To know or not to know?

Attitudes towards receiving genetic information among patients and the general public.

Katharina Wolff

Dissertation for the degree of Philosophiae Doctor (PhD)

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List of Papers.

Paper 1

Wolff, K., Nordin, K., Brun, W., Berglund, G., & Kvale, G. (in press). Affective and cognitive attitudes, uncertainty avoidance and intention to obtain genetic testing: An extension of the Theory of Planned Behaviour. *Psychology and Health*

Paper 2

Wolff, K., Brun, W., Kvale, G., & Nordin, K. (2007). Confidentiality versus duty to inform—An empirical study on attitudes towards the handling of genetic information. *American Journal of Medical Genetics Part A, 143A(2),* 142–148. doi:10.1002/ajmg.a.31467

Paper 3

Wolff, K., Brun, W., Kvale, G., Ehrencrona, H., Soller, M., & Nordin, K. (in press). How to handle genetic information - A comparison of attitudes among patients and the general population. *Public Health Genomics,* doi:10.1159/000313458

Abstract

Progress in the field of molecular genetics has made it possible to identify individuals with an increased risk for a variety of hereditary diseases. To ensure successful implementation of genetic testing and counselling according to patients best interests the attitudes and motives behind testing intentions are important to consider. The main aim of the current thesis was to investigate factors which might facilitate the uptake of genetic testing. Furthermore this research investigated whether potential relatives want to be informed about the existence of hereditary conditions within their family, and under which conditions they want healthcare providers to breach confidentiality in order be informed. Finally, the thesis compares the attitudes of patients with hereditary conditions within their family to the attitudes of the general public concerning these issues. It was hypothesized that interest in receiving genetic risk information would be influenced by both characteristics of the individual and by characteristics of the disease. The role of disease characteristics was studied by using scenarios with systematically varied disease features, namely fatality, treatability and penetrance. Individual factors were investigated by using an extended version of Ajzen's (1985; 1991) Theory of Planned Behaviour, as well as several individual difference measures such as uncertainty avoidance, worry, coping, self-efficacy, consideration for future consequences, knowledge about genetics, and familiarity with genetic testing. Results showed interest in learning about ones genetic risk to be relatively high, as was the acceptance of confidentiality breaches. Among the predictors the most important disease characteristic was the treatability of the disease, and the most important individual predictor was uncertainty avoidance. Patients were found to be more positive towards receiving genetic risk information and towards breaches of confidentiality then the general population.

Introduction

Background

Genetic tests include a variety of laboratory techniques to determine whether a person has a hereditary condition or disease, or is likely to develop one. Tests can be used to confirm a diagnosis (diagnostic testing), to establish whether a couple is at risk of having a child with a genetic disorder (carrier testing), or to detect changes in a fetus's genes or chromosomes before birth (prenatal testing). The current thesis focuses on predictive testing, also called presymptomatic or susceptibility testing, i.e. genetic testing used to determine whether a symptom free individual has an increased risk of developing a genetic disorder later on in life. Currently more than a 1000 tests for different genetic conditions are available, many of which are very rare. For the time being tests are mainly offered to individuals with a family history of certain diseases, in later years, however, genetic testing services are becoming available to everybody over the internet. To ensure successful implementation of genetic testing at the population level and to promote counseling according to patients best interests the attitudes and motives behind testing intentions are important to consider. Some limited research attention has been directed toward estimating interest in genetic testing and actual test taking behaviour. These studies show a fairly high interest in predictive testing for hereditary cancers, ranging from 32% (Bunn, Bosompra, Ashikaga, Flynn, & Worden, 2002) up to 90% (Bosompra, Ashikaga, Flynn, Worden, & Solomon, 2001), and a somewhat lower interest for diseases such as Huntington's disease (Binedell & Soldan, 1997) or Alzheimer's disease (Frost, Myers, & Newman, 2001). However, less is known about the motivating factors behind these test taking intentions. More knowledge about these factors could improve counseling as well as the accuracy of estimates of upcoming and changing demands for genetic testing services.

