To compare across diseases, we performed paired samples t-tests to assess the difference between perceived risk of diabetes and perceived risk of CVD and differences in causal beliefs.

Results

The study sample

Sample characteristics are presented in Table 1. Most participants were either married or lived together, education level was generally low, and a high percentage of participants was overweight.

Table 1: Self-reported characteristics of participants (n=255)

<table>
<thead>
<tr>
<th>Demographic variables</th>
<th>N (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age, mean (sd), range</td>
<td>66 (5), 57-79</td>
</tr>
<tr>
<td>≥ 65 years</td>
<td>156 (61)</td>
</tr>
<tr>
<td>Male</td>
<td>132 (52)</td>
</tr>
<tr>
<td>Married/ Living together</td>
<td>200 (78)</td>
</tr>
<tr>
<td>Education^</td>
<td></td>
</tr>
<tr>
<td>Low</td>
<td>140 (55)</td>
</tr>
<tr>
<td>Medium</td>
<td>71 (28)</td>
</tr>
<tr>
<td>High</td>
<td>35 (14)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Risk factors</th>
<th>N (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physiological risk factors</td>
<td></td>
</tr>
<tr>
<td>Raised cholesterol levels</td>
<td>78 (31)</td>
</tr>
<tr>
<td>Raised blood pressure</td>
<td>125 (49)</td>
</tr>
<tr>
<td>Overweight (BMI^ ≥ 25)</td>
<td>201 (79)</td>
</tr>
<tr>
<td>Obese (BMI ≥ 30) (% of overweight)</td>
<td>88 (44)</td>
</tr>
<tr>
<td>Behavioural risk factors</td>
<td></td>
</tr>
<tr>
<td>Unhealthy diet or eating habits^</td>
<td>84 (33)</td>
</tr>
<tr>
<td>Lack of physical activity^</td>
<td>97 (38)</td>
</tr>
<tr>
<td>Current smoker (%)</td>
<td>35 (14)</td>
</tr>
<tr>
<td>Family disease history^</td>
<td></td>
</tr>
<tr>
<td>Diabetes</td>
<td>112 (44)</td>
</tr>
<tr>
<td>CVD</td>
<td>149 (58)</td>
</tr>
<tr>
<td>Both diabetes and CVD</td>
<td>81 (32)</td>
</tr>
</tbody>
</table>

^ Low: primary school, lower level of secondary school or lower vocational training. Medium: higher level of secondary school, or intermediate vocational training. High: higher vocational training or university.

^ Body Mass Index = kg/m^2

^ Lack of agreement with the statement “I attend to my diet and see to it that I eat healthy every day”

^ Lack of agreement with the statement “I see to it that I am sufficiently physical active everyday”

^ At least one 1st degree relative with diabetes and/or cardiovascular disease (CVD).
Descriptives of perceived risk of diabetes and CVD and predicted value of each of the risk factors on perceived risk of diabetes and CVD are presented in Table 2. Mean scores of both measures of perceived risk measures were relatively low (below scale midpoint). Mean scores for perceived risk of diabetes were significantly lower than means scores for perceived risk of CVD.

Table 2: Simple and multiple regression analyses for perceived risk of diabetes and cardiovascular disease (CVD) on risk factors

<table>
<thead>
<tr>
<th>Risk factors</th>
<th>Perceived risk of diabetes</th>
<th>Perceived risk of CVD</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean (sd), range 1-7</td>
<td>t(235) = 7.97; p &lt; .001</td>
</tr>
<tr>
<td></td>
<td>Difference t(df)</td>
<td>t(df)</td>
</tr>
<tr>
<td>Advanced age³</td>
<td>-1.7</td>
<td>.03</td>
</tr>
<tr>
<td>Raised cholesterol levels</td>
<td>.08</td>
<td>.12</td>
</tr>
<tr>
<td>Raised blood pressure</td>
<td>.06</td>
<td>.13</td>
</tr>
<tr>
<td>BMI²</td>
<td>.05</td>
<td>.00</td>
</tr>
<tr>
<td>Unhealthy diet/eating habits</td>
<td>.11</td>
<td>.01</td>
</tr>
<tr>
<td>Lack of physical activity</td>
<td>.16</td>
<td>.03</td>
</tr>
<tr>
<td>Current smoker</td>
<td>-.05</td>
<td>.16</td>
</tr>
<tr>
<td>Family history diabetes³</td>
<td>.22</td>
<td>.00</td>
</tr>
<tr>
<td>Family history CVD³</td>
<td>.02</td>
<td>.18</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Risk factors</th>
<th>Perceived risk of diabetes</th>
<th>Perceived risk of CVD</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean (sd), range 1-7</td>
<td>t(222) = 3.03; p = .000</td>
</tr>
<tr>
<td></td>
<td>Difference t(df)</td>
<td>t(df)</td>
</tr>
<tr>
<td>Advanced age³</td>
<td>-.13</td>
<td>.05</td>
</tr>
<tr>
<td>Raised cholesterol levels</td>
<td>.06</td>
<td>.36</td>
</tr>
<tr>
<td>Raised blood pressure</td>
<td>.05</td>
<td>.44</td>
</tr>
<tr>
<td>BMI²</td>
<td>.01</td>
<td>.91</td>
</tr>
<tr>
<td>Unhealthy diet/eating habits</td>
<td>.07</td>
<td>.28</td>
</tr>
<tr>
<td>Lack of physical activity²</td>
<td>.11</td>
<td>.08</td>
</tr>
<tr>
<td>Current smoker</td>
<td>-.04</td>
<td>.08</td>
</tr>
<tr>
<td>Family history diabetes³</td>
<td>.23</td>
<td>.00</td>
</tr>
<tr>
<td>Family history CVD³</td>
<td>-.01</td>
<td>.29</td>
</tr>
</tbody>
</table>

Explained variance of all risk factors:
\( R^2 = .11 \) for diabetes
\( R^2 = .18 \) for CVD

³ Age in years < 64 = 0; ≥ 65 = 1
² Body Mass Index (kg/m²) < 25 = 0, ≥ 25 = 1, ≥ 30 = 2
³ Lack of agreement with the statement “I attend to my diet and see to it that I eat healthy every day”
² Lack of agreement with the statement “I see to it that I am sufficiently physical active everyday”
⁴ Number of affected 1st degree relatives: 0, 1, ≥ 2

A positive family history of diabetes and lack of physical activity were positively associated with perceived diabetes risk, and being 65 years or older was negatively associated with perceived diabetes risk. Raised cholesterol levels, family history of CVD, unhealthy diet and eating habits, and lack of physical activity were positively associated with perceived CVD risk. Being 65 years or older and family history of diabetes were negatively associated with perceived CVD risk. Taken together,
self-reported risk factors accounted for 11% of the variance in perceived risk of diabetes and 18% of the variance in perceived risk of CVD (see Table 2).

**Causal beliefs and the relationship between risk factors and perceived disease risk**

Descriptives of causal beliefs (scale 1-5) of diabetes and CVD are presented in Table 3. Causal beliefs concerning behavioural factors (unhealthy diet, lack of physical activity and smoking) were less strong for diabetes than for CVD. Mean scores for causal beliefs about diabetes ranged from 2.9 (sd = 1.2) for “smoking” to 4.0 (sd = 1.0) for “being overweight”. Means scores for beliefs about CVD were all ≥ 4.0, except for “advancing age” (M= 3.7, sd = 0.7) and “having diabetes” (M= 3.4, sd = 1.1).

**Table 3: Causal beliefs for diabetes and cardiovascular disease**

<table>
<thead>
<tr>
<th>Causes and risk factors</th>
<th>Diabetes</th>
<th>Cardiovascular disease</th>
<th>Difference (t-test)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Means (sd)</td>
<td>Means (sd)</td>
<td>t(df)</td>
</tr>
<tr>
<td></td>
<td>range 1-5</td>
<td>range 1-5</td>
<td></td>
</tr>
<tr>
<td>Advanced age</td>
<td>3.8 (1.0)</td>
<td>3.7 (0.9)</td>
<td>t(232) = –1.07</td>
</tr>
<tr>
<td>Raised cholesterol levels</td>
<td>4.0 (1.0)</td>
<td>4.1 (1.0)</td>
<td>–</td>
</tr>
<tr>
<td>Raised blood pressure</td>
<td>4.1 (1.0)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Being overweight</td>
<td>4.0 (1.0)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Unhealthy diet/eating habits</td>
<td>3.8 (1.2)</td>
<td>4.1 (1.1)</td>
<td>t(237) = 3.94</td>
</tr>
<tr>
<td>Lack of physical activity</td>
<td>3.4 (1.2)</td>
<td>4.1 (1.0)</td>
<td>t(238) = 7.93</td>
</tr>
<tr>
<td>Current smoker</td>
<td>2.9 (1.2)</td>
<td>4.0 (1.2)</td>
<td>t(235) = 13.17</td>
</tr>
<tr>
<td>Heredity; it runs in the family</td>
<td>3.6 (1.3)</td>
<td>4.0 (1.1)</td>
<td>t(236) = 3.77</td>
</tr>
<tr>
<td>Having diabetes</td>
<td>3.4 (1.1)</td>
<td>–</td>
<td>–</td>
</tr>
</tbody>
</table>

*Participants indicated for each cause the extent to which they believed it could be a cause (or risk factor) of diabetes (cardiovascular disease)

For diabetes, we found significant risk factor - causal belief interaction effects for advancing age and being overweight (see Table 4a). For advanced age, explained variance in perceived risk of diabetes increased from 3% for age category alone (see Table 2) to 6% when the interaction term was added, and for being overweight, explained variance in perceived risk of diabetes increased from 0% (see Table 2) to 3%. For CVD, we found significant risk factor causal belief interaction effects for smoking and heredity (see Table 4b). For smoking, explained variance in perceived risk of CVD increased from 0 % (see Table 2) to 3% and for family history explained variance increased from 3% (see table 2) to 8%. Other causal beliefs for diabetes and CVD did not significantly moderate the relationship between risk factor and perceived disease risk.
### Table 4: Hierarchical stepwise multiple regression analyses for perceived disease risk on risk factors: the moderating role of causal beliefs

#### 4a Perceived risk of diabetes

<table>
<thead>
<tr>
<th>Causes and risk factors</th>
<th>Perceived risk of diabetes</th>
<th>1st step</th>
<th>2nd step</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>β</td>
<td>p-value</td>
<td>R²</td>
</tr>
<tr>
<td>Advanced age&lt;sup&gt;a&lt;/sup&gt;</td>
<td>−.17</td>
<td>.01</td>
<td>.04</td>
</tr>
<tr>
<td>Causal belief</td>
<td>.11</td>
<td>.10</td>
<td>.28</td>
</tr>
<tr>
<td>Interaction term</td>
<td>−.55</td>
<td>.04</td>
<td></td>
</tr>
<tr>
<td>BMI&lt;sup&gt;b&lt;/sup&gt;</td>
<td>.04</td>
<td>.54</td>
<td>.00</td>
</tr>
<tr>
<td>Causal belief</td>
<td>.04</td>
<td>.60</td>
<td>.18</td>
</tr>
<tr>
<td>Interaction term</td>
<td>.75</td>
<td>.02</td>
<td></td>
</tr>
<tr>
<td>Unhealthy diet/eating habits&lt;sup&gt;c&lt;/sup&gt;</td>
<td>.11</td>
<td>.10</td>
<td>.01</td>
</tr>
<tr>
<td>Causal belief</td>
<td>.03</td>
<td>.64</td>
<td>.03</td>
</tr>
<tr>
<td>Interaction term</td>
<td>.25</td>
<td>.26</td>
<td></td>
</tr>
<tr>
<td>Lack of physical activity&lt;sup&gt;d&lt;/sup&gt;</td>
<td>.15</td>
<td>.02</td>
<td>.03</td>
</tr>
<tr>
<td>Causal belief</td>
<td>.06</td>
<td>.32</td>
<td>.08</td>
</tr>
<tr>
<td>Interaction term</td>
<td>−.09</td>
<td>.68</td>
<td></td>
</tr>
<tr>
<td>Current smoker</td>
<td>−.04</td>
<td>.52</td>
<td>.00</td>
</tr>
<tr>
<td>Causal belief</td>
<td>.05</td>
<td>.50</td>
<td>.01</td>
</tr>
<tr>
<td>Interaction term</td>
<td>.25</td>
<td>.16</td>
<td></td>
</tr>
<tr>
<td>Family history of diabetes&lt;sup&gt;f&lt;/sup&gt;</td>
<td>.20</td>
<td>.002</td>
<td>.06</td>
</tr>
<tr>
<td>Causal belief</td>
<td>.09</td>
<td>.16</td>
<td>.08</td>
</tr>
<tr>
<td>Interaction term</td>
<td>.06</td>
<td>.82</td>
<td></td>
</tr>
</tbody>
</table>

#### 4b Perceived risk of cardiovascular disease

<table>
<thead>
<tr>
<th>Causes and risk factors</th>
<th>Perceived risk of cardiovascular disease</th>
<th>1st step</th>
<th>2nd step</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>β</td>
<td>p-value</td>
<td>R²</td>
</tr>
<tr>
<td>Advanced age&lt;sup&gt;a&lt;/sup&gt;</td>
<td>−.12</td>
<td>.07</td>
<td>.03</td>
</tr>
<tr>
<td>Causal belief</td>
<td>.10</td>
<td>.13</td>
<td>.03</td>
</tr>
<tr>
<td>Interaction term</td>
<td>.42</td>
<td>.17</td>
<td></td>
</tr>
<tr>
<td>Raised cholesterol levels</td>
<td>.26</td>
<td>&lt;.001</td>
<td>.08</td>
</tr>
<tr>
<td>Causal belief</td>
<td>.05</td>
<td>.41</td>
<td>.00</td>
</tr>
<tr>
<td>Interaction term</td>
<td>.63</td>
<td>.08</td>
<td></td>
</tr>
<tr>
<td>Raised blood pressure</td>
<td>.08</td>
<td>.24</td>
<td>.03</td>
</tr>
<tr>
<td>Causal belief</td>
<td>.13</td>
<td>.06</td>
<td>.09</td>
</tr>
<tr>
<td>Interaction term</td>
<td>.41</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Unhealthy diet/eating habits&lt;sup&gt;c&lt;/sup&gt;</td>
<td>.13</td>
<td>.04</td>
<td>.02</td>
</tr>
<tr>
<td>Causal belief</td>
<td>.01</td>
<td>.84</td>
<td>.04</td>
</tr>
<tr>
<td>Interaction term</td>
<td>.50</td>
<td>.10</td>
<td></td>
</tr>
<tr>
<td>Lack of physical activity&lt;sup&gt;d&lt;/sup&gt;</td>
<td>.16</td>
<td>.01</td>
<td>.03</td>
</tr>
<tr>
<td>Causal belief</td>
<td>−.03</td>
<td>.68</td>
<td>.01</td>
</tr>
<tr>
<td>Interaction term</td>
<td>−.13</td>
<td>.64</td>
<td></td>
</tr>
<tr>
<td>Current smoker</td>
<td>.05</td>
<td>.52</td>
<td>.00</td>
</tr>
<tr>
<td>Causal belief</td>
<td>.02</td>
<td>.50</td>
<td>.03</td>
</tr>
<tr>
<td>Interaction term</td>
<td>.63</td>
<td>.02</td>
<td></td>
</tr>
<tr>
<td>Family history of CVD&lt;sup&gt;f&lt;/sup&gt;</td>
<td>.16</td>
<td>.02</td>
<td>.04</td>
</tr>
<tr>
<td>Causal belief</td>
<td>.06</td>
<td>.38</td>
<td>.12</td>
</tr>
<tr>
<td>Interaction term</td>
<td>1.16</td>
<td>.001</td>
<td></td>
</tr>
</tbody>
</table>

---

<sup>a</sup> The extent to which participants believed this could be a cause (or risk factor) of disease

<sup>b</sup> Age in years < 64 = 0; ≥ 65 = 1

<sup>c</sup> Body Mass Index (kg/m²) < 25 = 0, ≥ 25 = 1, ≥ 30 = 2

<sup>d</sup> Lack of agreement with the statement “I attend to my diet and see to it that I eat healthy every day”

<sup>e</sup> Lack of agreement with the statement “I see to it that I am sufficiently physical active everyday”

<sup>f</sup> Number of affected 1<sup>st</sup> degree relatives: 0, 1, ≥ 2
To summarize, the results showed that participants only partially translated self-reported risk factor information into perceptions of risk. They seemed to have a somewhat better understanding of the association between risk factors and CVD risk than between risk factors and diabetes-risk. While for some risk factors causal beliefs strengthened the association between self-reported risk factor and perceived disease risk, (i.e. between advancing age and being overweight and perceived risk of diabetes, and between smoking and heredity and perceived risk of CVD), for other risk factors no moderator effects of causal beliefs were found.

Discussion

We sought to examine perceptions of diabetes and CVD risk in an elderly population at increased risk of diabetes and CVD. The results support earlier findings that family disease history is associated with perceived risk of diabetes (Harwell et al., 2001; Hariri et al., 2006; Adriaanse et al., 2008; Gallivan et al., 2009) and CVD (Hunt et al., 2000; Montgomery et al., 2003; Frijling et al., 2004). We found that believing more strongly that heredity could be a cause of disease strengthened the association between family history of CVD and perceived CVD risk but not between family history of diabetes and perceived diabetes risk. It is possible that the association between family history and perceived risk of diabetes is best explained by the salience of diabetes in memory, which increases with the number of and closeness to people they know with diabetes, rather than family history and heritability of diabetes per se (Walter et al., 2004). There was an unexpected negative association between family history of diabetes and perceived CVD risk. Any explanation is highly speculative and we need further studies to confirm these findings and to disentangle the exact relationship between family history of diabetes and perceptions of CVD risk.

In line with other studies (Harwell et al., 2001; Hariri et al., 2006; Adriaanse et al., 2008; Gallivant et al., 2009; Frijling et al., 2004), we found that people above the age of 65 had lower perceptions of risk than younger people. Considering that this age group has a higher objective risk of diabetes (Ruige et al., 1997) and CVD (Conroy et al., 2003), their lower risk perceptions may be viewed as misconceptions. More worrisome, the negative relationship between age and perceived diabetes risk was even more pronounced in those that viewed advancing age an important cause of diabetes. One explanation is that after a certain age people may think that if they were likely to develop a disease they would already have developed the disease. It could also be that people minimize their risk as a defensive mechanism.

In general, associations between risk factors and perceived disease were weak. Confirming the findings of previous studies (Hariri et al., 2006; Adriaanse et al., 2008), we found that being overweight and smoking were not associated with higher perceived disease risk of diabetes. We also
found that perceived risk of CVD was not associated with being a smoker, a finding that is inconsistent with previous findings (Frijling et al., 2004; van der Weijden et al., 2007). However, most participants indicated that the believed being overweight and smoking could cause diabetes and CVD, respectively. One explanation for this apparent paradox might be that in general, people show an optimistic bias when considering their own health risk (Weinstein, 1987). People believe that they are healthier than others not because they have inaccurate knowledge about the causes of disease but because they do not correctly apply this knowledge to themselves. In particular, they may believe their actions contributing to risk (e.g. smoking) are outweighed by their actions to prevent risk as (e.g. engaging in sufficient physical activity). Notably, being overweight and believing that overweight could be a cause of diabetes was associated with higher perceptions of diabetes risk. Similarly, being a smoker and believing that smoking could be a cause of CVD was associated with higher perceptions of CVD risk.

For other risk factors, causal beliefs did not strengthen the associations between risk factor and perceived disease risk. One explanation might be that causal beliefs were assessed with closed questions which tend to overestimate actual knowledge of disease causation (Pijl et al., 2009a). The absence of a moderator effect for causal beliefs concerning unhealthy diet and eating habits and lack of physical activity might also be explained by the phrasing of the items assessing these factors. Participants were not asked to report on their behaviour as such, but to provide qualitative judgments; “I attend to my diet and see to it that I eat healthy everyday” and “I see to it that I am sufficiently physical active everyday”. Disagreement with these items already indicates some awareness of a causal link between self-reported behaviour and health risk.

Previous studies comparing perceived disease risk of diabetes and CVD showed inconsistent results. While DiLorenzo et al. (2006) found that people perceive their risk of developing CVD as larger than the risk of developing diabetes, Wang et al. (2008) found no difference in perceived risk between diabetes and CVD. In our study, we found that participants perceived their risk for CVD as higher than for diabetes. Furthermore, self-reported risk factors explained more variance in perceived CVD risk than in perceived diabetes risk. Participants also demonstrated a better knowledge about the causes of CVD risk than about the causes of diabetes risk. It is likely that the general public is better informed about risk factors and causes for CVD, because it is the number one cause of death in the Netherlands and more prevalent than diabetes. Strikingly, having diabetes was not generally understood as a risk factor for CVD. Other studies also showed that even among diabetics, the link between having diabetes and higher risk of CVD is not clearly understood (Frijling et al., 2004; van der Weijden et al., 2007).

This study has some limitations that need to be addressed. First, the study presents cross-sectional data and therefore does not show the dynamic relationship between risk factors, perceived
disease risk and causal beliefs. If a personal risk factor is seen as increasing disease risk and or as an important cause of disease, a person may be motivated to engage in preventive behaviour, which in turn may reduce the number of risk factors. In addition, we note that people may not accurately report on risk factors. Also limiting the generalization of the results is the unknown representativeness of the study population. The participants participated in a population-based diabetes screening program some years earlier; therefore, their understanding of diabetes risk might have been better than that of the average at risk population. On the other hand, being tested for diabetes and not having diabetes could have minimized the belief that the risk factors had any personal relevance, thereby reducing the moderating effect of causal beliefs.

Our results confirm that people’s understanding of their risk of developing diabetes and/or CVD is limited. In general, people seem to have some knowledge of disease causation, in particular of CVD, but only partially use this knowledge to link personal risk factors to an increased disease risk. To address misconceptions and improve understanding of risk information, health professionals may need to educate people on the nature of their disease risk; explain the causes of disease and how personal risk factors can contribute to the development of diabetes and CVD.
Chapter 3

Perceived risk and representations of cardiovascular disease and preventive behaviour in people diagnosed with Familial Hypercholesterolemia

Abstract
Perceived risk and representations of cardiovascular disease (CVD), and preventive behaviour of people diagnosed with Familial Hypercholesterolemia by DNA testing (n=81) were assessed. In general, participants perceived their own CVD risk as being relatively low. While participants reported almost optimal medication adherence (99%), only 49% reported following recommendations concerning diet and physical activity. Family history of CVD was associated with both risk perception and the adoption of a healthy lifestyle. In their communications with FH-screened positives, health professionals should be aware that people may underestimate CVD risk, and should stress how behaviour change can reduce the risk.
Introduction

With the rapid developments in the field of health screening technology, people are increasingly being confronted with new information about their susceptibility to a range of diseases. In particular, advances in genetic testing have opened the door for new possibilities and responsibilities in healthcare and prevention. Whether or not people benefit from genetic testing may depend on how they interpret DNA-based risk information and translate it into behavioural responses. In the present study, we examine how people who are diagnosed with Familial Hypercholesterolemia (FH) by DNA testing interpret, and respond to their risk of developing cardiovascular disease (CVD). FH is a monogenic disease characterized by a family history of CVD, impaired metabolism of low-density lipoprotein cholesterol, and increased risk of developing premature CVD. In Western countries, the estimated frequency of FH is 1 in 400 - 500 people (Austin et al., 2004). People diagnosed with FH can reduce their risk of developing CVD through the use of cholesterol-lowering medication and the adoption of a healthy lifestyle (not smoking, healthy diet and sufficient physical activity) (Civeira, 2004). However, FH patients may not understand information based on DNA results and may feel that there is little that can be done to reduce a genetic risk. As a result, they may not engage in the recommended actions.

A core construct in most health behavioural theories explaining responses to health risk information (e.g. the health belief model and protection motivation theory) is risk perception (the perceived susceptibility to a health threat). It is assumed that when people form accurate risk perceptions, they will be motivated to engage in behaviour that is appropriate to their actual risk. However, in general, studies have found only weak associations between risk perception and preventive behaviour (Brewer et al., 2007). One possible explanation is that people do not form accurate risk perceptions (Leventhal, Kelly, & Leventhal, 1999; Windschitl, 2002). Often, individuals do not receive accurate personalized risk information, possibly because the information for accurate risk assessment may not be available. In addition, risk perception may not reflect well-formed stable beliefs that are stored in memory. Instead, it has been suggested that risk perceptions can be regarded as ‘ad hoc’ constructions, made ‘on the spot’, based on accessible personal beliefs about the threat, rather than retrieved from memory (Leventhal et al., 1999; Windschitl, 2002; Cameron, 2003; Cameron 2008).

The self-regulation model of health and illness (SRM) offers a framework for identifying the content of these personal beliefs (Leventhal et al., 1997) and may help to examine the mismatch between risk perceptions and preventive behaviours. Although research in which the SRM is used has mainly focused on individuals affected by an illness (Hagger & Orbell, 2003), it has also been applied to understand responses of unaffected individuals at risk of developing an illness (Decruyenaere et al., 2000; Rees et al., 2004; van Oostrom et al., 2007; Kaptein et al., 2007; Cameron, 2008). According
to the SRM, people construct mental models (or representations) of health threats based on both internal (e.g. physical signs) and external stimuli (e.g. experiencing an illness of a close relative, risk information). New information concerning a specific threat, such as a positive DNA test result, will be integrated into the existing cognitive and emotional representations of that threat. Cognitive illness-risk representations are based on personal theories about different aspects of the threat ordered into five logical themes or dimensions: identity, causal beliefs, timeline, consequences, and control. The identity dimension includes abstract labels associated with the threat as well as concrete signs and symptoms. Given that diagnosis of FH is based on DNA testing, family history of CVD and cholesterol levels, for people diagnosed with FH these factors are likely to be included in their CVD risk representations (Leventhal, 1999; Shiloh, 2006). Causal beliefs concern factors that place one at risk (e.g. a genetic susceptibility). The timeline dimension refers to beliefs about how the threat will develop over time. The consequences dimension concerns beliefs regarding possible outcomes of the threat (reduced life expectancy, physical and psychosocial effects) and may serve as a base for perceived seriousness. The control dimension contains an evaluation of the perceived options to reduce the threat, which are suggested to be linked to causal beliefs (Marteau & Weinman, 2006). In particular, if an illness is attributed to lifestyle factors, a healthy lifestyle is likely to be considered an effective way of reducing the threat. In addition, if an illness is attributed to genetic factors, a healthy lifestyle may not be considered an effective way of reducing the threat. Emotional illness-risk representations interact with the cognitive representations and most commonly reflect fear-related responses, such as worry.

Both risk perception and preventive behaviour can be considered as products of the underlying illness-risk representations (Leventhal et al., 1999; Cameron, 2003; Cameron 2008). The cognitive representations contributing to risk perception (identity, causal beliefs, and timeline) may differ, however, from the representations producing preventive behaviour (identity, timeline, and beliefs about control) (Leventhal et al., 1999; Cameron, 2003; Cameron 2008). Emotional responses may shape risk perception and trigger emotion regulation processes, which may lead to preventive behaviour but can also generate maladaptive responses, such as denial and avoidance (Cameron, 2003).

Providing people with genetic risk information based on DNA testing may affect perceived risk and representations, and preventive behaviour. It has the potential to raise risk awareness and motivation to engage in risk-reducing actions. However, if a person believes that genetic risks are immutable, it may strengthen beliefs that little can be done to prevent or control the disease, thereby adversely affecting motivation (Alper & Beckwith, 1993; Marteau & Lerman, 2001; Collins et al., 2003). Insight into these processes and how they may affect preventive behaviour may be
beneficial to health professionals in their communications with, and treatment of, screened positives.

From the studies reporting on perceived risk and representations of CVD and behavioural responses of those diagnosed with FH, a picture is beginning to emerge. Regarding the understanding of the genetic risk information, people diagnosed with FH were found to underestimate their risk (van Maarle et al., 2003a), refer to their CVD risk as “relatively low” (Senior et al., 2002), and have perceptions of CVD risk similar to those of clinically diagnosed FH patients in whom no mutation has been found (Marteau et al. 2004). In a qualitative study by Senior et al. (2002), people diagnosed with FH often referred to their CVD risk as “nothing to worry about” and studies by van Maarle et al. (2003b), and Marteau et al. (2004) showed that screened positives did not become more anxious after receiving the test results. For preventive behaviour, the results were more ambiguous. According to a review of the first five years of screening for FH in the Netherlands, 93% of screened positives were on cholesterol-lowering treatment a year after being tested (Umans-Eckenhausen et al., 2001). Senior, Marteau and Weinman (2004) reported high levels of medication adherence in a clinically diagnosed population in the UK. Van Maarle et al. (2002) showed that overall clinical outcome quality, based on cholesterol levels, BMI and smoking status, of screened positives, had improved considerably at follow-up (18 months after receiving the test results). However, this study also showed that the achieved level of care still fell below the current guidelines in almost half of the cases; 15% did not use cholesterol-lowering medication while hypercholesterolemic, and 24% continued smoking. Marteau et al. (2004) showed that finding a mutation in a population already clinically diagnosed had no effect on adherence to risk-reducing behaviours (smoking, diet, activity level, and cholesterol-lowering medication adherence). It did, however, seem to weaken perceptions of the effectiveness of behavioural means and strengthen perceived effectiveness of medication as a way of reducing risk. In a later study, Senior and Marteau (2007) found that attributions to genetic factors were associated with perceived effectiveness of medication, and attributions to behavioural factors were associated with perceived effectiveness of dietary changes.

Although some of the previous studies indicated that perceived risk and representations of CVD and preventive behaviour of screened positives may be linked (Senior et al., 2002; van Maarle et al., 2003a/b; Marteau et al., 2004; Senior & Marteau, 2007), little is known about how they are connected. The present paper reports the findings from a cross-sectional questionnaire study exploring the relationships between perceived risk and representations of CVD and preventive behaviour of people diagnosed with FH by DNA testing. The objectives of the study were to examine a) perceived risk of CVD and their relationship with representations of CVD; b) the adoption of recommended behaviour concerning medication and lifestyle and their relationship with
representations of CVD, in particular with beliefs concerning identity, consequences, timeline and control (efficacy of medication and efficacy of a healthy lifestyle); and c) causal beliefs (attributions of CVD risk) and their relationship with control beliefs.

Methods

Participants and procedure

In the Netherlands, family members of clinically diagnosed FH patients are traced in a nationwide family cascade screening program by the StOEH (Foundation for Tracing Hereditary Hypercholesterolemia - Dutch acronym). Located family members are invited to participate in the screening program. If a person decides to participate, a genetic field worker visits him or her at home, takes a blood sample for cholesterol measurement and DNA analyses, and provides information about the condition and the treatment options. The sample is tested for the mutation in the LDL receptor gene found in the index patient. Results are communicated by letter. Screened positives are advised to consult their general practitioner (GP) and/or a vascular specialist; a letter to inform the participants’ GP is enclosed with this mailing. In 2006, the StOEH randomly selected 200 addresses of men and women who met the following inclusion criteria: screened positive for FH in the preceding two years, and 18 years of age or over. In order to have a representative sample, the addresses were also balanced for gender, place of residence, and type of mutation. To each address, the StOEH sent information about the purpose of the study, inviting participation in the study by the signing and returning of the accompanying informed consent form. Non-respondents were sent a reminder after three weeks. Participants were sent a postal questionnaire asking about perceived risk and representations of CVD and preventive behaviour. The Medical Ethical Committee of the VU University Medical Center approved the study protocol.

Measures

General characteristics of participants. Self-reported data were obtained on demographic variables: age, gender, marital status, educational level and Body Mass Index (BMI = kg/m²).

Perceived risk of CVD

A common method for assessing risk perception is to solicit a single numerical estimate. However, people often have difficulty understanding and providing numerical estimates of probabilities (Windschitl, 2002). Therefore, both numerical estimates and verbal expressions of risk were assessed. To assess numerical risk perception, participants were asked to complete the following statement: “I think I have a chance of ... in a 100 of getting CVD within the next 10 years”. In addition, estimations of age-related population risk were assessed (“I think that of every 100 men/women my
age on average ... will get CVD within the next 10 years”) to compare numerical risk perception to perceptions of population risk. Perceived risk in verbal terms was assessed with three items: “How likely do you think it is that you will get CVD within the next 10 years?” (very likely (1) - very unlikely (7)), “Based on your feelings, how big is the chance of you developing CVD within the next 10 years?” (very low (1) - very high (7)), “What do you think the chance is of you developing CVD compared to an average man/woman your age?” (a lot lower (1) - a lot higher (7)).

Representations of CVD

Identity. Participants were asked to indicate how many (and which) of their first-degree family members were affected by CVD. Based on their answers, a family history variable was created (categories: 0, 1, and 2 or more affected family members). In addition, participants were asked: “Have you been told your cholesterol level in your blood is too high?” using two response categories: “yes” or “no (not that I can remember)”.

Causal beliefs. Participants were presented a list of 20 possible causes of CVD based on the revised Illness Perception Questionnaire (IPQ-r) (Moss-Morris et al., 2002) and asked to indicate for each cause to which extent they believed it could be a cause of CVD (range: definitely not (1) - definitely (5)). For the present study, we identified two separate causal beliefs: heredity (two items: “Heredity, CVD runs in the family” and “a predisposition”, and unhealthy lifestyle (three items: unhealthy diet, lack of physical activity and smoking).

Timeline was assessed with two items: “If I should get CVD, I will have that for the rest of my life” and “If I should get CVD, the symptoms will become less severe in time”, range: completely disagree (1)–completely agree (5)).

The consequences subscale of the IPQ-r was used to assess the consequences dimension (six items, e.g. “If I should get CVD, it will shorten my life considerably”, range: completely disagree (1)–completely agree (5)).

The control dimension was assessed with two variables: Efficacy of medication (one item: “When I use my medication as prescribed, it will reduce my health risk”) and Efficacy of a healthy lifestyle which was explained in the instruction to participants as eating food with little saturated fat and at least two pieces of fruit and 200 grams of vegetables every day and at least half an hour of moderate intense physical activity, such as walking, biking, swimming and gardening, five days a week (two items: “Eating healthily every day would reduce my health risk” and “Taking
sufficient exercise at least five times a week would reduce my health risk”), a third item was added for current smokers: “stopping smoking would reduce my risk”. Responses were assessed on five-point rating scales; completely disagree (1) – completely agree (5)).

Emotional representation of CVD risk

CVD risk worry was assessed with two items on a seven-point rating scale range (“When I think of my chances of developing CVD, I feel ...”; not anxious at all (1) - very anxious (7) / not worried at all (1) - very worried (7)).

Preventive behaviour

Smoking status was assessed by asking participants to describe their smoking behaviour (response categories: “I am a smoker”, “I stopped smoking less than 2 years ago”, “I stopped smoking more than 2 years ago”, “I have never smoked (with any regularity)”).

Adoption of a healthy diet and being sufficiently active (explained in the instruction to participants, see efficacy of a healthy lifestyle) was assessed with two items measured on a five-point rating scale (“I attend to my diet and see to it that I eat healthily everyday” and “I see to it that I am sufficiently physically active every day” - completely disagree (1) - completely agree (5)).

The use of cholesterol-lowering drugs was assessed with one item: “Do you use cholesterol-lowering drugs?” (response categories: “Yes, as prescribed”, “Yes, but I don’t (always) use them as prescribed”, “No, they are prescribed but I don’t use them”, “No, they are not prescribed”).

Statistical analyses

Exploratory analyses were performed to test the assumptions of normality. To correct for non-normality of the distribution, the first three response categories for the items assessing causal beliefs and the two control measures were collapsed (response categories 1, 2 and 3 were recoded as 1, response category 4 was recoded as 2, and 5 recoded as 3). No outliers were detected. A composite variable was created by combining participants’ responses to the two items assessing adoption of a healthy diet and being sufficiently active (response categories 1, 2 and 3 were recoded as 0, response category 4 was recoded as 1, and 5 recoded as 2) and the item assessing smoking behaviour (non-smokers were assigned a score of 2 and smokers a score of 0). Frequencies, means, standard deviations and reliability measures were generated to describe the data. T-tests were performed to test for significant differences between numerical risk perception and estimations of age-related
population risk, hereditary and unhealthy lifestyle causal beliefs and perceived efficacy of medication and healthy lifestyle. Pearson’s correlations (and Spearman’s correlation coefficients for Family history of CVD and point-biserial correlations for Raised cholesterol) were calculated to examine the relationships between risk perceptions, the different dimensions of CVD risk representations, and preventive behaviour. Regression analyses were employed to further explore the predictive value of beliefs concerning identity, consequences of CVD, timeline and control (efficacy of medication) and emotional representations on adoption of a healthy lifestyle and adherence to medication.

Results

Recruitment, response and population characteristics
Of the 200 letters that were sent out, 13 were undeliverable. Of the remaining study population, 140 responded (75% of 187), of whom 86 agreed to participate in the study and 81 returned a completed questionnaire (43% of 187). The mean age of participants was 48 years (sd = 16), 48% were men, 80% married or co-habiting, 36% had completed either higher vocational training or university, and 57% were either overweight or obese (BMI ≥25). There were no significant differences in age or gender between the participants and the study population.

Perceived risk and representations of CVD, and preventive behaviour
Descriptives of perceived risk and representations of CVD, and preventive behaviour are presented in Table 1. Correlations between these measures are presented in Table 2.

Perceived risk of CVD and the relationship with representations of CVD
On average, perceived risk of CVD of participants was relatively low. The average numerical risk perception was 26.88 in 100 (sd = 24.33), although the distribution showed a spike at 50 (mode response of 19% of the participants). A t-test did not show a significant difference (p > .05) between numerical risk perceptions and participants’ estimations of age-related population risk (M= 27.63, sd = 17.83). Perceived risk in verbal terms (M= 3.59, sd = 1.44) was significantly below the scale midpoint (t (77) = -2.545, p = .013, 95% CI [-.74, -.09]).

Numerical risk perceptions and perceived risk in verbal terms were moderately correlated. Numerical risk perceptions were positively correlated with family history of CVD. Associations with being told as having high cholesterol, causal beliefs, timeline and emotional representations were not significant. Numerical risk perceptions were negatively correlated with perceived consequences of having CVD. Perceived risk in verbal terms was positively correlated with both measures of identity (family history of CVD, raised cholesterol), timeline (seeing CVD as a permanent condition) and CVD risk worry. Associations with causal beliefs and emotional representations were not significant.
### Table 1: Perceived risk and representations of CVD, and self-reported preventive behaviour (n=81)

<table>
<thead>
<tr>
<th></th>
<th>Reliability</th>
<th>Means (sd)</th>
<th>%</th>
<th>N</th>
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<tr>
<td><strong>Perceived risk</strong></td>
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<tr>
<td>Numerical</td>
<td></td>
<td>26.88 (24.33)</td>
<td>77</td>
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<tr>
<td>Estimated age related population risk</td>
<td></td>
<td>27.63 (17.83)</td>
<td>78</td>
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<tr>
<td>In verbal terms (scale 1-7)</td>
<td></td>
<td>88 3.59 (1.44)</td>
<td>78</td>
<td></td>
</tr>
<tr>
<td><strong>Identity</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>Family history of CVD¹</td>
<td></td>
<td>2.02 (2.24)</td>
<td>30</td>
<td>81</td>
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<tr>
<td>None</td>
<td></td>
<td>28 24</td>
<td></td>
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<tr>
<td>1 member</td>
<td></td>
<td>42 23</td>
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<tr>
<td>2 or more members</td>
<td></td>
<td>93 34</td>
<td></td>
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<tr>
<td>Raised cholesterol</td>
<td></td>
<td>75</td>
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<td></td>
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<tr>
<td><strong>Causal beliefs (scale 1-3)</strong></td>
<td></td>
<td>.55 2.52 (0.53)</td>
<td>81</td>
<td></td>
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<tr>
<td>Hereditary</td>
<td></td>
<td>.88 2.39 (0.71)</td>
<td>81</td>
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<tr>
<td>Unhealthy lifestyle</td>
<td></td>
<td></td>
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<tr>
<td><strong>Timeline</strong></td>
<td></td>
<td>.54 3.50 (0.65)</td>
<td>81</td>
<td></td>
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<tr>
<td><strong>Consequences (scale 1-5)</strong></td>
<td></td>
<td>.77 3.18 (0.62)</td>
<td>81</td>
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<td><strong>Control (scale 1-3)</strong></td>
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<tr>
<td>Efficacy of medication²</td>
<td></td>
<td>2.41 (0.61)</td>
<td>66</td>
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<tr>
<td>Efficacy healthy lifestyle</td>
<td></td>
<td>.65 2.18 (0.53)</td>
<td>80</td>
<td></td>
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<tr>
<td><strong>CVD-risk worry (scale 1-7)</strong></td>
<td></td>
<td>.87 3.81 (1.48)</td>
<td>76</td>
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<tr>
<td></td>
<td></td>
<td>3.31 (1.39)</td>
<td>79</td>
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<tr>
<td><strong>Adoption of a healthy lifestyle (range 0- 6)</strong></td>
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<td>Smoking status</td>
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<tr>
<td>Current smoker</td>
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<td>11 9</td>
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<tr>
<td>Ex-smoker</td>
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<td>33 27</td>
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<tr>
<td>Never smoked</td>
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<td>53 43</td>
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<td>Healthy diet/ sufficiently active (range 0 - 4)</td>
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<td>.54 1.56 (1.07)</td>
<td>49³</td>
<td>80</td>
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<td></td>
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<td>75</td>
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<td><strong>Use of cholesterol lowering drugs³</strong></td>
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<tr>
<td>Yes, as prescribed</td>
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<td>88 66</td>
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<tr>
<td>Yes, but I don’t (always) use them as prescribed</td>
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<td>1 1</td>
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<td>No, they are prescribed but I don’t use them</td>
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<td>0 0</td>
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<tr>
<td>No, they are not prescribed</td>
<td></td>
<td>9 7</td>
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</table>

¹ Number of 1st degree relatives affected by CVD: 51% of participants reported that their father was affected with CVD, 20% mother, 31% brother(s), 25% sister(s) and 17% children
² Only assessed in those prescribed cholesterol-lowering medication (n=66)
³ Only assessed in those with raised cholesterol (n=75)
⁴ Agreement with both statements concerning diet and physical activity (response categories 4 and 5)

**The adoption of recommended behaviour and the relationship with representations of CVD**

Of those with raised cholesterol (93% of all participants), 9% (7 of 75) reported that they were not prescribed medication, 88% (66 of 75) reported using cholesterol-lowering drugs as prescribed, only 1% (1 of 75) reported partial adherence, and none of the participants reported complete non-
adherence. Eighty-nine percent of participants reported that they were non-smokers and 49% agreed with both statements concerning the adoption of a healthy diet and physical activity.

Because frequencies for partial and complete non-adherence were too small to detect statistical significant effects, no further analyses were performed for medication adherence. The adoption of a healthy lifestyle was positively associated with a family history of CVD and efficacy of a healthy lifestyle. Associations with raised cholesterol, timeline, consequences and CVD risk worry were not significant.

Table 2: Correlations$^1$ for perceived risk and representations of CVD, and preventive behaviour$^2$ (n=81)

<table>
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<tr>
<th></th>
<th>1b</th>
<th>2a$^1$</th>
<th>2b$^1$</th>
<th>3a</th>
<th>3b</th>
<th>4</th>
<th>5</th>
<th>6a</th>
<th>6b</th>
<th>7</th>
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<tbody>
<tr>
<td><strong>1. Risk perception</strong></td>
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<tr>
<td>b. In verbal terms</td>
<td>.225</td>
<td>.228</td>
<td>- .033</td>
<td>- .143</td>
<td>.335</td>
<td>.042</td>
<td>- .001</td>
<td>.095</td>
<td>.449</td>
<td>.012</td>
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<td><strong>2. Identity</strong></td>
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<tr>
<td>b. Raised cholesterol$^1$</td>
<td>.105</td>
<td>.041</td>
<td>.157</td>
<td>.063</td>
<td>.335</td>
<td>.137</td>
<td>.091</td>
<td>.140</td>
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<td><strong>3. Causal beliefs</strong></td>
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<tr>
<td>a. Hereditary</td>
<td>.248</td>
<td>.264</td>
<td>- .084</td>
<td>.195</td>
<td>.268</td>
<td>- .099</td>
<td>.061</td>
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<td>- .149</td>
<td>.113</td>
<td>.344</td>
<td>.267</td>
<td>- .117</td>
<td>- .033</td>
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<td><strong>4. Timeline</strong></td>
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<td>- .117</td>
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<td><strong>5. Consequences</strong></td>
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<td>.008</td>
<td>.003</td>
<td>.189</td>
<td>- .048</td>
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<td><strong>6. Control</strong></td>
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<td>a. Efficacy medication</td>
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<td><strong>7. CVD-risk worry</strong></td>
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<td></td>
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<td>- .094</td>
</tr>
</tbody>
</table>

Correlations in bold are significant at the .05 level (2-tailed)

$^1$ Pearson’s correlations were calculated except for Family history of CVD (Spearman’s correlation coefficients) and for Raised cholesterol (point-biserial correlation)

$^2$ For medication adherence no correlation coefficients are presented, because frequency for non-adherence is 1, which is too small to provide statistical significance

$^3$ Constant; efficacy of medication was only assessed in participants who reported high cholesterol

A stepwise multiple regression analysis was employed to explore which and how well CVD risk representations predicted the adoption of a healthy lifestyle. Demographic variables (age, gender and education) and the relevant representations of CVD (family history of CVD, raised cholesterol, timeline, consequences and efficacy of a healthy lifestyle) were entered into the regression equation. Efficacy of a healthy lifestyle (st. β = .345; p = .001), family history (st. β = .248, p = .014) and gender
(men < women, st. β = -.228; p= .024) explained 25% of the variance in adoption of a healthy lifestyle (F(3,77) = 8.467; p <.001).