Genetic information differs to some extend from other medical information in that it is personal, sensitive, familial, and potentially discriminatory. Genetic testing reveals risk information not only about the individual but also about the individual's family members. Patients who test positive for a disease causing mutation are strongly encouraged to inform their family, so that testing can be offered to all at-risk relatives, and the disease can either be prevented or the prognosis improved by early detection. The existence of this sensitive personal- yet at the same time familial- information raises new ethical and legal questions (Falk, Dugan, O'Riordan, Matthews, & Robin, 2003; Wertz & Fletcher 1989; 1991). One dilemma is how to protect the individual's right *not* to know this information. Another questions concerns health care providers' rights and possibly obligations to disclose this information to at-risk relatives. In the assumingly rare cases where patients fail to disclose risk information to family members, healthcare professionals face an ethical dilemma between the principle of confidentiality on one side, and the duty to warn at-risk individuals on the other. This problem is particularly pressing for physicians caring for several members of the same family (Chan-Smutko, Patel, Shannon, & Ryan, 2008).

The question of confidentiality versus duty to warn has been discussed in the literature and there is some limited empirical research looking at patients and professionals attitudes towards the issue. In the medical ethics literature a spectrum of opinions about the stringency of confidentiality exists, ranging from commentators focusing on the sensitivity of genetic information, calling for more stringent confidentiality measures, on one end of the spectrum (e.g. Denbo, 2006; McGuire et al., 2008), to others focusing on the non-individualistic cohort ownership of genetic information questioning the practice of withholding information from potentially affected family (e.g. Taub, Morin, Spillman, Sade, & Riddick, 2004; Gilbar, 2007) on the other end. Regarding empirical research, findings show that the vast majority of patients perceive it to be their duty to inform their family (d'Agincourt-Canning 2001; Wilson

et al. 2004), and most patients *do* in fact disclose their genetic status to relevant family members (Clarke et al. 2005). Research on family communication about risk for Huntington Disease shows that some patients pursue genetic testing *solely* to provide risk information to other family members (Etchegary & Fowler, 2008) whilst others decline testing out of fear that a positive test result would be harmful to family members (d'Agincourt-Canning, 2006). With regard to health care providers attitudes, there seems to be little consensus as to which circumstances call for confidentiality breaches (Wertz, Fletcher, & Mulvihill, 1990). Findings also show that about half of all clinicians have faced the dilemma of patients refusing to inform family members in their practice; and about ¼ has seriously considered informing at-risk relatives against their patient's wishes. Cases where clinicians actually do inform relatives without consent are rare, despite the fact that the majority of clinicians (63 to 69%) believe they have an obligation to ensure that at-risk relatives are being informed (Clarke et al. 2005; Dugan et al. 2003; Falk et al. 2003).

The situation to date remains challenging. Clinicians are facing the dilemma of whether or not to warn at-risk relatives, but there is little consensus or established practise on how to tackle the issue. The lack of research looking at whether or not relatives actually want to be informed is therefore striking. To my knowledge there are heretofore no empirical studies looking at whether potential relatives want to be informed about the existence of hereditary conditions within their family, and under which conditions they want healthcare providers to breach confidentiality to inform them. It is important to investigate, not only the attitudes of the general population, but also attitudes among affected groups, i.e. people with an increased risk for hereditary diseases. This is because affected groups constitute a minority within the general public. Public opinion, however, might have a decisive influence on an issue that is personally relevant first and foremost to the affected group.

Research Aims

The present research set out to study the predictors and motivating factors of intentions to undergo genetic testing. Furthermore, it investigated whether and under which conditions people want to be informed about the existence of hereditary condition within their family. And finally it compared the attitudes of individuals who have experience with genetic conditions within their family (i.e. patients with an increased risk for hereditary cancer) to attitudes of the general public regarding these questions. Interest in ones genetic risk status is likely to be predicted by both characteristics of the individual and by characteristics of the disease, and even by the interplay between the two. Therefore the research to be presented in this thesis investigates the role of disease characteristics by using hypothetical scenarios with systematically varying disease characteristics; as well as the role of individual characteristics by using an extended version of Ajzen's (1985; 1991) Theory of Planned Behaviour (TPB) and several individual difference measures.