**Causal beliefs and the relationship with control beliefs**
Heredity (M = 2.52, sd = .53) and an unhealthy lifestyle (M = 2.39, sd = .71) were generally perceived as likely causes of CVD (differences were not significant, p>.05). Although on average, participants believed strongly in the efficacy of the recommended actions, participants (those who reported being prescribed cholesterol-lowering medication) perceived the adoption of a healthy lifestyle (M = 2.18, sd = .53) as somewhat less effective than medication (M = 2.41 sd = 0.61) in reducing the risk of CVD (t (65) = -2.920, p = .005, 95% CI [-.36, -.07]). Both considering an unhealthy lifestyle and heredity as a likely cause for CVD were positively correlated with the efficacy of a healthy lifestyle. In addition, considering an unhealthy lifestyle as a likely cause for CVD was positively associated with efficacy of medication. The association between considering heredity as a likely cause for CVD and efficacy of medication however was not significant.

**Discussion**
The aim of this study was to examine how those recently diagnosed with FH following DNA testing interpret their risk of developing CVD. We found that in general, participants perceived their own risk of having CVD as relatively low. Participants equally endorsed both genetic and lifestyle attributions of CVD. While participants reported high adherence to medication, adherence to recommendations concerning lifestyle, with the exception of non-smoking, seemed less optimal. As proposed by the SRM, both perceived risk and preventive behaviour were linked to representations of CVD. Having a positive family history of CVD, being informed of high cholesterol, considering CVD as a permanent condition, and CVD risk worry were all positively associated with verbal expressions of perceived risk. Besides considering a healthy lifestyle as an effective way to reduce health risks, having a family history of CVD, and gender explained some of the variance in self-reported adherence to lifestyle recommendations. Although considering heredity as a likely cause for CVD, was positively associated with a positive evaluation of the effectiveness of a healthy lifestyle it was not significantly associated with a positive evaluation of the effectiveness of medication. Considering an unhealthy lifestyle a likely cause of CVD was associated with positive evaluations of both preventive options.

Previous studies have also found relatively low perceived risk in people diagnosed with FH (Senior et al. 2002; van Maarle et al., 2003a). It may be that people diagnosed with FH construct accurate risk perceptions that are based on a correct understanding of their condition, and the accurate belief that the use of cholesterol-lowering drugs is effective in reducing their CVD risk. However, given that the incidence of CVD in people diagnosed with FH, including those on
cholesterol-lowering medication, is still higher than the incidence of CVD in non-carrier family members (Umans-Eckenhausen et al., 2002) and the general population (Morherschadtl et al, 2004), our findings suggest that a considerable proportion of screened positives underestimate their own risk of developing CVD. A common explanation might be that in general, people show an optimistic bias when considering their own health risk (Weinstein, 1987). Alternatively, in communicating with FH-screened positives, health professionals may be especially motivated to focus on the controllability of the disease, in particular the effectiveness of cholesterol-lowering drugs. Explaining the actual CVD risk of having an FH-mutation and the remaining risk with effective treatment may not routinely be part of the communication. In fact, it is rather difficult to provide accurate personalized information on the actual risk for CVD. In the Netherlands today, almost 500 mutations in the LDL receptor gene have been identified, varying widely in expression and response to medication (JoJoGenetics, 2008).

With regard to preventive behaviour, the results are mixed. In line with most other studies, we found a high adherence to prescribed cholesterol-lowering medication (Umans-Eckenhausen et al., 2001; Senior et al., 2004). Interestingly, participants with more first-degree family members affected with CVD were more likely to follow recommendations concerning lifestyle. There is some evidence from other studies that family history information may have a positive effect on motivation to adopt a healthy lifestyle. For example, Hariri and colleagues (2006) found that a family history of diabetes was positively associated with making lifestyle changes to prevent diabetes. Having witnessed CVD in a close relative may have enhanced the salience and immediacy of the CVD threat and, as a result, increased these behaviours. In addition, people with CVD in the family may also have adopted a healthy lifestyle in the family context. A plausible explanation for the difference in adherence to medication and adherence to lifestyle recommendations is the relative ease of taking medication compared to making lifestyle adjustments (Burke, Dunbar-Jacob & Hill, 1997). Alternatively, because participants considered a healthy lifestyle as being less effective in reducing CVD-risk, they may be less inclined to follow recommendations concerning lifestyle. It has been suggested that providing people with genetic risk information reinforces attributions to heredity, thereby strengthening beliefs in the efficacy of medication, and weakens beliefs in the efficacy of a healthy lifestyle Marteau 2004; Senior et al. 2005; Marteau & Weinman, 2006; Senior & Marteau, 2007). However, we found no evidence for a positive link between attributions to heredity and perceived efficacy of medication. Moreover, our findings suggest a positive link between attributions to heredity and perceived efficacy of a healthy lifestyle.

In conclusion, persons with diagnosed FH are aware of their increased risk on CVD but still seem to underestimate the risk and do not always show adequate adherence to lifestyle
recommendations. This might be due to overconfidence in the efficacy of medication as a means of reducing the risk.

Further research is needed to examine the impact of screening and to disentangle causal relations between perceived risk and representations of CVD, and preventive behaviour. In order to assess the actual effects of screening, it will be necessary to measure perceived risk and representations of CVD and preventive behaviour in a prospective study with follow-up. A prospective study may also offer more insight into the relationship between perceived risk and behaviour. This relationship is likely to be reciprocal, i.e. a person who feels at risk may decide to engage in preventive behaviour, and this behaviour may reduce perceived risk.

Factors limiting the generalizability of the findings are the small sample size, response rate and the unknown representativeness of the study sample. It should also be noted that while self-report measures may be appropriate for assessing perceived risk, and representations of an illness, they are less suitable for assessing preventive behaviour.

Mindful of these limitations, the results of this study may have several potential practical implications. We would suggest that health professionals, such as GPs and vascular specialists, should be aware that FH- screened positives may underestimate their CVD risk and have relatively little confidence in the effectiveness of lifestyle changes to reduce their CVD risks. In their communications with people diagnosed with FH, health professionals should emphasize that maintaining an unhealthy lifestyle increases CVD risk. In addition, they may need to clarify that although medication can be effective in lowering cholesterol levels, it will not completely reduce CVD risk to population levels, and therefore appropriate lifestyle adjustments may still be necessary. Our findings suggest that family history of CVD plays a role in the construction of risk perceptions and in the decision to adopt a healthy lifestyle. New cases, in particular young people, may not have a first-degree relative with CVD (yet) and may therefore be less aware of the high risk of having an FH mutation and will therefore be less likely to adopt and maintain a healthy lifestyle.
Chapter 4

Being at risk for cardiovascular disease: perceptions and preventive behaviour in people with and without a known genetic predisposition

Abstract

This study compares and explains differences in perceptions of cardiovascular disease (CVD) risk and preventive behaviours in people with and without a known genetic predisposition to CVD.

A cross-sectional study using two samples was performed. The first sample (genetic predisposition; n=55) consisted of individuals recently diagnosed with Familial Hypercholesterolemia (FH) through DNA testing. The second sample (no genetic predisposition; n=49) was recruited among patients with CVD-risk profiles based on family history of CVD, cholesterol levels and blood pressure, registered at general practices. Participants filled out a postal questionnaire asking about their perceived risk, causal attributions (i.e. genetic and lifestyle), and about perceived efficacy and adoption of preventive behaviour (i.e. medication adherence and adoption of a healthy diet and being sufficiently active).

Perceived risk, genetic attributions of CVD and perceived efficacy of medication were higher in the ‘genetic predisposition’ sample than in the ‘no genetic predisposition’ sample. The samples did not differ on lifestyle attributions, efficacy of a healthy lifestyle, or preventive behaviour. Individual differences in perceived risk, genetic attributions and perceived efficacy of medication were best explained by family history of CVD.

For people diagnosed with a single-gene disorder characterized by a family disease history such as FH, family disease history may be more important than DNA information in explaining perceptions of and responses to risk.
Introduction

Cardiovascular disease (CVD) is the leading cause of death in almost every country in the world (WHO, 2005). Most national and international guidelines for CVD prevention recommend the appropriate use of medication and adoption of a healthy lifestyle such as sufficient levels of physical activity in individuals with increased risk for CVD (Smith et al., 2004). Risk assessment is primarily based on gender, age, tobacco use, blood pressure, cholesterol levels and body weight (e.g. Anderson et al., 1991; Conroy et al., 2003; Backer et al., 2003). In addition, CVD-risk assessment can be based on (self-reported) family history of CVD (Murabito et al., 2005; Nasir et al., 2004), reflecting a genetic susceptibility and shared environmental and behavioural factors. Rapid advances in genetic screening technology increase possibilities for using DNA tests for CVD-risk assessment. At present, such tests are mostly used to detect single-gene disorders. The most common single-gene disorder associated with an increased risk of CVD at an early age is Familial Hypercholesterolemia (FH). FH is characterized by a family history of CVD and impaired metabolism of low-density lipoprotein (LDL) cholesterol. In Western countries, the estimated frequency of FH is 1 in 400 - 500 people (Austin et al., 2004). A DNA-based genetic test for FH became available in the early nineties (Hobbs, Brown & Goldstein, 1992).

When using family history information and/ or DNA test results in addition to risk factors such as raised blood pressure and cholesterol levels to estimate CVD risk, it is important to understand how this type of information is perceived and affects people’s preventive behaviour. According to the self-regulation model of health and illness, people construct mental representations of an illness threat based on existing knowledge and beliefs about that threat (Cameron & Leventhal, 2003). These representations may influence the motivation to engage in preventive behaviour. It can be expected that experiencing an illness in the family and/or receiving positive DNA test results establishing a genetic predisposition to a disease will raise the anticipation of a negative health outcome. This may increase motivation to engage in preventive behaviour. On the other hand, since both family history information as well as positive DNA test results specifically refer to a potential genetic origin of the risk, this information might also strengthen genetic attributions of risk, which in turn may weaken attributions to behavioural causes and reduce confidence in the efficacy of behavioural recommendations. People who strongly believe in a genetic origin of risk may have more confidence in biologically-based methods (such as taking medication) to reduce disease risk (Marteau & Weinman, 2006; Senior & Marteau, 2007).

Evidence concerning people’s perceptions of having a family history of CVD and/or an identified genetic predisposition to CVD is limited. Some studies show that having a family history of CVD (Hunt, Davison, Emslie & Ford, 2000; Montgomery et al., 2003; Acheson et al., 2010) or an identified genetic predisposition to CVD (van Maarle, Stouthard & Bonsel, 2003) has the potential to
increase perceived risk. There is also some support that identifying a genetic predisposition to CVD strengthens genetic attributions of CVD while weakening attributions to behavioural causes (Senior, Weinman & Marteau, 2000; Marteau et al., 2004). There is some evidence that the provision of genetic risk information increases perceived efficacy of medication (Wright, Weinman & Marteau, 2003; Marteau et al., 2004; Senior, Marteau & Weinman, 2005; Phelan, Yang, & Cruz-Rojas, 2006; Senior & Marteau, 2007) and decreases the perceived efficacy of a healthy lifestyle (Marteau et al., 2004; Senior & Marteau, 2007). Only a few studies have examined the role of having a family history of CVD and/or an identified genetic predisposition to CVD in relation to preventive behaviour. Some studies found positive associations with medication adherence but no relation with lifestyle adjustments (van Maarle, Stouthard & Bonsel, 2003; McCusker et al., 2004) while others showed no relationship with either medication adherence or lifestyle adjustments (Marteau et al., 2004; Senior & Marteau, 2007; Wright et al., 2007; Elis et al., 2008).

The present paper reports the findings from a study examining the differences in perceived risk and causal attributions of CVD, and in perceived efficacy and adoption of preventive behaviour (medication adherence, adoption of a healthy diet and being sufficiently active) between individuals at risk for CVD, with and without a known genetic predisposition to CVD. In line with the literature, we hypothesized that individuals with a known genetic predisposition to CVD compared to those without a known genetic predisposition to CVD
- have higher perceptions of risk,
- attribute CVD risk more strongly to genetics and less to unhealthy lifestyle,
- have more confidence in the efficacy of medication and less confidence in the efficacy of a healthy lifestyle to reduce risk.

No hypotheses were formulated for preventive behaviour.

Methods
Participants

Two separate samples of participants with increased risk for CVD were used. The ‘genetic predisposition’ sample (n =55; aged 30 - 80 years) consisted of individuals with a known genetic predisposition to CVD, and was recruited among individuals recently diagnosed with FH through DNA testing traced in a nationwide family cascade screening programme by the StOEH (Foundation for Tracing Hereditary Hypercholesterolemia - Dutch acronym). Before testing, participants had received extensive written information about FH (e.g. the consequences and the chance of inheriting the mutation) and been visited by a genetic field worker who provided additional information and discussed treatment options. Test results were communicated by letter, explaining the results in
relation to CVD risk; an information sheet to inform the participants’ GP was enclosed with this letter.

The ‘no genetic predisposition’ sample (n = 49; aged 30-75 years) was recruited among participants in a larger ongoing intervention study. This intervention study was aimed at improving adherence to lifestyle advice in individuals at risk for CVD registered at general practices (Koelewijn et al., 2008). Inclusion criteria were based on the Dutch guidelines for CVD risk management (NHG. M84, 2006). Patients with FH or existing CVD were excluded. In the intervention, CVD risk was assessed and communicated, and preventive options were discussed with each participant.

Participants filled out a postal questionnaire asking about CVD risk factors, perceived risk and causal attributions of CVD, and about perceived efficacy and adoption of preventive behaviour (medication adherence, adoption of a healthy diet and being sufficiently active). Examples of CVD (e.g. heart attack and stroke) were provided in the instruction.

**Measures**

**General characteristics of participants**

Self-reported data were obtained on age, gender, educational level, cholesterol levels, blood pressure and number of first-degree relatives with CVD.

**Perceived risk of CVD**

Two measures were employed to assess perceived risk of CVD. Perceived susceptibility was assessed using two items: “How likely do you think it is that you will get CVD within the next 10 years?” (very likely (1) - very unlikely (7)). “Based on your feelings, what is your chance of you developing CVD within the next 10 years?” (very low (1) - very high (7)). The reliability of this scale (Spearman-brown coefficient ($r_{sh}$)) was .91. Comparative risk was assessed using one item: “What do you think the chance is of you developing CVD within the next 10 years compared to an average man/woman your age?” (a lot lower (1) - a lot higher (7)).

**Causal attributions**

Participants were presented a list possible causes of CVD based on the revised Illness Perception Questionnaire (Moss-Morris et al., 2002) and asked to indicate for each cause the extent to which they believed it could be a cause of CVD (range: definitely not (1) - definitely (5)). For the present study we only selected causal beliefs relating to established risk factors: genetic attributions (two items, “Hereditary, CVD runs in the family” and “predisposition”, $r_{sh} = .61$) and lifestyle attributions (two items to assess attributions of CVD to “unhealthy diet”, and “lack of exercise”, $r_{sh} = .86$), and a separate item to assess attributions to “smoking”.

49
Perceived efficacy of preventive behaviour

Perceived efficacy of medication was assessed in those participants who were prescribed medication (either cholesterol-lowering or antihypertensive medication) with one item: “When I use my medication as prescribed, it will reduce my health risk”). Perceived efficacy of a healthy lifestyle was assessed using two items for exercise and dietary behaviour: “Eating healthily every day would reduce my health risk” and “Taking sufficient exercise at least five times a week would reduce my health risk”, \( r_{sh} = .67 \). Current smokers rated an additional item: “If I quit smoking it would reduce my risk”. Responses were assessed on five-point rating scales; completely disagree (1) – completely agree (5).

Preventive behaviour

Adherence to medication was assessed using two items. In those who reported having high cholesterol: “Do you use cholesterol-lowering drugs?” and in those who reported having high blood pressure: “Do you use blood pressure-lowering drugs?” (response categories: 1= “Yes, as prescribed”, 2= “Yes, but I don’t (always) use them as prescribed”, 3= “No, they are prescribed but I don’t use them”, 4= “No, they are not prescribed”). For each group the level of adherence was calculated by dividing frequencies for response category 1 by the sum of frequencies for response categories 1, 2 and 3.

Adoption of a healthy diet and being sufficiently active was assessed by the sum of participants’ responses to the following items: “I attend to my diet and see to it that I eat healthily every day” and “I see to it that I am sufficiently physical active every day”. Responses were assessed on five-point rating scales (completely disagree (1) – completely agree (5)).

Smoking behaviour was assessed by asking participants to select which best described their smoking behaviour (response categories: “I am a smoker”, “I stopped smoking less than 2 years ago”, “I stopped smoking more than 2 years ago”, “I have never smoked (with any regularity)”. The four categories were dichotomised; smokers were assigned a score of 1, and former and non-smokers a score of 0.

Statistical analyses

Means, standard deviations and frequencies were generated to describe general characteristics of the two samples. T- and \( \chi^2 \)-tests were performed to analyze differences between samples. Exploratory analyses were performed to evaluate missing data and test the assumptions of normality and homogeneity of variance for the outcome measures. Both samples showed low occurrence of missing data (0-4%) for all variables, and no systematic missing data. To correct for non-normality of the distribution (positive skew), the first three response categories (indicating the disagreement with
the statements and the neutral position) for the items assessing causal attributions, perceived efficacy and adoption of a healthy diet and being sufficiently active measures were collapsed (1 through 3 = 1, 4 = 2, 5 = 3).

To assess differences between the samples on perceived risk and causal attributions of CVD and on perceived efficacy and adoption of preventive behaviour \( t \)- and \( \chi^2 \)-tests (for medication adherence and non smoking) were performed. To examine differences between samples in further detail, in particular to assess the contribution of having a known genetic predisposition, in addition to having a family history of CVD and/or to other risk factors, covariance were analysed (using logistic regression analyses for medication adherence and non smoking). ANCOVA and logistic regressions were performed over complete cases with self-reported raised cholesterol levels and raised blood pressure, family history of CVD (0, 1, 2 and more first degrees relatives with CVD) as covariates.

Results

Table 1: General characteristics of participants

<table>
<thead>
<tr>
<th>Genetec predisposition</th>
<th>No genetic predisposition</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>N = 55 (%)</strong></td>
<td><strong>N = 49 (%)</strong></td>
<td></td>
</tr>
<tr>
<td>Age</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mean (sd)</td>
<td>52 (2)</td>
<td>55 (1)</td>
</tr>
<tr>
<td>Gender</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Men (%)</td>
<td>26 (47)</td>
<td>26 (53)</td>
</tr>
<tr>
<td>Education level:</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Low (%)</td>
<td>19 (35)</td>
<td>23 (42)</td>
</tr>
<tr>
<td>Medium (%)</td>
<td>16 (29)</td>
<td>14 (29)</td>
</tr>
<tr>
<td>High (%)</td>
<td>19 (35)</td>
<td>10 (20)</td>
</tr>
<tr>
<td>1st degree relatives with CVD:</td>
<td></td>
<td></td>
</tr>
<tr>
<td>median (range)</td>
<td>2 (0 – 8)</td>
<td>1 (0 – 4)</td>
</tr>
<tr>
<td>0</td>
<td>10 (18)</td>
<td>21 (43)</td>
</tr>
<tr>
<td>1</td>
<td>17 (31)</td>
<td>16 (33)</td>
</tr>
<tr>
<td>2 or more</td>
<td>28 (51)</td>
<td>12 (25)</td>
</tr>
<tr>
<td>Raised cholesterol (%)</td>
<td>53 (96)</td>
<td>22 (45)</td>
</tr>
<tr>
<td>Raised blood pressure (%)</td>
<td>11 (20)</td>
<td>36 (73)</td>
</tr>
</tbody>
</table>

f1 \( \chi^2 \)-test; f2 \( t \)-test; f3 \( \chi^2 \)-test

- Individuals with a known genetic predisposition to CVD, recruited among individuals recently diagnosed with Familial Hypercholesterolemia through DNA testing
- Patients at risk for CVD registered at general practices
- Low: primary school, lower level of secondary school or lower vocational training. Medium: higher level of secondary school, or intermediate vocational training. High: higher vocational training or University
- Assessed by asking participants which, and how many, of their close relatives (parent(s), sibling(s) and/or child(ren)) had CVD
- Assessed by asking participants if they were told their cholesterol level (blood pressure) was too high; using response categories ‘yes’ or ‘no (not that I can remember’

Note: Table 1 includes gender and education level which were not included in the analysis due to small sample sizes.
**Sample characteristics**

General characteristics of the samples are presented in Table 1. Significant differences with regard to self-reported raised blood pressure, raised cholesterol and number of first-degree relatives with CVD were found between samples. Differences in age, gender and education and medication prescription levels were not significant.

**Sample differences in perceptions and preventive behaviour**

Means and standard deviations of perceived risk and causal attributions of CVD and of perceived efficacy and adoption of preventive behaviour of the two samples are presented in Table 2. Perceived comparative risk and genetic attributions of CVD, and perceived efficacy of medication were higher in the ‘genetic predisposition’ sample than in the ‘no genetic predisposition’ sample. The samples did not significantly differ on lifestyle attributions, efficacy of a healthy lifestyle or preventive behaviour.