On Attitudes

In order to be able to study the effect of different disease characteristics in combination with individual characteristics, hypothetical scenarios were employed throughout this research. This implies that the dependent variables of the current research can not be measures of actual genetic testing behaviour, but are measures of attitudes and intentions towards genetic testing and towards being informed about the existence of hereditary conditions within ones family. Attitudes are usually conceptualized to include both an affective and a cognitive evaluation of an object as well as a behavioural tendency towards the object. Most modern definitions of the attitude concept are unidimensional, including an evaluation of the attitude object which is both cognitive and affective, but excluding behaviour from the definition of attitude (e.g. Eagly & Chaiken, 1993; Petty, Wegener &

Fabrigar, 1997). Attitudes are commonly found to predict behaviour moderately well (e.g. Glasman & Albarracín, 2006).

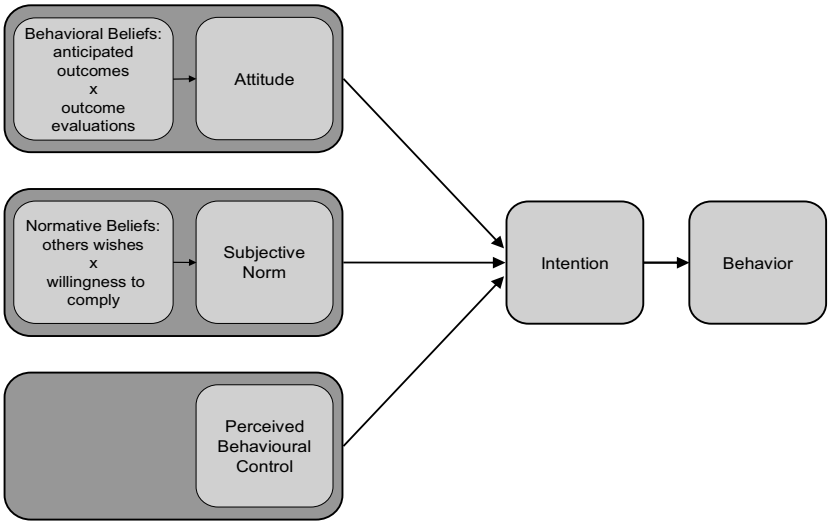
The research presented in the current thesis employed Ajzens (1985, 1991) operationalization of the attitude concept, which defines an attitude as a cognitive and affective evaluation of a given behaviour, which is separate from, but predictive of the intention to perform that behaviour. While the affective evaluation is a defining component of an attitude, according to Ajzen (1985, 1991, 2002), his model, the Theory of Planned Behaviour (TPB), has still been criticized for not assessing this component sufficiently (Manstead & Parker, 1995; Richard, van der Pligt, & de Vries, 1995). The present research therefore tried to extend the TPB (Ajzens, 1985, 1991) by incorporating a measure of the affective attitude component.

Individual Characteristics Predicting Interest in Genetic Risk Information

Potential individual predictors were assessed using an extended version of Ajzen's (1985; 1991) Theory of Planned Behaviour (TPB) as well as several individual difference measures. The TPB is an expectancy-value model designed to predict and explain human behaviour in specific contexts (Ajzen, 1985, 1991). Its utility in predicting and explaining intentions as well as actual behaviour has been demonstrated in a number of health-related behaviours including genetic testing intentions (Nordin, Bjork & Berglund, 2004). The model depicts intention as the direct antecedent of behaviour. Intentions are in turn determined by *attitudes*, *subjective norms*, and *perceived behavioural control*. Attitudes are an individual's evaluation of the behaviour as either positive or negative, subjective norms represent the individual's perceived social pressure to perform the behaviour, and perceived behavioural control refers to the individual's perception of control over performing the behaviour. The model also specifies the antecedents of attitudes and subjective norms. For any given attitude

these are a small set of specific salient behavioural beliefs, i.e., anticipated outcomes of the behaviour, weighted by an evaluation of each of these outcomes. Correspondingly the antecedents of subjective norm are a set of beliefs about how others want one to behave weighted by ones willingness to comply with these wishes (Ajzen, 1985, 1991).