Table 2: Perceived risk, causal attributions, efficacy of and adoption of preventive behaviour of participants

<table>
<thead>
<tr>
<th></th>
<th>Genetic predisposition</th>
<th>No genetic predisposition</th>
<th>p (^d)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>N =55(^a)</td>
<td>N =49(^b)</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Mean, sd</td>
<td>Mean, sd</td>
<td></td>
</tr>
<tr>
<td><strong>Perceived risk</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Susceptibility</td>
<td>3.46, 1.61</td>
<td>3.24, 1.23</td>
<td>.447</td>
</tr>
<tr>
<td>Comparative risk</td>
<td>4.25, 1.66</td>
<td>3.35, 1.25</td>
<td>.003</td>
</tr>
<tr>
<td><strong>Causal attributions</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Genetic attributions</td>
<td>2.58, 0.47</td>
<td>2.30, 0.62</td>
<td>.009</td>
</tr>
<tr>
<td>Lifestyle attributions</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Unhealthy diet/insufficiently active</td>
<td>2.41, 0.72</td>
<td>2.41, 0.65</td>
<td>.995</td>
</tr>
<tr>
<td>Smoking</td>
<td>2.47, 0.79</td>
<td>2.39, 0.73</td>
<td>.572</td>
</tr>
<tr>
<td><strong>Efficacy of preventive behaviour</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Medication(^b)</td>
<td>2.85, 0.35</td>
<td>2.45, 0.62</td>
<td>.001</td>
</tr>
<tr>
<td>Healthy lifestyle</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Healthy diet/Sufficiently active</td>
<td>2.33, 0.55</td>
<td>2.07, 0.54</td>
<td>.139</td>
</tr>
<tr>
<td>Quit smoking(^c)</td>
<td>2.20, 0.84</td>
<td>2.33, 0.50</td>
<td>.712</td>
</tr>
<tr>
<td><strong>Preventive behaviour</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Medication (in % of prescribed)(^c)</td>
<td>98 (49)</td>
<td>97 (29)</td>
<td>.612</td>
</tr>
<tr>
<td>Healthy lifestyle</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Healthy diet/ Sufficiently active</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Non-smoking (%)</td>
<td>1.67, 1.08</td>
<td>1.39, 1.12</td>
<td>.201</td>
</tr>
<tr>
<td></td>
<td>91 (50)</td>
<td>82 (40)</td>
<td>.145</td>
</tr>
</tbody>
</table>

\(^a\) Individuals with a known genetic predisposition to CVD, recruited among individuals recently diagnosed with Familial Hypercholesterolemia through DNA testing

\(^b\) Patients at risk for CVD registered at general practices

\(^c\) Only assessed in participants that were prescribed either cholesterol-lowering or antihypertensive medication

\(^d\) p-value t-test
Analyses of covariance showed that there were no significant differences between samples when differences in other risk factors, i.e. raised cholesterol, raised blood pressure and family history were taken into account. Individual differences in perceived risk, genetic attributions and perceived efficacy of medication were best explained by family history of CVD. A higher number of first-degree relatives with CVD was positively associated with higher perceived susceptibility risk ($F(1, 92) = 6.28$, $p = .014$, $\eta^2_{p} = .064$), stronger genetic attributions of CVD ($F(1,94 = 5.40$, $p = .022$, $\eta^2_{p} = .054$), and increased confidence in the efficacy of medication ($F(1,84) = 5.58$, $p = .020$, $\eta^2_{p} = .062$).

**Discussion**

This study compared and explained differences in perceptions and responses to CVD risk between individuals at risk for CVD, with and without a known genetic predisposition to CVD. Our results showed that perceived risk, genetic attributions of CVD and perceived efficacy of medication were higher in the sample recently diagnosed with FH through DNA testing than in the sample without a known genetic predisposition to CVD. Individual differences in perceptions were best explained by differences in the number of first-degree relatives that had CVD.

In line with previous studies, we found that participants recently diagnosed with FH through DNA testing perceived their risk of developing CVD as higher (Hunt, Davison, Emslie & Ford, 2000; Montgomery et al., 2003; Acheson et al., 2010), had stronger genetic attributions of CVD risk, and believed more strongly in the efficacy of medication as a means to reduce risk than those with risk profiles based on family history of CVD, cholesterol levels and blood pressure only (Wright et al., 2003; Marteau et al., 2004; Senior, Weinman & Marteau, 2005; Phelan, Yang, & Cruz-Rojas, 2006). These differences however, were best explained by differences in family history of CVD. One explanation is that prior to genetic testing, screenees will already have developed a personal sense of vulnerability that was informed by the salience of their family history (Walter et al., 2004). It is thus feasible that a positive DNA test result did not affect perceptions and responses to risk because it only confirmed existing knowledge and beliefs about risk or because the information was not correctly understood.

In contrast to earlier research (Marteau et al, 2004; Senior & Marteau, 2007), we found no evidence that genetic risk information weakened attributions to lifestyle factors or lowered perceived efficacy of, and adherence to lifestyle recommendations. In our study, participants either with or without a genetic predisposition seemed to be well aware of the multifactorial nature of the risk, and that maintaining a healthy lifestyle in addition to taking medication can be effective in reducing CVD risk. Moreover, a known predisposition to CVD and/or family history was not associated with reduced adherence to lifestyle recommendations.
Considering the role of family history in explaining perceptions of disease risk, family history information might be used as a tool to communicate disease risk. A few studies have assessed the effectiveness of communicating familial disease risk for disease prevention (Qureshi & Kai, 2008; Pijl et al., 2009b; Yoon et al., 2009). There is some evidence suggesting that providing people feedback on their family history information can raise risk awareness and increases adherence to preventive recommendations. However, more studies are needed before a definite conclusion about the effectiveness of the use of family history as a tool for disease prevention can be drawn (Claassen et al., 2010).

Genetic screening technology can be useful as a tool for risk assessment. In the case of single-gene disorders such as FH, DNA testing might exclude a genetic predisposition in people with a strong family history. If the results are correctly understood, genetic testing may also benefit individuals that were previously unaware of their risk (e.g. because of an absence of family disease history), for example by starting early treatment. In those already aware of risk prior to testing, a positive test result may not be novel information and therefore may not alter perceptions of, and responses to, risk. With the rapid increase in testing for multiple susceptibility genes for common chronic diseases, it seems relevant to study the impact of communicating risk based on multiple low-risk gene variants (McBride et al., 2010). For an individual previously unaware of an increased genetic risk, risk information based on DNA test results will be novel information that is likely to affect perceptions and responses to disease risk.

The size and the comparability of the samples limit the generalisability of the results of this study. A primary concern is the exposure of both samples to different types of risk communication. This may have influenced our findings. In the 'non genetic predisposition’ sample, the intervention provided information directly related to this study's subject matter and outcome measures. The ‘genetic predisposition’ sample did not receive such an intervention. However, the ‘genetic predisposition’ sample received similar information; CVD risk was assessed and communicated, and preventive options were discussed with each participant. It seems unlikely that differences in risk communication alone can explain the pattern of our findings. Another limitation of this study is that, we employed a rather broad definition of family history. A stricter definition (e.g. having a first-degree relative diagnosed with CVD before the age of 60) could have provided more accurate risk profiles (Wilson et al., 2009). It should also be noted that while self-reported measures are appropriate for assessing perceived risk, causal attributions and perceived efficacy of preventive behaviour, they might be less suitable for assessing risk factors, and actual behaviour (e.g. because of unawareness, recall bias or social desirability issues) (Newell et al., 1999).

Although these limitations should be considered in the interpretation of the results, the findings suggest that genetic risk information can increase risk perception, strengthen genetic
attributions of risk and the confidence in the efficacy of medication as a means to reduce risk without weakening lifestyle attributions or reducing perceived efficacy of, and adherence to, a healthy lifestyle. For people diagnosed with a single-gene disorder characterized by a strong family disease history such as FH, family disease history may be more important than DNA information in explaining perceptions of, and responses to, risk.
Chapter 5

Fatalistic responses to different types of genetic risk information: exploring the role of Self-Malleability

Abstract

Providing people with genetic risk information may induce a sense of fatalism, the belief that little can be done to reduce the risk. We postulated that fatalism is a function of health risk information and individual differences in self-perception. DNA-based risk information was hypothesized to generate more fatalism than risk information based on family history or non-genetic risk information. Moreover, people who view themselves as more rather than less able to change self-attributes, were hypothesized to respond least fatalistically.

Factor analyses in separate samples were used to construct a five-item “Malleability of Self” measure. Predictive validity of the measure was tested using a within-subjects analogue design. Participants responded to three scenario vignettes in which they were informed of an increased risk of cardiovascular disease (CVD) In Scenario 1, risk was ascertained by DNA-testing, family history and cholesterol testing; in Scenario 2, it was ascertained by family history and cholesterol testing; in Scenario 3, risk was ascertained by cholesterol testing alone. Scenario 1 was associated with least perceived control over cholesterol level and CVD-risk. People who viewed themselves as more able to change self-attributes experienced more control in all three Scenarios.
Introduction

Advances in genomics research have opened the door to new possibilities for prevention, early diagnoses, and treatment in health care. Providing people with information about their susceptibility to a disease may motivate them to engage in risk-reducing behaviours. Because of its personalized nature, genetic information, based on DNA-testing, family history, or both, has the potential to be more motivating than other types of risk-information (Marteau & Lerman, 2001; Collins, Green, Guttmacher, & Guyer, 2003). Alternatively, a genetic susceptibility may be perceived as a fixed, unchangeable self-attribute, especially when it is established by DNA-testing. If so, genetic information and DNA-based information in particular, might trigger feelings of fatalism, the belief that little can be done to change the risk, and adversely affect motivation to engage in risk reducing behaviour (Alper & Beckwith, 1993). Evidence concerning responses to genetic risk information is limited and inconclusive. Some studies show that genetic risks are perceived as less controllable and less preventable (Senior, Marteau, & Peters, 1999; Senior, Marteau, & Weinman, 2000; Marteau & Lerman, 2001; van Maarle, Stouthard, & Bonsel, 2003), while others find no support for this (Wright, Weinman, & Marteau, 2003; Marteau et al., 2004). One emerging finding from this literature is that using DNA to assess risk, make a diagnosis or tailor treatments, may weaken beliefs in the efficacy of preventive behaviour and reinforce biological ways of reducing risk, resulting in a preference for medication as opposed to behavioural means to control or reduce risk. Marteau et al. (2004) found that participants in whom a mutation for Familial Hypercholesterolemia was found believed less strongly in the efficacy of a diet and showed a trend in believing more strongly in the efficacy of cholesterol-lowering drugs, than either participants in whom no mutation was found, or participants who underwent a non-genetic diagnosis. Additional evidence that genetic testing will weaken beliefs in the efficacy of preventive behavior and reinforce biological ways of reducing risk comes from analogue studies. Wright et al. (2003) found that when smokers learn of a genetic predisposition to nicotine addiction they are more likely to choose pharmacological treatment and less likely to use their own willpower when quitting. In addition, (Phelan, Yang, & Cruz-Rojas, 2006) found that genetic attributions for a mental illness were related to increased recommendations for medical treatment and hospitalization.

The extent to which people show fatalistic responses to genetic information is likely to vary among individuals. Whether or not people actually benefit from being informed of their risk status and in turn change their behaviour to reduce their health risks may depend on how they process and evaluate this information. The processing of self-relevant information is guided by the self-concept, a highly interconnected knowledge structure, or mental model, in which different self-beliefs, concerning physical and psychological attributes, are loosely organized with inconsistent and even conflicting beliefs coexisting (Markus & Sentis, 1982; Linville, 1987; Campbell et al., 1996;
Baumeister, 1998). The organization of the self-concept may reflect fundamentally different perspectives on human nature (Chiu, Hong, & Dweck, 1997; Levy, Plaks, & Dweck, 1999; Dweck, 2000). According to Dweck, people with a deterministic, static perspective on human nature see their physical and psychological attributes as fixed over time and situations. Others have a more dynamic perspective and analyze and understand themselves in terms of goals, needs and states of mind and put more emphasis on situational influences. These differences in perspective can result in different patterns of self-relevant information processing. Those with a static perspective selectively attend to self-relevant information that is consistent with a deterministic perspective, and facilitates trait-based judgments about the self, while those with a dynamic perspective are more attentive to information that is consistent with a dynamic perspective (Levy et al., 1999). Processing information about potential health problems may follow the same pattern. Those with a static perspective will be more likely than those with a dynamic perspective to regard a (genetic) susceptibility to a disease and as a physical attribute that is fixed and unchangeable.

In health research measures of perceived control are often used to explain individual differences in responses to health risk information. Control beliefs can be conceptualized at different levels of specificity of generality depending on the context. As a rule, the more specific a construct is to a particular response the stronger the association between those two is (Ajzen & Fishbein, 1974). The disadvantage of using specific measures is that they are tied to a single response and fail to capture the wide range of possible responses to health risk information. Global constructs are more broadly applicable across a wide area of human functioning. Most commonly used in the health domain is the Multidimensional Health Locus of Control scale (MHLC), which can be regarded as a domain specific measure with a medium level of specificity, referring to a broad spectrum of health events and behaviours. The MHLC was developed to tap beliefs concerning the source of control over health and illness; primarily internal or primarily external, specifically a matter of chance (expressing fatalistic beliefs) or under the control of physicians (Wallston, Wallston, & DeVellis, 1978). It is argued that people with strong feelings of internal control over their health are more likely to engage in a various health-promoting behaviours. However, the MHLC has not performed well when applied to predict behaviour (Norman, 1995; Wallston, 2005; Luszczynska & Schwarzer, 2005).

In the present study responses to different types of genetic health risk-information and the link between these responses and individual differences in self-perception are addressed. The first objective of the study was to construct a scale that captures the extent to which people view themselves as able to change self-attributes, the Malleability of Self questionnaire, based on the measure that was developed by Dweck and her colleagues. It can be expected that Malleability of Self is positively correlated with the Internal control scale of the MHLC, i.e. people who view themselves as more able to change self-attributes may also have higher levels of perceived internal
control over health and illness. In addition, correlations with other theoretically different psychological constructs, such as optimism, neuroticism, depression and self-esteem, are expected to be low (Dweck, Chiu, & Hong, 1995). These constructs have been used in some studies explaining perceptions relating to control (Mirowsky & Ross, 1990; Scheier, Carver, & Bridges, 1994b; Judge, Erez, Bono, & Thoresen, 2002).

The second objective was to examine responses to scenario vignettes describing different types of health risk information and to assess the predictive validity of Malleability of Self in explaining these responses. It is hypothesized that responses to health risk information vary with the type of information. If the information includes positive DNA-test results, it is predicted to generate more fatalism, manifested in lower perceptions of control over health risk and a stronger preference for medication as opposed to behavioural means to control or reduce the risk, than when the information is based on family history and/or non-genetic information alone. We also predict an effect of perceived control over health risk on preferences for medication; people who experience more control are more likely to prefer behavioural means to control or reduce the risk.

In addition, it is hypothesized that confronted with a health risk, people who view themselves as more able to change self-attributes experience more control and have weaker preferences for medication as opposed to behavioural means to control or reduce risk than do people who view themselves as less able to change self-attributes. Moreover, it is expected that the amount of fatalism generated by health risk information is moderated by Malleability of Self. Furthermore, we also examine the contribution of Malleability of Self in explaining responses to health risk information relative to the contributions of the MHLC subscales.

**Methods**

**Study design**

Responses to health risk information and predictive validity of Malleability of Self were assessed by examining student responses to scenario vignettes, describing three different levels of genetic health risk information. Scenarios vignettes were developed and questionnaire items were selected based on literature, consensus of experts and pre-tests in a small convenient sample. The vignettes were presented in counter balanced order (to compensate for order effects). After answering the accompanying questions the students were asked to complete measures of the MHLC, optimism, neuroticism, depression and self-esteem. In addition, they were asked to complete a questionnaire intended to capture the extent to which people view themselves as able to change self-attributes, which contained eight items based on Dweck’s “kind of person – self form” (Dweck, 2000), and 19 other items relating to self-perception. This 27–item questionnaire was also administered in a separate out-patient sample. Based on psychometric properties, explorative factor analyses in the
student sample and confirmative factor analysis in the out-patient sample, a Malleability of Self scale was constructed.

Participants
Student sample
A total of 94 students (31 men and 63 women) from the VU University, Amsterdam participated (mean age 21.5 years; range 17-65 years) recruited students from all faculties and years. Participants were informed that the purpose of the study was to examine how people deal with health risks and were then asked to complete a questionnaire. Each participant was paid €5 in gift vouchers after completing the questionnaire.

Out-patient sample (for confirmatory factor analysis only)
An out-patient sample (n=96), representing people at an increased health risk, was recruited from three different medical settings, 32 patients visiting a general practice in a municipality in the northern part of the Netherlands (12 men and 20 women, mean age 50, range 22-71 years), 26 patients visiting the out-patient ophthalmology clinic in a hospital in a city in the southern part of the Netherlands (11 men and 15 women, mean age 67, range 58-81 years), and 38 people visiting an out-patient obstetrics clinic in the VU University Medical Center in Amsterdam (2 men and 36 women, mean age 36, range 20-56 years). Participants were asked to complete a short questionnaire and handed an accompanying letter informing them of the purpose of the study, namely, to examine how people deal with health risks. No payment was offered.

Materials
Measures
Malleability of Self. Roughly following instructions by Dweck (2000) for the “kind of person – self form”, we compiled an eight item scale to capture the extent to which people view themselves as able to change self-attributes (see Table 1). Response categories ranged from 1 = strongly disagree, 2 = disagree, 3 = mildly disagree, 4 = mildly agree, 5 = agree, 6 = strongly agree. Although there are no published data on the validity of this ‘self form’ measure, there is some evidence for the internal and test-retest reliability and construct validity of a “Kind of Person” measure, consisting of three items depicting a static perspective on human nature, e.g. “The kind of person someone is, is something very basic about them and it can’t be changed very much” (Dweck et al., 1995). This original “Kind of Person” measure did not include items depicting a dynamic perspective because the researchers found that many participants who endorsed items depicting a static perspective also endorsed items
of the opposite perspective and shifted toward dynamic choices over items. An expanded measure, containing additional items presenting a strong form of a dynamic perspective, was employed in later studies, e.g. “All people can change even their most basic qualities” (Chiu, Hong, & Dweck, 1997b). This expanded eight item measure proved to be relatively stable over time and situations as well (Dweck, 2000).

Perceived control over health. The 18 item Multidimensional Health Locus of Control Scale (MHLC) validated in Dutch was used to assess control beliefs relating to health and illness on a six-point rating scale (Halfens & Philipson, 1988). The MHLC consist of three six-item subscales: internal control (IHLC), a matter of chance (CHLC) and under the control of physicians (PHLC). Cronbach’s alphas (α) for the subscales for the present sample are: IHLC α = .68, CHLC α = .70 and PHLC α = .73.

Depression. Depression was measured by using the Depression subscale of the Hospital Anxiety Depression Scale, validated in Dutch (Spinhoven et al., 1997). This seven-item measure, using a four-point rating scale, assesses self-reported depressive symptoms. Reliability in the present sample was α = .74.

Self Esteem. The 10-item Rosenberg Self Esteem Scale (Rosenberg, 1965) was used to measure global attitudes about the self. Participants were asked to state to what extent they agreed with positively and negatively worded items, assessed on a four-point rating scale. Reliability in the present sample was α = .83.

Neuroticism. The Dutch version of the neuroticism scale, taken from the Neo-Five Factors Inventory (NEOFFI) (Hoekstra, Ormel, & De Fruyt, 1996), was used to assess neuroticism. The neuroticism scale of the NEO-FFI contains 12 items, scored on a five-point rating scale (sample item “Often I feel tense and nervous”). Reliability in the present sample was α = .81.

Optimism. Optimism was assessed using the Revised Life Orientation Test (Scheier, Carver, & Bridges, 1994a). This 10-item scale (including two filler items) measures generalized expectancies for positive versus negative outcomes, using five-point rating scales. Reliability in the present sample was α = .71.

Vignettes
Participants were first presented with general information about cholesterol and CVD risks. Subsequently they were asked to imagine themselves in three different health situations, presented in counter balanced order, and to answer the accompanying questions.
- In the DNA-scenario (1) participants were first given information concerning an inherited predisposition to developing high levels of cholesterol, entailing a heightened risk for CVD. Next, they were asked to imagine that this condition ran in their own family (established by DNA-testing). The affected family members were prescribed cholesterol lowering drugs and advised to change lifestyle. Subsequently, participants had to imagine they themselves being tested for the mutation and for cholesterol and that both the mutation was found as well as a high level of cholesterol levels.

- In the Family History scenario (2) participants had to imagine a family history of CVD having (not confirmed by DNA-testing). The affected family members were prescribed cholesterol lowering drugs and advised to change lifestyle. The participant was also asked to imagine that this had made him or her decide to check their own cholesterol; high cholesterol levels were found.

- The Cholesterol scenario (3) stated that recently a close (male) friend suffered a heart attack. This friend was prescribed cholesterol lowering drugs and advised to change lifestyle. The participant was again asked to imagine that the incident has made him or her decide to check his or her own cholesterol; high cholesterol levels were found.

**Outcome measures for each vignette**

*Perceived control over cholesterol and CVD-risk.* Perceived ability to lower cholesterol level and prevent CVD, was assessed using six-items, based on the personal control items of the revised Illness Perception Questionnaire (Moss-Morris et al., 2002). Participants were asked to indicate the degree of agreement using a five-point rating scale (e.g. “There is little I can do to prevent me from getting cardiovascular diseases”). Reliability of the control scales was α = .85 for Scenario 1, α = .82 for Scenario 2 and α = .80 in Scenario 3.

*Preferences for medication as means to control cholesterol and CVD-risk.* Pre-tests showed that people are inclined to equally endorse the prevention options that are presented to them. Therefore, we decided to force participants to discriminate between the two prevention options, using only one item. Participants were asked to indicate what they would prefer to do in each situation using a one-item seven-point bipolar scale, labelled at either end: 1 = “I would rather adopt a healthy lifestyle” and 7 = “I would rather use cholesterol lowering drugs”. Because preferences for medication showed skewed results for all threes scenario vignettes, with low frequencies for response categories 5, 6 and 7, the item was transformed into a five-point rating scale, combining the last three categories.
Statistical analyses

An explorative (EFA) and confirmatory factor analysis (CFA) were performed using the Mplus computer program 3.13 (Muthen & Muthen, 2005) which allows for missing data and categorical data. EFA, followed by promax rotation, was conducted over the eight items based on Dweck’s “kind of person-self form” using a selection criterion of eigenvalues >1, on data of the student sample. The other 19 items relating to self-perceptions were not used in the analyses. Since the items are measured on an ordinal categorical scale, we used analyses building on polychoric correlations. To test factor generalizability across samples, a series of multigroup maximum likelihood CFA for categorical variables were performed on the data of the out-patient sample using the factor solution from the student sample (Muthen & Muthen, 1998 -2007). This multigroup analysis approach was used to examine measurement invariance between the student and the out-patient sample in two ways: invariance of factor loadings and item thresholds. Overall fit was tested using goodness-of-fit indices ($\chi^2$/df < 2, Comparative Fit Index (CFI) and Tucker-Lewis Index (TLI) >.95, Root Mean Square Error of Approximation (RMSEA) ≤ .06 and Weighted Root Mean Square Residual (WRMR) < .90) as criteria for acceptance (Hu & Bentler, 1999, Yu, 2002). Based on the interpretation of the results EFA and CFA we constructed a Malleability of Self scale.

Based on missing values pattern in the student sample we excluded one participant from further analyses. In the remaining sample (n=93) the overall percentage of missing was <.001. A response function imputation was performed on the remaining missing values (Van Ginkel & van den Ark, 2007).

Pearsons’ correlations between Malleability of Self and scores on the MHLC subscales were computed to assess convergent validity. Pearsons’ correlations between Malleability of Self, optimism, neuroticism, depression and self-esteem were computed to assess discriminant validity.

The data were restructured so that the outcome variables for each vignette were collapsed into two response variables: perceived control over cholesterol and CVD-risk and preferences for medication with three cases, for each vignette, per participant. A Linear Mixed Models (LMM) approach (SPSS 14.0) was used to examine the influence of scenario and the covariates, Malleability of Self and the three subscales of the MHLC, on perceived control and to examine the influence of scenario and the covariates, perceived control over cholesterol level and CVD-risk, Malleability of Self and the subscales of the MHLC, on preferences for medication. Main effects and two-way interaction effects (between scenario and covariates) were estimated via Maximum Likelihood procedures, under an unstructured repeated covariance structure. We started with the complete model and proceeded by a stepwise removal of non-significant interaction terms, followed by a stepwise removal of non-significant covariates. Bonferroni comparisons were performed to examine the differences between scenarios.
Results

Construction of the Malleability of Self-scale

The five negative phrased items of Malleability of Self, depicting a static perspective, were recoded so that high scores depicted a more dynamic perspective and low scores depicted a more static perspective. After considering the proportions in the six response categories in the student sample, the extreme categories on both ends of the scale were combined. Response categories were thus recoded into four categories. EFA yielded a two-factor solution, explaining 68.6% of the variance. The correlation between the two factors was $r = .60$. Descriptives, factor structure and factor loadings are listed in Table 1.

Table 1: Malleability of Self; descriptives, factor structure and promax rotated factor loadings (student sample, n=94)

<table>
<thead>
<tr>
<th>Items (scale 1-4)</th>
<th>Means</th>
<th>sd</th>
<th>Factor 1</th>
<th>Factor 2</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. The kind of person I am, is something very basic about me and it can’t be changed very much</td>
<td>2.31</td>
<td>1.14</td>
<td>.62</td>
<td>.14</td>
</tr>
<tr>
<td>2. I can do things differently but who I am can’t really be changed*a</td>
<td>2.94</td>
<td>1.04</td>
<td>.63</td>
<td>.20</td>
</tr>
<tr>
<td>3. As much as I hate to admit it, you can’t teach an old dog new tricks; I can’t change my deepest attributes*b</td>
<td>2.18</td>
<td>1.14</td>
<td>.64</td>
<td>.19</td>
</tr>
<tr>
<td>4. I am but who I am and can do only little to change that*c</td>
<td>2.14</td>
<td>1.04</td>
<td>1.03</td>
<td>.19</td>
</tr>
<tr>
<td>5. The kind of person I am, is changing all the time</td>
<td>2.00</td>
<td>.99</td>
<td>.04</td>
<td>.60</td>
</tr>
<tr>
<td>6. I am able to change my basic qualities substantially</td>
<td>2.27</td>
<td>1.07</td>
<td>.11</td>
<td>.59</td>
</tr>
<tr>
<td>7. I can always change the kind of person I am</td>
<td>2.27</td>
<td>1.05</td>
<td>.13</td>
<td>.92</td>
</tr>
<tr>
<td>8. I can’t change myself*d</td>
<td>1.79</td>
<td>.97</td>
<td>.83</td>
<td>.02</td>
</tr>
</tbody>
</table>

Factor correlation: .60

*a Recoded item

All five negative phrased items loaded highly (> .60) on the first factor and the three positive phrased items loaded high (> .58) on the second factor. Subsequently, a CFA was performed on the outpatients sample using the factor solution from the student sample, setting some of the factor loadings to zero and constraining factor loadings and item thresholds to be equal across groups. Goodness-of-fit indices showed a poor fit for this model. Based on the modification indices we decide to remove one of the items (“The kind of person I am, is changing all the time”) and run a second CFA. Although fit indices improved considerably, the modified model still did not fit the data acceptably. Modification indices for the two remaining items of the second factor suggested that the model fit would improve without this factor. Therefore we decided to perform a CFA on a one-factor model with the remaining five negative phrased items. Overall, goodness of fit indices for the third...
model were superior to the fit-indices of the other two models. Although RMSEA was .08, indicative of only a marginal acceptable fit, other indices pointed to good fit of the model ($\chi^2/df$ (15) = 24, $p = .06$, CFI= .993, TLI = .993 and WRMR =.89). Results for the confirmatory factor analyses are presented in Table 2. Based on the findings from the factor analyses, a five-item Malleability of Self scale was constructed (M = 2.27, sd = .82, in the student sample, and M = 2.41 sd =.93 in the outpatient sample). Reliability of the Malleability of Self scale was $\alpha = .83$ in the student sample, and $\alpha = .85$ in the out-patient sample).

Table 2: Confirmatory multigroup factor analyses; Goodness-of-Fit Indices (student sample, n=94, out-patient sample, n=96)

<table>
<thead>
<tr>
<th></th>
<th>$\chi^2$</th>
<th>df</th>
<th>P-value</th>
<th>$\chi^2$/df</th>
<th>CFI</th>
<th>TLI</th>
<th>RMSEA</th>
<th>WRMR</th>
</tr>
</thead>
<tbody>
<tr>
<td>Two-factor model (8 items)</td>
<td>66.6</td>
<td>31</td>
<td>.0002</td>
<td>2.1</td>
<td>.948</td>
<td>.973</td>
<td>.110</td>
<td>1.148</td>
</tr>
<tr>
<td>Two-factor model (7 items)</td>
<td>43.1</td>
<td>25</td>
<td>.014</td>
<td>1.7</td>
<td>.974</td>
<td>.987</td>
<td>.087</td>
<td>.956</td>
</tr>
<tr>
<td>One factor model (5 items)</td>
<td>24.0</td>
<td>15</td>
<td>.06</td>
<td>1.6</td>
<td>.993</td>
<td>.993</td>
<td>.080</td>
<td>.890</td>
</tr>
</tbody>
</table>

Note: CFI = Comparative Fit Index, TLI = Tucker-Lewis Index, RMSEA = Root Mean Square Error of Approximation, WRMR = Weighted Root Mean Square Residual. Criteria for acceptance: $\chi^2$/df < 2, CFI, TLI, > .95, RMSEA ≤ .06, WRMR < .90

*a Recruited in a General Practice (n=32), an Ophthalmology Clinic (n=26) and an Obstetrics clinic (n=38)

Convergent and discriminant validity of Malleability of Self

Descriptives and correlations between Malleability of Self, the three subscales of the MHLC, optimism, neuroticism, depression and self-esteem are presented in Table 3. Contrary to expectation, the correlation between Malleability of Self and IHLC was not significant. Associations between Malleability of Self and the other two subscale of the MHLC were non-significant as well. Although there was a substantial overlap between optimism, neuroticism, depression and self-esteem, correlations between Malleability of Self and these constructs were weak at best.

Table 3: Descriptives and correlations\(^a\) between Malleability of Self and other scales

<table>
<thead>
<tr>
<th>Range</th>
<th>Malleability of Self</th>
<th>MHLC(^c)</th>
<th>Depression</th>
<th>Self-esteem</th>
<th>Neuroticism</th>
<th>Optimism</th>
</tr>
</thead>
<tbody>
<tr>
<td>1-5</td>
<td>2.27 .82 -.13 -.07 -.28 .09 -.17 .13</td>
<td>4.08 .60 -.38 -.32 -.04 .23 -.11 .13</td>
<td>2.62 .70 -.36 .10 -.19 .10 -.16</td>
<td>2.94 .69 -.03 .00 .04 -.03</td>
<td>1.47 .44 -.41 .42 -.31</td>
<td></td>
</tr>
<tr>
<td>1-4</td>
<td>3.16 .42 -.66 .66</td>
<td>2.58 .68 -.67</td>
<td>3.62 .52</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
</tbody>
</table>

\(^a\) Correlations in bold are significant at the 0.05 level (2-tailed)

\(^b\) Range of response categories of scales are indicated between brackets

\(^c\) Multidimensional Health Locus of Control
Responses to the three vignettes and Malleability of Self

There was a main within subject effect of scenario on perceptions of control over cholesterol and CVD-risk ($F(2,93) = 18.8$, $p < .001$). Differences between scenarios were significant for all pair wise comparisons, with lowest perceptions of control in the DNA scenario and highest perceptions of control in the Cholesterol scenario. In addition, we found a positive between-subjects effect of Malleability of Self on control ($\beta = 119$, $t(93) = 2.1$, $p = .037$). That is, students who viewed themselves as more able to change self-attributes experienced more control. However, we didn’t find an interaction effect of Malleability of Self and scenario on perceived control over cholesterol and CVD-risk. Between subject effects of the MHLC-subscale and interaction effects of the MHLC-subscale and scenario on perceived control were all non-significant.

Table 4: Responses to health scenarios: descriptives and summary of Linear Mixed Models analyses, estimates of fixed effects (student sample, $n = 93$)

<table>
<thead>
<tr>
<th>Scenario</th>
<th>Control over cholesterol and CVD risk</th>
<th>Preferences for medication $^b$</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Obs. mean (sd)</td>
<td>Est.</td>
</tr>
<tr>
<td>Intercept</td>
<td>4.530</td>
<td></td>
</tr>
<tr>
<td>DNA</td>
<td>3.85 (0.67)</td>
<td>-.411</td>
</tr>
<tr>
<td>Family History</td>
<td>3.98 (0.60)</td>
<td>-.275</td>
</tr>
<tr>
<td>Cholesterol</td>
<td>4.25 (0.51)</td>
<td>-</td>
</tr>
<tr>
<td>Malleability of Self</td>
<td>-</td>
<td>-.119</td>
</tr>
<tr>
<td>MHLC subscales</td>
<td>-</td>
<td>Not in the final model</td>
</tr>
<tr>
<td>Control over cholesterol and CVD risk</td>
<td>-</td>
<td>-.345</td>
</tr>
</tbody>
</table>

$^a$Scenarios assessment of CVD risk:
1. based on DNA-testing, family history and cholesterol testing
2. based on family history and cholesterol testing
3. based on cholesterol testing

$^b$The Cholesterol scenario is the reference category; the estimates for the first two scenarios contrast the effect of the third scenario.

There was a main within subject effect of scenario ($F(2,100.4) = 13.9$, $p < .001$) on preferences for medication as means to control cholesterol and CVD-risk. With strongest preferences for medication in the DNA scenario compared to the Family History scenario and the Cholesterol scenario. Pair wise comparisons showed no significant difference between the Family History scenario and the Cholesterol scenario. We also found a negative main between subject effect of perceived control on preferences for medication ($\beta = -.345$, $t(248.6) = -2.5$, $p = .013$). That is, students who experienced more control over cholesterol and CVD risk were less likely to prefer medication as means to control...
cholesterol and CVD risk. Between subject effects of *Malleability of Self* and the MHLC-subscases and interaction effects of *Malleability of Self* and the MHLC-subscases and scenario on preferences for medication were all non significant. A summary of the results of the mixed models analyses is presented in Table 4.

**Discussion**

People may react differently to different types of health risk information depending on how they process self-relevant information. The present study provides support for the hypotheses that different types of hypothetical risk information generate differences in perceptions of control over the health risk and behavioural preferences concerning the means to control the risk. Our findings also provide support for the validity of a *Malleability of Self* scale, a viable instrument for capturing the extent to which people view themselves as able to change self-attributes, and partial support for the hypothesis that *Malleability of Self* has predictive validity in explaining individual differences in fatalistic responses to health risk information. However, we found no support for a moderating effect of *Malleability of Self* on differences in responses generated by different types of health risk information.

Using an analogue within subject design, we showed that scenario vignettes, depicting different types of CVD risk information, were effective as a means to induce variance in perceptions of control and behavioural preferences. Overall, average levels of perceived control were relatively high (above scale midpoints), indicating that people did not seem to be very fatalistic in response to the vignettes. However, health risk information that included positive DNA-results generated lower perceptions of control and stronger preferences for medication compared to risk information based on family history and/or non–genetic risk information alone.

Explorative and confirmative factor analysis supported a five-item *Malleability of Self* scale (constructed following instructions by Dweck, 2000). We found no support for an expanded measure containing positive phrased items, presenting a strong form of a dynamic perspective. The lack of shared variance between *Malleability of Self*, the MHLC, and other psychological constructs, including optimism, neuroticism, depression and self-esteem, suggest that *Malleability of Self* may be an independent theoretical construct. Contrary to our hypothesis, we found that people with high *Malleability of Self* scores were not more likely to perceive more control over health and illness, as was assessed with the internal subscale of the MHLC. In addition, low *Malleability of Self* scores did not predict preferences for medication as opposed to behavioural means to control or reduce risk. However, we found that *Malleability of Self* showed some predictive validity in explaining control over cholesterol and CVD-risk. Moreover, people who experienced less control over cholesterol and CVD–risk were more likely to prefer medication as means to control or reduce the risk. None of the
subscales of the MHLC showed predictive value in explaining responses. This is in line previous studies which failed to show a strong link between MHLC and preventive behaviour or avoiding health risks (Wallston, 2005; Luszczynska & Schwarzer, 2005). Therefore, although Malleability of Self may not be an appropriate measure to assess general feelings of control in the health domain, as is measured by the subscales of the MHLC, it may be a better predictor of perceptions of control in response to health risk information which in turn may be the best predictor of specific behavioural responses.

Previous research show mixed results regarding the impact of genetic testing on perceived control and preferences for means to control or reduce risk (Senior, Marteau, & Peters, 1999; Senior et al., 2000; van Maarle et al., 2003; Wright, Weinman, & Marteau, 2003; Marteau et al., 2004; Phelan, Yang, & Cruz-Rojas, 2006). However, none of these studies included variables concerning individual differences as presented in the current study. Based on the present study it is expected that differences in self-beliefs, specifically the extent to which people belief themselves as able to change self-attributes, may help explain some of the variance in perceived control over health risk in other settings.

Our study has some limitations. First, although the vignettes were presented in counterbalanced order, to compensate for order effects, the within-subjects design may have enlarged perceived differences between the vignettes through comparison. Second, it is an analogue study in which young and healthy people were asked to imagine themselves to be at risk for CVD. Although the descriptions of the vignettes can be considered as reasonable approximations of real life situations, for most of the participants in our study these situations will not occur in the near future. It remains to be determined how far the student responses can be generalized to the general population and/or real life situations, in which people respond to risk information concerning their own health. We also need to determine whether our findings can be extrapolated to other clinical contexts and conditions. Today, people can be tested for a large number of genetic conditions, all varying in seriousness and controllability. People may respond differently to health risk information concerning genetic conditions for which preventive options are limited (e.g. early onset Alzheimer’s disease) than to health risk information concerning a genetic condition, like Familial Hypercholesterolemia, with a higher potential for prevention. Third, although according to Dweck (1995, 2000) peoples perspective on human nature, either static or dynamic, is relatively stable over time and situations, we have no data on test-retest stability of our Malleability of Self-scale. In addition, convergent validity was not established. Further research is needed to provide additional evidence for the validity of Malleability of Self.

The results of the current study support the findings from previous studies that giving people feedback on their genotype may induce some (but not complete) fatalism, which may
adversely affect motivation to engage in risk reducing behaviour. Our findings also imply that people vary in how they respond to the information. Therefore, it is important to find ways of communicating health risk information to people without demotivating them to engage in recommended preventive behaviours. Moreover, health professionals providing such information should be aware of individual differences in responses to health risk information. *Malleability of Self,* a measure that assesses the extent to which people believe themselves as able to change self-attributes, may help explain these differences. It has the potential to identify those who are more vulnerable to the fatalistic impact of health risk information in different contexts. This identification may offer a starting point for tailoring health risk information to maximize its behavioural impact. These individuals may need a different approach when it comes to tailoring health risk information.
Chapter 6

Perceived control over diabetes risk and preventive behaviour: the role of family history and Self-Malleability

Abstract
Having a family history of disease may raise risk awareness, thereby promoting preventive behaviour, but it might also induce a sense of fatalism. How people perceive their risk may also depend on Self-Malleability, i.e. the extent to which people view themselves as being able to change. To evaluate the use of family history information as a tool for diabetes prevention, we examined the potential impact of family history and Self-Malleability on control beliefs and preventive behaviour.

The sample consisted of individuals (n=212) at increased risk for type 2 diabetes, aged 57 to 75 years. Participants completed a postal questionnaire assessing family history of diabetes, perceived control over diabetes risk, perceived efficacy and adoption of a healthy diet and sufficient exercise, and Self-Malleability. A structural equation modelling approach was used to analyze relationships between the constructs.

Family history was positively associated with the adoption of healthy diet and sufficient exercise but was not associated with perceived control over diabetes risk or perceived efficacy of preventive behaviour. Higher Self-Malleability was associated with higher perceived control.

Our findings suggest that having a family history of diabetes motivates people to engage in preventive behaviour and does not induce fatalism. However, people with lower Self-Malleability seem more fatalistic towards being able to prevent the disease.
**Introduction**

Epidemiological studies show that a family history is a strong and independent risk factor for type 2 diabetes (Harrison et al., 2003; Valdez et al., 2007). Family history information not only reflects the consequences of multiple genetic factors, but also captures the complex interactions between genetic, environmental, and behavioral factors. Interventions aimed at lifestyle modifications to reduce the risk of diabetes are increasingly being targeted at individuals with a family history of diabetes (Lindstrom et al., 2006; Kinmonth et al., 2008; Pijl et al., 2009b). It has also been suggested to use family history information as a tool to personalize health messages which are potentially more powerful motivators than standardized health messages (Yoon, et al., 2002; Harrison et al., 2003; Claassen et al., 2010b). To evaluate the use of family history as a tool for diabetes prevention it is relevant to understand how people with a family history perceive their risk and how this affects preventive behaviour.

Several health behaviour theories suggest that motivation to engage in preventive behaviour arises from the awareness of risk and the expectation that something can be done to reduce this risk (Weinstein, 1993). This expectation involves beliefs concerning whether and how the illness can be prevented, including beliefs about one’s own capability to change risk and the efficacy of preventive options (e.g. dietary changes). A family history of disease can raise risk awareness and therefore may motivate people to engage in preventive behaviour. However, it has been hypothesized that due to the genetic nature of the risk, family history information may also induce a sense of fatalism, the belief that little can be done to reduce the risk (Alper & Beckwith, 1993; Harrison, et al., 2003; Walter et al., 2005), and/or reduce the confidence in the effectiveness of lifestyle changes in reducing risk (Marteau & Weinman, 2006).

How people perceive their risk and whether or not they actually adapt their behaviour to reduce risk may also depend on how they process and evaluate self-relevant information about disease risk. The way people perceive themselves can affect this process. Basically there are two fundamentally different perspectives on human nature (Dweck, Chiu & Hong, 1995; Chiu et al., 1997; Levy et al., 1999; Dweck, 2000). According to Dweck, people with a deterministic, static perspective see their physical and psychological attributes (e.g. fitness and self-esteem) as fixed over time and situations. Others have a more dynamic, malleable self-perspective. Differences in self-perspective (Self-Malleability) can result in different patterns of self-relevant information processing, including the processing of information about potential health problems (Levy et al., 1999; Claassen et al., 2010). Those with a static self-perspective (low Self-Malleability) will be more likely to regard a susceptibility to a disease as a physical attribute that is fixed and unchangeable than those with a dynamic perspective (high Self-Malleability).
To evaluate the use of family history information as a tool for diabetes prevention, we examined the potential impact of family history and Self-Malleability on control beliefs and preventive behaviour. Based on the theory described above, a model was constructed to describe the relationships between these constructs in cohort of people at increased risk for type 2 diabetes (see Figure 1). The paths represent the proposed relationships between the constructs. In particular, we sought to understand the potential impact of family history on preventive behaviour and control beliefs in people at risk for type 2 diabetes. Having a family history of disease may raise risk awareness, thereby promoting preventive behaviour (path 1), but it might also reduce perceptions of control (path 2a and path 2b). We proposed a positive association between perceived control over diabetes risk and perceived efficacy of preventive behaviour (path 3a) and between perceived efficacy and the adoption of preventive behaviour (path 3b). In addition, we hypothesize that people’s perceptions of control over disease are positively linked to perceived Self-Malleability (path 4).

Figure 1: The proposed model

Methods

Study sample and procedure

The sample consisted of individuals that previously participated in a population-based screening program to identify people with undiagnosed type 2 diabetes (Spijkerman et al., 2002a). Participants in this program first filled out a Symptom Risk Questionnaire. Screen positives (scores ≥ 6) were subsequently biochemically tested for diabetes. For the present study, performed five years after screening, screen positives who did not have diabetes at the time of testing were approached (n=319). Those who gave informed consent and stated that they did not have diabetes were sent a postal questionnaire; 255 individuals returned the questionnaire. The VU University Medical Center
Ethical Committee in Amsterdam approved the study protocol. Participants aged 75 years and older were excluded from the analyses because of the high prevalence of missing data in this age group.

**Measurements**

*Family history of diabetes*
Participants were asked to indicate how many of their first-degree family members were affected by diabetes.

*Preventive behaviour: diet and exercise*
Diet and exercise behaviour was assessed using two items (“I attend to my diet and see to it that I eat healthily every day” and “I see to it that I am sufficiently physically active every day”) (completely disagree (1) - completely agree (5)). In the instructions given to participants, a healthy diet and taking sufficient exercise was explained as eating food with little saturated fat and at least two pieces of fruit and 200 grams of vegetables every day and at least half an hour of moderately intense physical activity, such as walking, biking, swimming and gardening, five days a week.

*Beliefs about control over risk*

*Perceived control over diabetes risk*
Participants were asked to indicate the degree to which they agreed with the statements, based on the revised Illness Perception Questionnaire (Moss-Morris et al., 2002): “There is a lot I can do to prevent me getting diabetes” and “Whatever I do, it will not affect my diabetes risk” (completely disagree (1) - completely agree (5), responses to the latter item were reversed, so that high scores represent more control over risk).

*Perceived efficacy of preventive behaviour*
Perceived efficacy of a healthy diet and sufficient exercise was assessed using two items: “Eating healthily every day would reduce my health risk” and “Taking sufficient exercise at least five times a week would reduce my health risk”. Response categories ranged from 1 = strongly disagree to 5 = strongly agree.

*Self-Malleability*
Perceived Self-Malleability was assessed using a five item questionnaire (e.g. “I am who I am and can do little to change that”), intended to capture the extent to which people view themselves as being able to change (Claassen et al., 2010; Dweck, 2000). Response categories ranged from 1 = strongly disagree to 6 = strongly agree. As indicated by previous research, the first two and the last two
response categories were collapsed and subsequently recoded so that higher scores represented a more dynamic perspective (Claassen et al., 2010).

**Statistical analyses**

Means, standard deviations and frequencies were generated to describe the characteristics of the sample. The extent to which the proposed model described the sample data was tested using Structural Equation Modelling (SEM) in LISREL 8.72 (Joreskog & Sorbom, 2004). With SEM it is possible to simultaneously test multiple dependence relationships (where a dependent variable becomes an independent variable in subsequent relationships within the same analysis). Exploratory analyses were performed to check whether the data met the assumptions of SEM. To correct for non-normality of item distributions, response categories with low frequencies were collapsed (for family history: 0 = 0, 1 = 1, 2 = 2, 3 through higher = 3, for perceived control and adoption of preventive behaviour: 1 and 2 = 1, 3 = 2, 4 = 3, 5 = 4, and for perceived efficacy of preventive behaviour: 1 through 3 = 1, 4 = 2, 5 = 3). Missing value analyses showed low occurrence of missing values for all items (0 - 2%) and no systematic missing data. As recommended by Joreskog and Sorbom (2004), missing values were imputed using the Expectation Maximization approach, a computational algorithm for multiple imputations. For SEM a two-stage approach was employed (recommended by Anderson & Gerbing, 1988). First, in a confirmatory factor analysis, the measurement model was fitted to the data, testing whether the items were adequate indicators for the constructs (latent variables). The measurement model consisted of four latent variables: diet and exercise behaviour, control over diabetes, efficacy of diet and exercise, and Self-Malleability. Items were specified to load on the corresponding variable, and all latent variables were allowed to correlate. Family history was defined as an exogenous variable and was not entered into the measurement model. Adequacy of the measurement model was assessed using multiple fit indices (with Normed Chi-square ($\chi^2/df$) < 2; $p > .05$, Comparative Fit Index (CFI) ≥ .96 and Standardized Root Mean square Residual (SRMR) ≤ .08, as criteria for acceptance (Hu & Bentler, 1999)) and analyses of standard residuals and modification indices. Measurement model fit was further evaluated by examining the estimated factor loadings of the items, correlations between and reliability of the latent variables. Parameter estimates should be significant ($p < .05$; $t$-value = 1.96) and correlations above .80 are indicative for multicollinearity. Reliability of the latent variables was assessed by computing composite reliability (recommended value > .70) and variance extracted (recommended value: > .50). In the second step, the proposed structural model was fitted to the data. The fit of the structural model was assessed with multiple measures of fit ($\chi^2/df < 2$, CFI > .95, SRMR ≤ .08) and significance of parameter estimates.
Results

Sample

Sample characteristics (n = 212) are presented in Table 1. A majority of participants was over 65 years of age (58%) and had a relatively low level of education, i.e. completed primary school, lower level of secondary school or lower vocational training (59%). Almost half of participants (46%) had at least one first-degree family member with diabetes.

Table 1: Self-reported characteristics (n = 212)

<table>
<thead>
<tr>
<th></th>
<th>N (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age - mean (sd), range</td>
<td>66 (5), 57-74</td>
</tr>
<tr>
<td>≥ 65 years</td>
<td>96 (42)</td>
</tr>
<tr>
<td>Gender</td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>112 (53)</td>
</tr>
<tr>
<td>Education¹</td>
<td></td>
</tr>
<tr>
<td>Low</td>
<td>120 (59)</td>
</tr>
<tr>
<td>Medium</td>
<td>58 (28)</td>
</tr>
<tr>
<td>High</td>
<td>26 (13)</td>
</tr>
<tr>
<td>Family history of diabetes²</td>
<td>97 (46)</td>
</tr>
<tr>
<td>Median (range)³</td>
<td>1 (0 – 7)</td>
</tr>
</tbody>
</table>

¹ Low: primary school, lower level of secondary school or lower vocational training. Medium: higher level of secondary school, or intermediate vocational training. High: higher vocational training or university.
² First-degree relative(s) with diabetes.
³ Number of first-degree relative(s) with diabetes.

Fitting the measurement model

The initial measurement model demonstrated poor fit ($\chi^2 = 244.48$, $\chi^2$/df(52) = 4.70; $p < .001$, CFI = .81; SRMR = .087). Analyses of standard residuals and modification indices suggested the elimination of one item assessing Self-Malleability: “The kind of person I am is something very basic about me and can’t be changed very much”. Elimination of this item led to a substantial improvement in fit of the model ($\chi^2 = 48.09$, $\chi^2$/df (39) = 1.23; $p = .151$, CFI = .99; SRMR = .051) supporting acceptance of the revised measurement model. Estimated factor loadings of the items were all significant, indicating that the items were adequate indicators for the corresponding latent variable. Estimated correlations between the latent variables were small to moderate, too low to indicate multicollinearity. Reliability of Self-Malleability and perceived efficacy of a healthy diet and sufficient exercise was adequate. Reliability of perceived control over risk and adoption of a healthy diet and sufficient exercise was below the recommended value, indicating internal inconsistency.
between items. Descriptives, correlations and reliability of the latent constructs in the measurement model are presented in Table 2.

Table 2: Psychometric properties of the measurement model

<table>
<thead>
<tr>
<th>Descriptives</th>
<th>Correlations</th>
<th>Reliability</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean (sd), range</td>
<td>1</td>
</tr>
<tr>
<td>Diet and exercise behaviour</td>
<td>2.7 (0.7), 1 – 4</td>
<td>-</td>
</tr>
<tr>
<td>Control over diabetes</td>
<td>2.6 (0.7), 1 – 4</td>
<td>-</td>
</tr>
<tr>
<td>Efficacy of diet and exercise</td>
<td>1.8 (0.4), 1 – 3</td>
<td>-</td>
</tr>
<tr>
<td>Self-Malleability</td>
<td>1.9 (0.8), 1 – 4</td>
<td>-</td>
</tr>
</tbody>
</table>

*Correlations in bold are significant at the 0.05 level

Fitting the structural model

The proposed structural model was fitted to the data. For the latent variables perceived control and adoption of a healthy diet and sufficient exercise, factor loadings of the corresponding items were fixed to 0.80 and measurement errors allowed to correlate to correct for internal inconsistency. The model indicated an adequate fit ($\chi^2 = 56.47$, df (42) = 1.34; $p = .067$, CFI = .98; SRMR = .072). The results are presented in Figure 2. The strengths of the associations between the constructs are presented through standardized path coefficients.

Figure 2: The structural model

Arrow 1Arrows represent the proposed relationships among measures. Numbers and arrows in bold represent significant relationships (standard path coefficients, $p < .05$)

Except for the paths between family history and perceived control over diabetes risk, and between family history and perceived efficacy of a healthy diet and sufficient exercise, all hypothesized paths in the structural model were statistically significant. The number of family members affected with
diabetes was positively associated with self-reported adoption of a healthy diet and sufficient exercise but not associated with beliefs about control over risk. Perceived control over diabetes risk was positively associated with perceived efficacy of a healthy diet and sufficient exercise, which in turn was positively associated with the adoption of a healthy diet and sufficient exercise. Self-Malleability was positively associated with perceived control over diabetes risk.

Discussion

Respondents with a higher familial risk of diabetes were more likely to report eating healthily and taking sufficient exercise. They were not less likely to believe that getting diabetes could be prevented and disease risk could be reduced by diet and exercise. Individuals however differed regarding the amount of control they believe they have in preventing diabetes, which in turn was related to diet and exercise behaviour. These differences in control are related to personality differences between people, i.e. differences in Self-Malleability.

Previous studies that examined relationships between self-reported family history of diabetes, preventive behaviour, and perceived control show mixed results. In a study by Goetsch and Forsythe (1997), people with a family history reported more attempts to lose weight than those without a family history. Reports on diet and exercise behaviour, however, did not differ between the two groups. Hariri et al. (2006) showed that people with a high familial risk of diabetes were more likely to report making ‘lifestyle changes’ but not more likely to meet recommended exercise guidelines. Baptiste-Roberts et al. (2007) found that African Americans with a family history of diabetes were more likely to consume the recommended amount of fruit and vegetables but were not more likely to attempt weight loss and reduce fat intake than those without a family history. As for perceived control, Acheson et al. (2010) also found that having a family history of diabetes was not associated with reduced perceived control of diabetes. However, other studies suggest that in people with a family history, diabetes is perceived as being less preventable and less controllable (Harwell et al., 2001; Pierce et al., 2001; Pijl et al., 2009a).

The results of the current study supported previous findings from an analogue study on Self-Malleability and control over cardiovascular disease risk (Claassen et al., 2010a). This analogue study showed that people who viewed themselves as being more able to change (higher Self-Malleability), perceived a higher control over disease risk in several scenarios. Moreover, those who experienced more control were more likely to prefer behavioural options as a means to reduce risk.

There are some limitations that should be considered when interpreting the results. Cross-sectional data were used to test the hypothesized model, making it impossible to establish causal relationships. Limiting the participial significance of the findings are the relatively small effects sizes of family history on preventive behaviour and Self-Malleability on perceived control. It should also be
noted that the items that were used to assess perceived control over risk and preventive behaviour may not sufficiently represent the corresponding construct.

Taking these limitations into account, our findings suggest that having a family history may motivate people to engage in preventive behaviour and does not induce a sense of fatalism. Considering the potential motivating impact, family history information may be useful as a tool to promote preventive behaviour in people at risk for diabetes. There is some evidence that providing people with feedback on their family history of diabetes to communicate diabetes risk may also motivate at-risk individuals to change and maintain healthy lifestyles (Qureshi & Kai, 2008; Pijl et al., 2009b). A large clinical trial assessing the effectiveness of a multidisease family history tool as a tool to prevent disease (including diabetes) is currently being evaluated (Yoon, et al., 2009). However, more research is needed before a definite answer to whether and how family history information should be given.

Health professionals providing feedback on family disease history to communicate disease risk should be aware of individual differences in responses to this information. Self-Malleability, a measure that assesses the extent to which people believe themselves as being able to change, may help to address these differences since it has the potential to identify those who have a more fatalistic view towards disease risk. This identification can offer a starting point to personalizing health messages to maximize behavioural impact. People with low Self-Malleability may first need to learn that they can do a lot to prevent themselves from developing a disease such as diabetes. In conclusion therefore, our findings suggest that having a family history of diabetes motivates people to engage in preventive behaviour and does not induce fatalism. However, people with lower Self-Malleability seem somewhat more fatalistic towards being able to prevent the disease.
Chapter 7

Using family history information to promote healthy lifestyles and prevent diseases

Abstract

A family history, reflecting genetic susceptibility as well as shared environmental and behavioural factors, is an important risk factor for common chronic multifactorial diseases such as cardiovascular diseases, type 2 diabetes and many cancers. The purpose of the present paper is to discuss the evidence for the use of family history as a tool for primary prevention of common chronic diseases, in particular for tailored interventions aimed at promoting healthy lifestyles. The following questions are addressed: (1) What is the value of family history information as a determinant of personal disease risk?; (2) How can family history information be used to motivate at-risk individuals to adopt and maintain healthy lifestyles in order to prevent disease?; and (3) What additional studies are needed to assess the potential value of family history information as a tool to promote a healthy lifestyle?

In addition to risk assessment, family history information can be used to personalize health messages, which are potentially more effective in promoting healthy lifestyles than standardized health messages. More research is needed on the evidence for the effectiveness of such a tool.
Background

Clinical trial evidence shows that lifestyle modifications (e.g. weight loss, eating more healthily, increased physical activity and smoking cessation) can reduce the incidence of cardiovascular diseases (Holme et al., 2006), type 2 diabetes (Knowler et al., 2002; Lindstrom et al., 2006) and some types of cancers (Friedenreich, 2001). However, it has become increasingly clear that general health education programs aimed at the whole population have limited effect (Kreuter, Strecher & Glassman, 1999). One way of increasing the effectiveness of these programs is to target interventions at individuals who are at increased risk of developing these diseases.

Most common chronic diseases are multifactorial in nature, resulting from interactions of genetic susceptibility, and behavioural and environmental influences. Susceptibility genes for common chronic diseases are newly discovered each day and advances in screening technology make it possible to screen large populations for multiple susceptibility genes (Hirschorn & Daly, 2005). Currently, several commercial companies, mostly in the US, already offer genomic profiling to the public (for an overview, see Williams, 2009). These companies claim that genetic profiling can provide accurate information about a person’s susceptibility to a range of diseases. They suggest that genetic profiling can support decisions concerning preventive actions, including lifestyle choices (Collins & McKusik, 2001). However, for most genes included in the commercial profiles, the evidence for significant gene-disease associations is insufficient (Janssens et al., 2008). Hence, these tests currently have limited value in predicting disease risk and for developing personalized prevention messages.

Another important source of risk information to which most people have access is the health status of their close relatives (family history). Compared to genetic profiling, family history information has the advantage that it not only reflects the consequences of multiple genetic factors, but also captures the complex interactions between genetic, environmental, and behavioural factors, and may therefore be a better determinant of disease risk than genetic profiling. Epidemiological studies show that a family history is a strong and independent risk factor for cardiovascular diseases (Nasir et al., 2004; Murabito et al., 2005), type 2 diabetes (Valdez et al., 2007), and many cancers (Pharaoh et al., 1997; Isaacs et al., 1995, Matakidou, Eisen & Houlston, 2005; Ziegas & Culver, 2003). In the clinical genetics setting, the value of systematically collecting and interpreting detailed family history information has long been recognized, i.e. for early diagnosis, decisions on genetic testing, and reproductive choices (Bennett, 1999; Wattendorf & Hadley, 2005). In primary care, the collection of family history information has been mainly used for diagnostic purposes in patients exhibiting disease symptoms, referrals for specialist evaluation (e.g. in the case of a suspected Mendelian disorder), and as a psychosocial tool to gain insight into family dynamics (McDaniel, 2005). It has been suggested that the systematic collection and interpretation of family history may also be used
as a tool for the prevention of common chronic diseases (Yoon et al., 2002). This information could not only be used to identify individuals at increased disease risk but also to raise risk awareness and motivate people to engage in risk-reducing behaviours. There is some evidence for the effectiveness of cancer screening on behaviour following family history risk assessment (Muarabito et al., 2001; Codori et al., 2001). However, only a few studies have examined the effectiveness of the use of family history information as a tool to promote a healthy lifestyle for primary disease prevention (Johnson et al., 2005; Qureshi & Kai, 2008; Pijl et al., 2009b; Yoon et al., 2009). The purpose of the present paper is to discuss the evidence for the use of family history as a tool for primary prevention of common chronic diseases, such as cardiovascular disease, in particular for tailored interventions aimed at promoting healthy lifestyles. The following questions are addressed: (1) What is the value of family history information as a determinant of personal disease risk?; (2) How can family history information be used to motivate at-risk individuals to adopt and maintain healthy lifestyles in order to prevent disease?; and (3) What additional studies are needed to assess the potential value of family history information as a tool to promote a healthy lifestyle?

Discussion

**What is the value of family history information as a determinant of personal disease risk?**

In order to determine personal disease risk, clear methods to assess the risk associated with a given family history are required. To this end, epidemiological data need to be translated to individual risk. Several methods have been developed to facilitate interpretation of family history information. A comprehensive risk assessment tool that categorizes individuals into risk strata (e.g. low-, moderate- and high risk) based on family history, has been developed by Scheuner et al. (1997). To stratify individuals into risk groups, detailed information is collected about the number of affected family members, kinship and age of onset of a specific disease. Such tools have been developed for several multifactorial diseases. For example, as part of a public health initiative, the Centers for Disease Control and Prevention (CDC) developed an interactive multi-generational web-based tool to assess familial risk for six diseases (coronary heart disease, stroke, diabetes, and colorectal, breast, and ovarian cancer) (Yoon et al., 2009). Recently, three systematic reviews evaluating family history collection tools for clinical use have been published (Reid et al., 2009, Qureshi et al., 2009, Valdez et al., 2010).

Besides risk stratification based on family history, this information can also be incorporated into a multifactorial risk assessment tool that includes other risk factors such as cholesterol levels and overweight. Examples include the QDScore, an algorithm to calculate diabetes risk (Hippisley-Cox et al., 2009), and the Reynolds Risk Score for cardiovascular disease risk assessment (Ridker et al., 2007). Family history information is generally assessed with a single yes or no question, asking if any
one of the first degree family members has the disease. To each factor associated with the disease, appropriate weights, based on sound epidemiological evidence, are assigned. This multifactorial risk assessment approach has also been incorporated into several guidelines, for example European guidelines on diabetes, pre-diabetes, and cardiovascular disease (EASD, 2007).

In order to use family history information as a determinant of an individual’s disease risk, the accuracy of self-reported family history should be ascertained. Studies show that for breast, colorectal and prostate cancers (Ziogas & Culver, 2003; Qureshi et al., 2007), as well as for cardiovascular diseases (Kardia, Modell & Peyser, 2003; Watt et al., 2000), relatives report with a reasonable degree of accuracy on the disease status of their close family members. The family history of other common chronic diseases, such as type 2 diabetes (Bensen et al., 1999) and ovarian cancer (Ziogas & Culver, 2003; Qureshi et al., 2007), and less close family members (second degree relatives) are often reported with a lower degree of accuracy. The accuracy of self-reports is restricted by awareness and understanding of a condition in a family member. Cultural variation in how family is conceptualized can also affect the accuracy (Burns, McGrath & Edwards, 2009). For example, in many cultures individuals place greater importance on, and have greater knowledge about, one side of the family. Raising public awareness of the importance of family history of cardiovascular diseases, type 2 diabetes and specific types of cancers (e.g. by mass-media campaigns) is likely to increase the accuracy of self-reporting of family history information. Assuming that it is possible to collect fairly accurate information about a person’s family history, more studies are needed to establish how this information should be interpreted to determine a person’s disease risk, and by which methods for translating this information into individual risk assessment are most useful.

**How can family history information be used to motivate at-risk individuals to adopt and maintain healthy lifestyles in order to prevent disease?**

Individuals who are at higher than average or population risk can be offered interventions to reduce or manage disease risk. In addition to medical recommendations, such as genetic testing, early screening (e.g. mammography) and medication, interventions may be aimed at promoting a healthy lifestyle. Often such interventions incorporate standard health messages (such as lose weight, eat more healthily, be more active, and stop smoking). However, the effectiveness of providing individuals who are at higher risk with these type of health messages is limited (Kreuter et al. 1999). Individualized messages tailored to specific characteristics and knowledge of individuals with an increased risk can be more persuasive than standardized (“one-size fits all”) messages (Kreuter et al., 1999; Salovey 2005; Rimer & Kreuter, 2006).
The identification of health and illness beliefs that contribute to an individual’s perception of disease risk may help to determine the key elements for tailoring individualized health messages. Most people with a family history have at least some relevant beliefs and knowledge about their disease risk. According to the Common Sense Model of the self-regulation of health and illness, these fragmentary beliefs are assembled into a mental model of personal disease risk (or illness representations), which people use in interpreting information (Morgan et al., 2002; Leventhal, Benyamini & Brownlee, 1997). Illness representations of people with a family history of a common disease can conflict with the epidemiological risk models of health professionals (Hunt et al., 2000; Walter & Emery, 2005). While lay understanding of personal disease risk may be based upon factors similar to epidemiological knowledge—such as the number of affected relatives, their age at diagnosis, and the level of kinship—other important factors have also been identified (Richard & Ponder 1996; Hunt, Emslie & Watt, 2001; Pierce et al., 2001; Walter et al., 2004; Walter & Emery, 2005, 2006; Pijl et al., 2009a). These include the experience of a relative’s illness, feelings of closeness to the affected relative, and perceived differences between themselves and the affected relative (e.g. gender, age, personality, lifestyle and physical characteristics). Illness representations include beliefs about disease causation and controllability, in particular the influence of genetic and behavioural factors. Often people are not aware that their family history places them at risk. The role of behavioural factors may also be misunderstood. People not only tend to underestimat energy intake and overestimate physical activity (Johansson et al., 1998) but also underestimate the potential consequences of maintaining their current lifestyle, and have little knowledge about preventive options (Pierce et al., 2001). Consequently, people may not be aware of a potential health problem and therefore may not see a need to change.

Prevention programs may be more likely to succeed if they incorporate an exploration of individual pre-existing illness representations alongside epidemiological risk factors (Hall et al., 2007). Since health messages can be tailored to key elements of people’s illness representations, one of the main challenges is to develop messages that fit within people’s illness representations (Marteau & Weinman, 2006). These messages should not only inform people about their disease risk based on what people already know and particularly addressing erroneous beliefs and gaps in knowledge, but should also motivate them to change their lifestyle.

To inform people about their disease risk, it is important that people receive objective and clear feedback on the risk associated with their family history and other risk factors. Often, risk information is communicated using verbal (e.g. high vs. low risk), numerical (frequencies and percentages), or visual (e.g. bar charts and icons) formats. These formats influence the effects on risk perception and subsequent behaviour (Timmermans, 1994; Gerrard, Gibbons & Reis-Bergan, 1999; Thomson, Edwards, Grey, 2005; Timmermans et al, 2004, 2008; Lipkus, 2007). Besides probability
information, people need to receive information about the nature of the risk (causes, consequences and severity) (Gerard et al., 1999), because this information may make people more aware of the consequences of maintaining their current lifestyle, and broaden their illness representations to incorporate a multi-causal explanation of disease risk. Subsequently, individualized prevention messages can be offered that match these explanations, thereby increasing people’s confidence in the effectiveness of specific prevention options (Leventhal et al., 1997). For example, providing smokers with a family history of cardiovascular disease with an explanation of how smoking increases their risk may strengthen their confidence in the effectiveness of smoking cessation in reducing their risk for cardiovascular disease.

There is conflicting evidence about whether being a member of a family with affected relatives already has a positive effect on motivation to adopt a healthy lifestyle (Hariri, 2006; McCusker, et al., 2004; Elis et al., 2008). Few studies have assessed the effectiveness of using family history information as a tool to communicate disease risk and to motivate at-risk individuals to change lifestyles and maintain healthy ones. One study evaluating the use of a family history assessment tool reported increases in yearly medical examinations and blood pressure checks in both high- and average-risk families but did not report on lifestyle modifications (Johnson et al, 2005). According to a cross-sectional study, people who were informed of their familial risk of diabetes (by their doctors) reported greater awareness of risk and lifestyle changes to reduce risk, such as diet and exercise, than those who were not informed of their familial risk (Qureshi & Kai, 2008). In a randomized controlled trial among high-risk individuals with a positive family history of type 2 diabetes, participants who had received additional information on familial risk reported more personal control over preventing diabetes and also reported more healthy eating habits than those who did not receive the additional information (Pijl et al., 2009b). A large clinical trial assessing the clinical utility of a family history tool developed by the CDC is currently being evaluated (Yoon et al., 2009). In this multicenter trial, the effects of the provision of both familial risk assessment and prevention messages on risk perceptions, disease-related attitudes and beliefs, as well as change in health behaviours on members of primary care practices in the U.S. are examined.

**What additional studies are needed to assess the potential value of family history information as a tool to promote a healthy lifestyle?**

A research agenda to evaluate the use of family history information in the prevention of common chronic diseases is currently being carried out in many governmental and academic institutions in the U.S. and elsewhere. This agenda addresses the key elements for assessing the added value of family history information, including the validity and interpretation of the information collected, and the practical barriers to implementation (see also Yoon, Scheuner, Khoury, 2003; Berg et al., 2010) for an
overview of specific research questions). However, this agenda does not specifically address how family history information can be used to motivate people to adopt and maintain a healthier lifestyle. To address the questions that were raised in this discussion, the research agenda should be refined and/or expanded.

In order to develop more effective personalized health messages that fit within people’s mental model of disease risk, further research is needed into their pre-existing illness representations. In addition to qualitative studies, which can provide insight into the content of lay beliefs about personal disease risk, quantitative studies are needed to assess how these beliefs are assembled into a mental model and how they affect preventive behaviour, for example how beliefs about family history and disease causation are linked to beliefs about the effectiveness of preventive options. More studies are also needed to assess whether and how these beliefs differ across individuals. Prior research has shown that beliefs vary across diseases and gender (Qureshi et al, 2005; Wang et al, 2008; Williams et al., 2001), but little is known about cultural and ethnic diversity and the effects of educational level (Hunt et al., 2001).

In order to motivate people to adopt a healthier lifestyle, it may be possible to build on interventions that already have proven successful in changing lifestyle in high-risk individuals (Holme et al., 2006; Knowler et al., 2002; Lindstrom et al., 2006; Friedenreich, 2001), such as in an extended follow-up study evaluating the effects of individualized counselling on lifestyle goals, such as reducing weight and increasing physical activity, in people at risk for diabetes (Lindstrom et al., 2006). Further research is needed to explore the effectiveness of differentiation of health messages based on family history information (high vs. low familial risk), other risk factor information, and to identify the components of the intervention that are most effective in achieving permanent lifestyle changes. It should also be noted that although providing information is usually a necessary prerequisite for behaviour change, it is rarely sufficient to promote change. When designing interventions other determinants, such as the social and physical environment, need to be considered.

In addition, more knowledge is needed about the best mode of delivery of family history risk information. Physicians have reported time restrictions, lack of reimbursement, and the complexity of familial risk interpretation as barriers to the routine and systematic collection and use of family history for disease prevention (Rich et al., 2004). Decision support systems and computer-aided tools that can be self-administered or administered by a nurse practitioner or physician assistant might reduce some of these barriers. In addition, internet-based interventions have the potential to reach more people at lower costs. However, they lack the social support that individuals receive from interpersonal counselling, while disparities in computer skills, internet access and public concerns about internet security may affect response.
Finally, there is the possibility that knowledge of family history will have adverse psychological effects. Some experts consider that informing people about an increased risk based on their family history could induce a sense of fatalism—the belief that little can be done to change the risk—which can decrease motivation to change behaviour (Marteau & Lerman, 2001; Marteau & Weinman, 2006; Claassen et al., 2010). The family history risk information may also evoke anxiety, which can induce maladaptive responses, such as avoidance or denial of the presented information (Case et al., 2005). False reassurance, either because a person has no family history or does not identify with the affected relative, is another potential adverse effect that needs careful examination (Marteau et al., 1996). So far, in studies where familial risk was discussed with people who had a family history of diabetes (Pijl et al., 2009b, Pierce et al., 2000; Qureshi et al., 2001) and colorectal cancer (Legatt et al., 2000; Rose et al., 2004) no (long term) adverse psychological effects were noted. This is in line with the larger literature on the emotional impact of a wide range of health risk assessments (see review by Shaw et al., 1999).

**Summary**

Detailed family history information can be used—along with personal risk factors such as weight and smoking status—as a simple, easily applied and cost-effective tool to determine a person’s disease risk. In addition to risk assessment, family history information can also be used to personalize health messages, which may be more effective in motivating people to adopt and maintain a health lifestyle than standardized health messages. A personalized health message should be phrased in such a way that it fits within the target’s pre-existing beliefs about current health status, possible causes and risk factors, age of onset and course of the disease, magnitude of and potential consequences of the risk, and ways to reduce the risk. In this way, personalized risk information can correct erroneous beliefs, fill knowledge gaps, and reinforce people’s confidence in their ability to change behaviour. The evidence for the effectiveness of using family history information as a personalized tool for disease prevention, in particular for raising motivation to adopt and maintain a healthy lifestyle, is very limited. More research is needed before a definite answer can be given to the question of whether and how family history information should be used and promoted as a tool to motivate people to change their behaviour.
Chapter 8

Discussion
Introduction
The central objective of this thesis was to gain understanding of how people perceive and respond to being at risk for type 2 diabetes (diabetes) and cardiovascular disease (CVD). In particular, how these perceptions and responses relate to awareness of genetic and other risk factors as well as to people’s self-concept (i.e. the way people see and describe themselves). The study project consisted of three complementary parts. First, to explore lay representations (or mental models) of being at risk, semi-structured interviews were held in a small sample of people at risk for diabetes (Pijl et al., 2009a) or CVD (unpublished data). Second, an analogue study was performed to validate a self-concept measure. Results of the interviews and the analogue study were then used to develop a self-administered questionnaire assessing risk factors, different aspects of people’s mental models of being at risk for diabetes and CVD, self-concept, and preventive behaviour. Questionnaire data were collected in three separate samples of people at risk for diabetes and/or CVD, based on family history and other risk factors or based on DNA test results.

In this final chapter, the main findings will be summarized and presented in a theoretical framework. The findings are then discussed in the context of the existing literature and the study’s strengths and limitations. After outlining recommendations for further studies, this chapter closes with a brief overall conclusion and five key messages.

Findings
In this section, the main findings concerning key dimensions of people’s mental model of being at risk, self-concept and preventive behaviour are summarized. The findings concerning the key dimensions of the mental model are structured around causal and control beliefs and perceived susceptibility. Other dimensions of the mental model, i.e. identity, consequences and time line, as well as affective responses, are not explicitly addressed.1 In the last part of this paragraph the findings are presented in a newly proposed theoretical framework.

Causal and control beliefs in relation to risk factors
The number of affected first degree relatives (even more than a positive DNA test result) was associated with stronger genetic attributions and a higher confidence in the efficacy of medication.

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1 It should be noted that beliefs about susceptibility to disease are closely linked to both cognitive dimensions of the mental model as well as to affective responses. The Identity dimension may be adequately described by the relationship between self-reported risk factors and perceived susceptibility, as there are no specific symptoms or specific labels (with the exception of a FH diagnosis) associated with being at risk. Also, perceived susceptibility of developing a disease is likely the most proximal consequence of being at risk. In addition, perceptions of susceptibility are assessed within a 10 years time line. Finally, affective responses such as fear and worry are emotional representations of perceived susceptibility.
The vignette study showed that hypothetical genetic information about personal risk can reduce feelings of control over risk (chapter 5). However, in the at-risk samples awareness of genetic risk factors (positive DNA test results or family history of disease) was not associated with a lowered control over disease risk or confidence in the efficacy of a healthy lifestyles (chapter 4 and 6) nor did it reduce lifestyle attributions (chapter 3 and 4). Causal beliefs were linked to beliefs about the appropriate means to control risk. Those who considered an unhealthy lifestyle an important cause of disease were more likely to consider the adoption of a healthy lifestyle an effective way to reduce disease risk (chapter 3 and 4).

**Perceived susceptibility, risk factors and the role of causal beliefs**

On average participants did not think it very likely that they would get CVD in the next ten years and thought it would even be less likely that they would get diabetes (chapter 2, 3 and 4). The most important risk factor in explaining perceived susceptibility to disease was the number of affected first degree relatives, even more so than a positive DNA test result (2, 3 and 4). Other risk factor information such as advanced age, being overweight and being a smoker, was poorly translated into increased perceived susceptibility to disease (chapter 2). For some risk factors, causal beliefs strengthened the relation between the particular risk factor and perceived susceptibility (chapter 2). Being overweight and believing more strongly that overweight could be a cause of diabetes was associated with increased perceived susceptibility to diabetes. Being a smoker and attributing CVD more strongly to smoking was associated with increased perceived susceptibility to CVD.

**Self-concept and control belief**

The findings from the analogue study provided support for the reliability of the Self-Malleability scale; a self-concept measure assessing the extent to which people view themselves as able to change self-attributes (chapter 5). The lack of shared variance between Self-Malleability and other psychological constructs such as Multi Health Locus of Control, optimism, neuroticism, depression and self-esteem, suggests that Self-Malleability may be an independent construct. Self-Malleability also predicted control over disease risk. That is, participants who viewed themselves as more able to change self-attributes experienced more control (chapter 5 and 6).

**Preventive behaviour**

Self reported medication adherence in those that were prescribed either cholesterol-lowering and/or antihypertensive medication was almost optimal (chapter 3 and 4). Medication adherence seemed higher in participants with genetic predisposition and/or family history of disease (chapter 4). A minority of participants reported being a smoker and less than half of all participants reported to
have adopted a healthy diet and eating habits and being sufficiently active (chapter 3, 4 and 6). The adoption of a healthy lifestyle was positively associated with confidence in the efficacy of a healthy lifestyle and family disease history (chapter 3 and 6). That is people who believed more strongly in the efficacy of a healthy lifestyle and/or had a higher number of affected first degree relatives were more likely to have adapted a healthy lifestyle.

A new theoretical framework
Based on the findings, a new theoretical framework of how people perceive and respond to disease risk is proposed. This framework integrates elements of the Common Sense Model of the self-regulation of health and illness (CSM), namely causal and control beliefs (Leventhal et al., 1997), with relevant constructs from other behavioural theories, perceived susceptibility and perceived control over risk. The CSM describes how people’s mental models of being at risk guide responses for dealing with this risk. In particular, the CSM proposes a symmetry between causal and control beliefs. Many health behavioural theories such as the Health Belief Model, Protection Motivation Theory and Theory of Planned Behaviour suggest that a person’s decision to engage in preventive behaviour arises from a perceived susceptibility to health threats and the expectation that something can be done to reduce this risk (Weinstein, 1993).

Figure 1: Key dimensions of mental models of being at risk for a common chronic multifactorial disease and the relationships with risk factors, self-concept and preventive behaviour: a theoretical framework
The new theoretical framework depicts the key dimensions of people’s mental models of being at risk for a common chronic multifactorial disease and the relationships between these dimensions and risk factors, self-concept and preventive behaviour (Figure 1).

**Proposed relationships**

- Awareness of genetic risk factors strengthens attributions of disease risk to genetic factors which in turn strengthens perceived efficacy of medication as a means to reduce risk.
- Awareness of genetic risk factors does not weaken attributions to an unhealthy lifestyle nor does it reduce perceived efficacy of a healthy lifestyle.
- Stronger attributions to an unhealthy lifestyle raises perceived efficacy of a healthy lifestyle.
- Stronger causal beliefs concerning a risk factor strengthen the relation between that risk factor and perceived susceptibility.
- People who view themselves as more able to change self-attributes (high Self-Malleability) experience more control over disease risk than people with who view themselves as less able to change self-attributes (low Self-Malleability).
- A stronger belief in the efficacy of medication as means to reduce risk contributes to medication adherence.
- A stronger belief in the efficacy of a healthy lifestyle as means to reduce risk contributes to the adoption of a healthy lifestyle.
- There is a dynamic relationship between perceived susceptibility, preventive behaviour, and risk factors. People will be more likely to engage in preventive behaviour if perceived susceptibility to disease is higher. In turn, the adoption of preventive behaviour will reduce risk and perceived susceptibility to disease.

**Discussion of the findings**

**Causal and control beliefs in relation to risk factors**

The results of the study project confirm the findings of other studies with non-patients on causal beliefs for diabetes and CVD showing that most people believe that both lifestyle factors and genetic factors could be a cause of these diseases (French et al., 2001; Walter et al., 2004; Walter & Emery, 2005; Pijl et al., 2009a, 2009b; Sanderson et al., 2010). According to the CSM, mental models of being at risk differ depending on the awareness of risk factors (Leventhal, Kelly & Leventhal, 1999). In particular, awareness of genetic risk factors are suggested to shape causal beliefs; by strengthening genetic attributions of risk and potentially weakening attributions to lifestyle factors (Marteau & Weinman, 2006; Dar-Nimrod & Heine, 2010). The present study project only provided partial
evidence for this hypothesis. Participants with an established genetic predisposition and/or positive family disease history believed more strongly in the genetic origin of risk than participants who did not report these risk factors but still seemed well aware of the contribution of unhealthy lifestyle to disease risk.

Some researchers have argued that, as genes are mostly immutable, being aware of a genetic predisposition or an increased genetic susceptibility to a disease (based on DNA test results and/or on family disease history) could enhance feelings of fatalism, i.e. the belief that little can be done to change the risk (Alper & Beckwith, 1993; Harrison, et al., 2003; Walter et al., 2005; Dar-Nimrod & Heine, 2010). The findings in this study project do not support this hypothesis. Previous studies show mixed results. Some studies suggest that genetic risks of diabetes (Harwell et al., 2001; Pierce et al., 2001; Pijl et al., 2009a) and CVD (Senior et al., 1999; Senior et al., 2005; Marteau & Lerman, 2001; van Maarle, Stouthard & Bonsel, 2003a) are perceived as less controllable and less preventable. Other studies did not support these findings (Marteau et al., 2004; Acheson et al., 2010; Pijl et al., 2009b; see also review Collins, Wright & Marteau, 2010). One explanation for these mixed findings is the heterogeneity in the assessment of perceived control. Consider for example “Now that I have Familial Hypercholesterolemia my cholesterol levels can never be low” (van Maarle et al., 2003a), which refers to control over a specific risk factor, opposed to “There is a lot I can do to prevent me from getting diabetes” (this study project), referring to prevention of disease in general.

As proposed by the CSM, control beliefs, in particular beliefs about the optimal means to reduce risk, were found to be linked to causal beliefs (Leventhal, Benyamini & Brownlee, 1997; Marteau & Weinman, 2006). In line with the findings of Senior and Marteau (2007) genetic attributions of disease risk seemed to strengthen the efficacy of taking medication which is assumed to affect the biological processes invoked by gene variations. Also, the adoption of a healthy lifestyle was considered more effective in reducing risk when an unhealthy lifestyle was perceived as an important cause of disease.

It has been suggested that awareness of a genetic risk may weaken the confidence in the efficacy of a healthy lifestyle as means to reduce risk (Marteau et al., 2004). However, the few studies evaluating the impact of communicating genetic risk information on perceived effectiveness of behavioural intentions have not demonstrated such adverse effects (see review Collins et al., 2010). In line with these findings, the present study project showed that participants were well aware of the fact that maintaining a healthy lifestyle in addition to taking medication could be effective in reducing risk, irrespective of the type of risk.

**Perceived susceptibility, risk factors and the role of causal beliefs**
Confirming findings in previous studies (Senior et al., 2002; van Maarle, Stouthard & Bonsel, 2003b; Adriaanse et al., 2008; Wang et al., 2009; Acheson et al. 2010), participants demonstrated relatively low perceptions of susceptibility to diabetes and CVD. Given that participants in the three samples were all at increased risk for either diabetes and/or CVD, the findings suggest that a considerable proportion underestimated their own susceptibility to disease. A common explanation is that in general, people show an optimistic bias when considering their own health risk (Weinstein, 1987). People believe that they are healthier than others, not because they have inaccurate knowledge about the major risk factors but because they do not correctly apply this knowledge to themselves. In particular, they believe their actions contributing to risk (e.g. smoking) are outweighed by their actions to prevent risk (e.g. engaging in sufficient physical activity). This underestimation of risk can have a negative impact on people’s decision to engage in prevention behaviour.

Family history of disease was the strongest factor in explaining susceptibility beliefs which corresponds with findings in previous studies on risk perceptions of diabetes (Forsyth & Goetsch, 1997; Harwell et al., 2001; Hariri et al., 2006; Adriaanse et al., 2008; Gallivan et al., 2009) and CVD (Marteau et al., 1995; Hunt et al., 2000; Montgomery et al., 2003; Frijling et al., 2004). Interestingly, additional risk information based on DNA test results did not contribute to susceptibility beliefs. A plausible explanation is that people receiving DNA test results have some difficulty understanding the implications of this information. Another possibility is that prior to genetic testing, screenees may already have developed a personal sense of vulnerability that is a mainly informed by their family history (Walter et al., 2004). It is thus feasible that a positive DNA test result did not affect perceptions to risk because it only confirmed existing knowledge and beliefs about risk.

Stronger causal beliefs concerning being overweight and being a smoker strengthened the relation between risk factor and perceived susceptibility to diabetes and CVD respectively. For other risk factors no such an effect was found. These findings partly support the hypothesis that self-reported risk factors only raise perceived susceptibility to disease if they fit within beliefs about disease causation (Leventhal et al., 1999; Weinstein, 1999; Cameron, 2003, 2008).

**Self-concept and control beliefs**

In health research, several measures of perceived control have been used to explain individual differences in perceptions and responses to being at risk. Control beliefs can be conceptualized at different levels of specificity of generality depending on the context. As a rule, the more specific a construct is to a particular response, the stronger the association between the two (Ajzen & Fishbein, 1974). The disadvantage of using specific measures is that they are tied to a single response and fail to capture the wide range of possible responses. Global constructs are more broadly applicable across a wide area of human functioning. The Multidimensional Health Locus of Control scale
(MHLC), a commonly used measure to explain perceived control in the health domain, has not performed well when applied in the context of preventive behaviour (Norman, 1995; Wallston, 2005; Luszczynska & Schwarzer, 2005). The findings of the present study project suggest that Self-Malleability, a self-concept measure, may be a marginally better predictor of perceptions of control over disease risk.

**Preventive behaviour**

The findings suggest that having a family history motivates people to engage in preventive behaviour by raising perceived susceptibility. The potential implications of these findings are discussed in chapter 7. There is some supporting evidence from other studies that having a family history raises motivation to adopt a healthy lifestyle. For example, Hariri and colleagues (2006) found that a family history of diabetes was positively associated with making lifestyle changes to prevent diabetes. Some studies also suggest that providing people with feedback on their family history to communicate disease risk may motivate people to change and maintain healthy lifestyles (Qureshi & Kai, 2008; Pijl et al., 2009b).

The adoption of preventive behaviour was not directly linked to perceived susceptibility. A likely explanation is that a person who is aware of risk will be motivated to reduce the impact of modifiable risk factors by engaging in preventive behaviour. This behaviour will reduce risk and perceived susceptibility.

A previous study assessing the effects of DNA based risk information compared to non-DNA information found positive effects on dietary behaviour but no effects on physical activity and medication (Marteau et al., 2004). In this study project, there was no evidence for an effect of DNA information in addition to family history information on preventive behaviour. As suggested above, it is feasible that DNA test results just confirmed existing knowledge and beliefs about risk and therefore did not affect responses.

**Strength and limitations**

**Strengths**

The CSM offers a robust theoretical framework for identifying the content of people’s mental models of being at risk, which may help to examine the potential mismatches between perceptions of and responses to risk (Leventhal et al., 1997). Only a few studies have applied the CSM to understand perceptions and responses of unaffected individuals at risk of developing an illness (Decruyenaere et al., 2000; Rees et al., 2004; van Oostrom et al., 2007; Kaptein et al., 2007; Cameron, 2008). This is first study project that employed the CSM as framework to assess, analyse and explain people’s perceptions and responses to being at risk for diabetes and CVD.
Another strength of the study project is that three separate samples of individuals at risk for diabetes and/or CVD were recruited. Therefore it was feasible to assess the impact of different types of risks (based on positive DNA test results establishing a genetic predisposition, family history and/or other risk factors) and to compare perceptions of and responses to risk across diseases.

**Limitations**

One limitation of the present study is that it is based on cross-sectional data and therefore does not show the dynamic and recursive relationships between people’s mental models of being at risk, risk factors, self-concept, and preventive behaviour. For example, if a personal risk factor is seen as increasing disease risk and/or as an important cause of disease, than a person may be motivated to engage in preventive behaviour, which in turn may reduce risk.

Limiting the generalization of the results is the unknown representativeness of the samples. For example, one sample consisted of low educated elderly individuals who participated in a population based diabetes screening program some years earlier. Their responses may be biased by the salience of diabetes risk in their memory and not accurately characterize responses in the general population.

Another limitation is the poor validity of some of the measurements employed in this study project. It should also be noted that while self-reported measures may be appropriate for assessing causal beliefs, control beliefs, and perceived susceptibility risk, they might be less suitable for assessing risk factors and actual preventive behaviour.

**Implications for further studies**

This thesis may help to understand how people perceive and respond to the being at risk for diabetes and/or CVD. However, more research is needed to further disentangle causal relations between mental models of being at risk for diabetes and CVD, risk factors, self-concept and preventive behaviour. For example, the evidence for the relation between genetic attributions, perceived efficacy of medication and medication adherence is not very strong. The underlying assumption that taking medication in particular can counteract the detrimental effects of gene variations needs closer examination. Another weak link in the framework is the role of self-concept (i.e. Self-Malleability). More studies are needed to assess whether and how key dimensions of mental models of being at risk differ across individuals. Although prior research showed only marginal effects of traits such as Health Locus on Control on control beliefs concerning disease risk, other personality traits or dispositions could be important in explaining individual differences (Contrada & Couper, 2003).

To examine actual effects of risk factor information on perceptions and responses, it is essential to assess the relevant aspects in a prospective study with follow-up. A prospective study
may especially offer more insight into the relationship between perceived susceptibility and preventive behaviour. This relationship is likely to be reciprocal, i.e. a person who feels at risk may decide to engage in preventive behaviour, and this behaviour may reduce perceived risk. Such prospective studies are also needed to explore the effects of differentiation of health messages based on DNA results, family history and other risk factor information.

Studies are also needed to determine whether the findings can be extrapolated to other multifactorial conditions, a broader clinical context and other populations. Today, people can be tested for a large number of genetic conditions, all varying in seriousness and controllability. People may respond differently to genetic risk information when preventive options are limited (e.g. early onset Alzheimer’s disease) than when there is a higher potential for prevention, like Familial Hypercholesterolemia. Study designs may not only include other monogenetic disorders, characterized by a strong family history (e.g. Maturity-Onset Diabetes of the Young and autosomal dominant inherited cardiomyopathies), but could also include genetic conditions that are not characterized by a strong family history (e.g. with an autosomal recessive inheritance pattern) or conditions that involve multiple low or intermediate risk gene variants.

Adverse effects of providing risk information also need further consideration. For example, studies are needed to assess the effects of negative (favourable) DNA test results that may inadvertently cause false reassurance, i.e. the belief that there is no disease risk, which can discourage risk reducing behaviour.

Conclusion
With the rapid developments in the field of health screening technology, people are increasingly confronted with new information about their susceptibility to a range of diseases, including genetic risk information based on DNA testing an/or family history. Little is known about how people make sense of this information. In addition, there is some debate on whether or not people benefit from information about their susceptibility to a disease. Specifically, it has been argued that once people consider an underlying genetic foundation for a condition, a particular set of genetic essentialist biases is triggered (Alper & Beckwith, 1993; Dar-Nimrod & Heine, 2010). These biases may lead people to perceive genes as the sole cause of the condition, and to view the risk for conditions associated with these genes as immutable The findings of this study project suggest that genetic risk information does not reduce attributions to lifestyle factors, nor does it induce a sense of fatalism or lower people’s confidence in the efficacy of preventive options. Moreover, having a positive family history of diabetes and/or CVD is likely to raise perceived susceptibility to disease which may increase motivation to engage in healthy lifestyles. There was no evidence for an effect of DNA information in addition to family history information on people’s perceptions of and responses to
being at risk. As for other risk factors, people only partially use their knowledge of disease causation to link self-reported risk factors to an increased susceptibility to diseases. Self-Malleability, a self-concept measure that assesses the extent to which people believe themselves as able to change self-attributes, does not seem to play an important role in explaining perceptions of control.

**Key messages from this study project**

Disease prevention programs or interventions are often ineffective. One way to increase the effectiveness is to select and deliver tailored or individualized messages (Noar, Benac & Harris, 2007). Such interventions may incorporate personal disease risk information to improve people’s understanding of their disease risk and help them to make better decisions regarding their health. Tailoring risk messages in this way may involve assessing key dimensions of people’s mental model of being at risk. Subsequently, a tailored message, prepared to match these dimensions, can be delivered to each individual (Lauver et al., 2002), for example, with the help of decision support systems and computer-aided tools, either administered in face-to-face consultations with a health professional or through the Internet (e.g.: Koelewijn-van Loon et al., 2009; Broekhuizen et al., 2010).

The precise content of an individualized intervention is not predetermined but rather develops as a result of a counsellor-counselee interaction

Five general inferences can be drawn from the present study project that could be considered when developing tailored and personalized risk messages:

1. **Awareness of a genetic susceptibility does not induce fatalism or reduce confidence in efficacy of a healthy lifestyle.**

   There have been some concerns that informing people about genetic risk factors (i.e. based on DNA test results and family disease history) may inadvertently reduce people’s confidence in the efficacy of risk reducing behaviour. There is little evidence to support these concerns.

2. **Improved knowledge about disease causation raises the awareness that a risk factor contributes to a higher disease risk.**

   To address misconceptions and improve understanding of risk information, people may need to be educated on the nature and causes of risk, in particular how risk factors can contribute to the development of disease.

3. **Having a family history increases risk awareness and motivates people to engage in preventive behaviour.**
The systematic collection and interpretation of family history might be used as a tool to identify individuals at increased disease risk, and to raise risk awareness and motivate people to engage in risk-reducing behaviours such as the use of medication and lifestyle modifications.

4. *For conditions characterized by a strong family history of disease a positive DNA-test result does not change risk awareness and motivations to engage in preventive behaviour.*
   A positive test result may not be novel information to people already aware of risk prior to testing. Genetic testing could benefit individuals that were previously unaware of their risk (e.g. because of an absence of family disease history), provided that the results are correctly understood. In addition, in the case of a suspected single-gene disorder, DNA testing might exclude a genetic predisposition.

5. *People with lower Self-Malleability seem somewhat more fatalistic towards being able to prevent the disease.*
   General measures of personality traits or dispositions such as Self-Malleability (the extent to which people believe themselves as able to change self-attributes), only explain a small amount of the differences in perceptions control. However, health professionals providing health information should be aware of individual differences in perceived ability to change risk. People may first need to learn that they have the ability to prevent themselves from developing a disease such as diabetes and CVD.
Summary
Summary

With the rapid developments in the field of health screening technology, people are increasingly confronted with new information about their susceptibility to a range of diseases. This information includes genetic risk information based on DNA testing or family disease history and information on other risk factors such as advanced age, raised cholesterol levels, high blood pressure, (central) obesity and being a smoker. Whether or not people benefit from these developments may not only depend on the changeability of risks, i.e. risk reducing options, but also on how people think of the risk and perceive themselves in relation to these risks ("The Risky Self"). People will try to make sense of their at-risk status by integrating new information concerning their health into a mental model that already exists in their minds. This model includes beliefs about the magnitude, nature, sources and controllability of the risk. The process of integrating risk information is influenced by people’s ideas about who they are as a person, i.e. the self-concept. People’s mental model of being at risk will also guide the identification and potential use of appropriate means to reduce risk. That is, people may only adhere to preventive recommendations if the recommendation corresponds with their representations of risk.

The central objective of the study project was to gain understanding on how people perceive and respond to being at risk for two common chronic diseases: type 2 diabetes (T2D) and cardiovascular disease (CVD). In this thesis the following questions are addressed:

1. How do people perceive being at risk for diabetes and CVD?
2. How is the awareness of different types of risk factors, in particular genetic versus other risk factors, and people’s self-concept related to mental models of being at risk for diabetes and CVD and to preventive behaviour?

Data were collected from three separate samples at risk for diabetes and/or CVD: individuals recently diagnosed with Familial Hypercholesterolemia (FH) based on DNA testing (sample 1; n = 81), individuals recruited among patients (registered with general practices) with a suspected high risk for CVD, participating in an ongoing intervention study aimed at improving patient adherence to lifestyle advice (sample 2; n = 49), and individuals who had taken part in a population based screening program to identify people with undiagnosed T2D, five years earlier. All participants were identified as at risk during the screening, based on family history and other risk factors, but did not have T2D at the time of testing, nor at recruitment in the present study project.

1 As most lay persons do not distinguish between different forms of diabetes we use the generic term diabetes when referring to people’s illness beliefs.
Chapter 2 describes a study that examined self-reported risk factors, causal beliefs and perceptions of diabetes and CVD risk in at-risk individuals (sample 3). In this study, participants demonstrated a better knowledge about the causes of CVD risk than about the causes of diabetes risk. Although family disease history was associated with higher perceptions of diabetes and CVD risk, in general, self-reported risk factors were only partially translated into perceptions of risk. In particular, being overweight and smoking was not associated with higher perceived disease risk of diabetes and CVD, respectively. However, being overweight and believing that overweight could be a cause of diabetes was associated with higher perceptions of diabetes risk. Similarly, believing that smoking could be a cause of CVD and being a smoker was associated with higher perceptions of CVD risk.

Chapter 3 describes the mental model of being at risk for CVD and preventive behaviour of individuals diagnosed with FH (sample 1). In general, participants in this study seemed to underestimate their risk of developing CVD. Although participants almost equally endorsed both genetic and lifestyle attributions of CVD, they viewed the adoption of a healthy lifestyle as somewhat less effective than medication in reducing risk of CVD. Moreover, while they reported almost optimal adherence to medication they did not always show adequate adherence to lifestyle recommendations. Participants with a stronger family history, i.e. a higher number of 1st degree relatives affected by CVD, perceived a higher risk and were more likely to adhere to lifestyle recommendations.

Chapter 4 describes a cross-sectional study that compared and examined differences in the mental model of being at risk for CVD and preventive behaviours of at risk individuals with (sample 1) and without an established genetic predisposition to CVD (sample 2). Risk perceptions, genetic attributions of CVD and efficacy of medication were higher in the sample with an established genetic predisposition than in the sample without an established genetic predisposition. However, these differences were best explained by individual differences in the number of first-degree relatives that had CVD and not by sample per se (i.e. positive DNA test result). The samples did not differ on lifestyle attributions, efficacy of a healthy lifestyle, or preventive behaviour.

Chapter 5 reports the results of an analogue study testing the hypothesis that fatalistic responses to risk information, i.e. the belief that little can be done to change risk, is a function of type of risk information and differences in self-concept (Self-Malleability). In particular, DNA-based risk information was assumed to generate more fatalism than risk information based on family history or non-genetic risk information only. Moreover, people who view themselves as more rather than less
able to change, were hypothesized to respond least fatalistically. Participants responded to three scenario vignettes in which they were informed about an increased risk of CVD: ascertained by DNA-testing, family history and cholesterol testing, by family history and cholesterol testing, and by cholesterol testing alone. The DNA-scenario triggered most fatalistic responses; it was associated with least perceived control over CVD-risk. People who viewed themselves as more able to change responded less fatalistically, they experienced more control in all three scenarios.

Chapter 6 describes the results of a study examining the potential impact of family history and Self-Malleability on control beliefs and preventive behaviour in individuals at risk for T2D (sample 3). The study employed a structural equation modelling approach to analyze all relationships between the constructs simultaneously. Family history was positively associated with the adoption of a healthy diet and sufficient exercise but was not associated with perceived control over diabetes risk or perceived efficacy of preventive behaviour. Higher Self-Malleability was associated with higher perceived control.

Chapter 7 discusses the evidence from other studies for the use of family history as a tool for primary prevention of common chronic diseases (i.e. T2D, CVD and several types of cancer), in particular for tailored interventions aimed at promoting healthy lifestyles. There is ample evidence that detailed family history information can be used - along personal risk factors such as weight and smoking status - as a simple, easily applied and cost-effective tool to determine a person’s disease risk. In addition to risk assessment, family history information may also be used to personalize health messages, which are potentially more effective in motivating people to adopt and maintain a healthy lifestyle than standardized health messages. A personalized health message should be phrased in such a way that it fits within the target’s pre-existing mental model of being at risk. In this way, personalized risk information could correct erroneous beliefs, fill knowledge gaps, and reinforce people’s confidence in their ability to reduce risk by changing behaviour. The evidence for the effectiveness of using family history information as a personalized tool for disease prevention, in particular for raising motivation to adopt and maintain a healthy lifestyle, is limited. More research is needed before a definite answer can be given to the question of whether and how family history information should be used and promoted as a tool to motivate people to change their behaviour.

In the final chapter of this thesis, the main findings are discussed. Overall, the study findings suggest that genetic risk information does not induce a sense of fatalism nor that it lowers people’s confidence in the efficacy of preventive options, in particular of adopting a healthy lifestyle. Moreover, having a positive family history of diabetes and/or CVD is likely to raise perceived
susceptibility to disease and increase motivation to engage in healthy lifestyles. There was no evidence for an effect of positive DNA information (confirming a genetic susceptibility) in addition to family history information. Besides family history, beliefs about disease causation have an impact on perceived susceptibility to diseases. Self-Malleability, a self-concept measure that assesses the extent to which people believe themselves as able to change self-attributes, plays a minor role in explaining perceptions of control. Although more research is needed our findings should be considered when developing tailored and personalized risk messages.
Samenvatting
Wat mensen denken over en doen met hun risico op diabetes en hart- en ziekten; de rol van genetische risico informatie en het zelfconcept

Door de snelle ontwikkelingen op het gebied van medische screeningstechnologie worden mensen in toenemende mate geconfronteerd met nieuwe informatie over allerlei mogelijke gezondheidsrisico’s. Ook genetische informatie, gebaseerd op DNA tests of familiegeschiedenis, maakt deel uit van deze nieuwe informatie. Of en in welke mate mensen baat hebben van deze ontwikkelingen, hangt niet alleen af van de mogelijkheden om de gezondheidsrisico’s te controleren maar hangt ook van de wijze waarop mensen zichzelf zien in relatie tot genetische en andere risico’s (‘The Risky Self’). Mensen proberen hun gezondheidsrisico’s te begrijpen door nieuwe informatie over hun gezondheid te integreren in het bestaande mentale model dat zij hebben gevormd over het risico. Dit model bevat ideeën over de hoogte, aard en oorsprong, en controleerbaarheid van het risico. Het verwerken van gezondheidsinformatie wordt beïnvloed door ideeën over wie men is als persoon: ‘het zelfconcept’. Hoe mensen over hun risico denken, heeft invloed op wat ze doen om het risico te verminderen. Aanbevelingen voor preventief gedrag worden eerder opgevolgd wanneer ze goed passen bij de ideeën over het risico. Met dit onderzoeksproject werd geprobeerd meer inzicht te krijgen in hoe mensen hun risico op diabetes en hart- en vaatziekten (HVZ) ervaren en er mee omgaan.

De gegevens voor het onderzoek werden verzameld uit drie verschillende datasets van gezonde personen met een verhoogd risico op diabetes en/of HVZ. Dataset 1 bestond uit 81 personen die recent met behulp van een DNA test gediagnosticeerd waren voor Familiaire Hypercholesterolemie (een erfelijke aandoening die geassocieerd is met een verhoogd risico op HVZ). Dataset 2 (50 personen) werd geworven onder personen met een verhoogd risico op HVZ vastgesteld in een interventie studie in verschillende huisartsenpraktijken. Dataset 3 (255 personen) werd geworven uit personen die vijf jaar eerder hadden deelgenomen in een screeningsprogramma, gericht op de identificatie van personen met ongediagnosticeerde diabetes. Deze deelnemers bleken destijds geen diabetes te hebben maar hadden wel een verhoogd risico op T2D (en HVZ).

In Hoofdstuk 2 wordt de relatie tussen risicofactoren, ideeën over oorzaken van diabetes en HVZ en het ervaren risico beschreven van personen met een verhoogd risico op diabetes en HVZ (dataset 3). In het algemeen waren de deelnemers beter op de hoogte van de oorzaken van HVZ dan van diabetes. Zelfgerapporteerde risicofactoren werden slechts gedeeltelijk vertaald in een hoger ervaren risico. Het ervaren risico op diabetes (respectievelijk HVZ) hing samen met het hebben van een familiegeschiedenis van diabetes (HVZ). Andere leefstijlgerelateerde risicofactoren zoals het hebben van overgewicht (een belangrijke risicofactor voor diabetes) en roken (een belangrijke risicofactor voor HVZ) werden doorgaans niet geassocieerd met een verhoogd risico. Echter, mensen
met overgewicht die zich realiseerden dat overgewicht een oorzaak kan zijn van diabetes schatten hun risico wel hoger in. Het zelfde geldt voor rokers; zij schatten hun risico op HVZ hoger wanneer ze zich er bewust van waren dat roker een oorzaak kan zijn van HVZ.

In **Hoofdstuk 3** wordt het mentale model voor het risico op HVZ bij mensen met een genetische aanleg voor HVZ (dataset 2) beschreven. De deelnemers noemen als mogelijk oorzaak voor het ontstaan van HVZ zowel genetische als leefstijl factoren (roken, te weinig bewegen, ongezonde voeding). Een gezonde leefstijl beschouwden ze als iets minder effectief in het verlagen van het eigen risico dan het trouw gebruiken van medicijnen. In het algemeen, leken de deelnemers hun risico op HVZ te onderschatten. De zelfgerapporteerde medicijntrouw was nagenoeg optimaal. Ook was het aantal rokers aanzienlijk lager dan in de algemene bevolking. Echter, minder dan de helft van de deelnemers gaf aan een gezonde leefstijl te hebben aangenomen. Opvallend was dat het hebben van een positieve familiegeschiedenis samenhangt met het onderhouden van een gezonde leefstijl. Dat wil zeggen dat mensen met meerdere eerstegraads familieleden met HVZ vaker een gezonde leefstijl rapporteerden dan mensen zonder of slechts één aangedaan familielid.

In **Hoofdstuk 4** wordt het mentale model voor het risico op HVZ en het preventief gedrag van mensen met een door DNA-test aangetoonde genetische gevoeligheid voor HVZ (dataset 1) vergeleken met mensen met een vergelijkbaar risico op HVZ maar zonder aangetoonde genetische aanleg (dataset 2). Bij de mensen met een aangetoonde genetische gevoeligheid was het ervaren risico hoger, werden genetische factoren vaker als belangrijke oorzaken van HVZ aangewezen, en was het vertrouwen in de effectiviteit van medicijnen groter dan in de groep zonder aangetoonde gevoeligheid. Deze verschillen werden nagenoeg volledig verklaard door verschillen in het aantal eerstegraads familieleden met HVZ. Er werden geen verschillen gevonden in leefstijl attributies, ervaren effectiviteit van leefstijl en in preventief gedrag.

In **Hoofdstuk 5** wordt een experimentele studie beschreven waarin de hypothese getest werd dat een fatalistische reactie op risico informatie (het idee dat er weinig gedaan kan worden om het risico te veranderen) afhangt van het soort risico informatie en van iemands zelfconcept (‘Self-Malleability’). Verondersteld werd dat informatie op basis van een DNA-test meer fatalisme opwekt dan informatie gebaseerd op familiegeschiedenis of niet-genetische informatie. Daarbij werd verondersteld dat mensen die denken zichzelf te kunnen veranderen minder fatalistisch op de informatie reageren dan mensen die zichzelf minder daartoe in staat achten. De hypotheses werden getest met behulp van drie verschillende scenario’s (vignetten) waarbij deelnemers zich moesten
voorstellen dat ze geïnformeerd werden over een verhoogd risico op HVZ. In Scenario 1 was het risico gebaseerd op een DNA-testuitslag, familiegeschiedenis en verhoogd cholesterol, in Scenario 2 was het risico alleen gebaseerd op familiegeschiedenis en verhoogd cholesterol, en in Scenario 3 was het alleen gebaseerd op verhoogd cholesterol. Zoals verwacht leidde Scenario 1 (DNA informatie) tot de minste ervaren controle over het HVZ risico. De mensen die zichzelf als meer veranderbaar beschouwen, ervoeren meer controle in alle drie de scenario’s.

**Hoofdstuk 6** beschrijft de resultaten van een studie naar de invloed van familiegeschiedenis en ‘zelf veranderbaarheid’ (‘Self-Malleability’) op ideeën over controle van het diabetes risico en op preventief gedrag bij personen met een verhoogd risico op diabetes (dataset 3). Familiegeschiedenis was positief geassocieerd met het zelfgerapporteerde preventieve gedrag (gezond voedingspatroon en voldoende beweging), maar was niet geassocieerd met de ervaren controle van het diabetes risico of de ervaren effectiviteit van het preventieve gedrag. Meer ‘zelf veranderbaarheid’ was wel geassocieerd met een grotere ervaren controle.

In **Hoofdstuk 7** wordt het gebruik van familiegeschiedenis als een instrument voor de preventie van veel voorkomende chronische ziekten (diabetes, HVZ en verschillende vormen van kanker) besproken. Uit eerdere studies blijkt voldoende bewijs dat gedetailleerde familiegeschiedenis informatie, in samenhang met andere persoonlijke risicofactoren, zoals overgewicht, gebruikt kan worden als een eenvoudige, gemakkelijk toe te passen en kosteneffectief instrument om het persoonlijke risico op ziekte(n) te bepalen. Daarnaast zou familiegeschiedenis informatie gebruikt kunnen worden om de gezondheidsinformatie te individualiseren. Dat kan effectiever zijn om mensen te motiveren een gezonde leefstijl aan te nemen dan algemene gezondheidsinformatie. Persoonlijke informatie zou zo moeten worden geformuleerd dat het goed aansluit bij het reeds bestaande mentale model van het risico van het betreffende individu. Op deze manier kunnen onjuiste overtuigingen gecorrigeerd worden, kennisleemtes worden aangevuld en het vertrouwen van mensen om hun gedrag te kunnen veranderen worden versterkt. Het bewijs voor de effectiviteit van het gebruik van persoonlijke informatie en uitleg over familiegeschiedenis voor ziektepreventie, met name voor het stimuleren van gedragsverandering, is echter nog beperkt.

In het laatste hoofdstuk van dit proefschrift worden de belangrijkste resultaten besproken en gepresenteerd in een theoretisch kader. Een belangrijke conclusie die uit dit onderzoek getrokken kan worden is dat genetische risico-informatie niet lijkt te leiden tot fatalistische reacties of tot een vermindering van het vertrouwen in de effectiviteit van preventief gedrag. De resultaten bevestigen ook dat het hebben van een positieve familiegeschiedenis van diabetes en/of HVZ het ervaren risico
op deze ziekten verhoogt en bovendien de motivatie voor een gezonde leefstijl vergroot. Er zijn geen aanwijzingen gevonden dat (bevestigende) DNA informatie, als aanvulling op een positieve familiegeschiedenis, effect heeft op ideeën over het risico of op gedrag. Naast familiegeschiedenis hebben ideeën over oorzaken van de ziekte invloed op het ervaren risico. Het zelfconcept, zoals gemeten met ‘zelf veranderbaarheid’, lijkt een beperkte rol te spelen in het verklaren van het gevoel van controle van het ziekte risico. Er zijn meer studies nodig om de impact van gezondheidsinformatie beter te begrijpen. Een deel van de studie bevindingen kunnen echter reeds gebruikt worden bij de ontwikkeling van persoonlijke risico informatie.
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