The Theory of Planned Behaviour (Ajzen, 2002)



A recent meta-analysis (Cooke & French, 2008) examining the models ability to predict intentions to attend screening programs (including genetic screening) as well as actual attendance in such programs found large sized relationships between attitudes and intentions, and medium sized relationships between intentions and actual attendance behaviour. The relationships of subjective norm and perceived behavioural control with intentions were also medium sized.

Despite the success of the TPB in predicting various behaviours, a substantial proportion of variance in health related behaviours remains unexplained. One possible

shortcoming of the TPB is its lacking focus on affective processes, in that it tends to emphazise cognitive or instrumental outcomes of behavioural actions (Manstead & Parker, 1995; Richard et al., 1995). However anticipated affective outcomes may be as important as instrumental outcomes in determining attitudes and intentions. Studies that have extended the TPB by measures of anticipated affective outcomes, especially anticipated regret, have been successful in increasing the predictive power of the model across different behaviours (see e.g. Parker, Manstead, Stradling, & Reason, 1992; Richard, de Vries, & van der Pligt, 1998; Abraham & Sheeran, 2004). A meta-analysis by Cooke and Sheeran (2004) found affective-cognitive consistency to moderate attitude-behaviour consistency, and reviewing the research, Conner & Armitage (1998) concluded that there is evidence supporting the inclusion of anticipated affective outcomes into the TPB-model. A more recent meta-analysis by Rivis, Sheeran, and Armitage (2009) found that the inclusion of anticipated affect increased the variance explained in intentions by 5%.

The relative lack of affective outcomes in the TPB may possibly be caused by the method used to elicit anticipated outcomes, namely by asking for advantages and disadvantages of a given behaviour. This might sample a predominantly cognitive subset of anticipated outcomes and fail to elicit beliefs which are more difficult to articulate, like e.g. affective outcome expectations (Conner & Armitage, 1998). In the present research the TPB was therefore extended to also include anticipated affective outcomes of the behaviour, i.e. affective behavioural beliefs.

Other individual difference measures employed in this research include measures of uncertainty avoidance, worry, coping, self-efficacy, consideration for future consequences, knowledge about genetics, and familiarity with genetic testing. Uncertainty avoidance as a possible motivator for genetic testing emerged in research on test uptake for Huntington's disease. Women who considered increased certainty as an advantage of genetic testing were

found to be more positive towards obtaining a test (Decruyenaere, Evers-Kiebooms, & Van den Berghe, 1993). Similarly Braithwaite, Sutton, and Steggles (2002) found that uncertainty avoidance predicted intention to be tested for hereditary cancer. Croyle, Dutson, Tran and Sun (1995) found women high in Need for Certainty to report greater interest in genetic testing, however when provided with additional information about the remaining cancer risk for women testing negative, they were less interested in obtaining a test. The opposite pattern was found for women low in Need for Certainty. Henderson, Maguire, Gray, and Morrison (2006) also found that the desire to resolve ambiguity motivated some of their participants to pursue genetic testing. The present research hypothesised that individuals high in uncertainty avoidance would be more interested in genetic testing. However, while the research discussed above used broad personality type measures to assess the concept of uncertainty avoidance, the current research employed a scale developed in the specific context of medical testing, where uncertainty avoidance is construed as a situation specific attitudinal measure (Braithwaite, Sutton, & Steggles, 2002). This measure is more specific than a personality measure, which in accordance with Ajzen's (1988) principle of compatibility should increase its predictive power.

Increasing amounts of research have shown that disease related worry may motivate a variety of health behaviours (Cameron, 2003). Worry has for example been associated with mammogram use (Diefenbach, Miller & Daly, 1999) and testicular self-examinations (Katz, Meyers & Walls, 1995). Cameron and Reeve (2006) found worry to be associated with perceived benefits of breast cancer testing. In fact, worry moderated the relationship of perceived benefits with testing interest, i.e. when worry was high interest was high regardless of the level of perceived benefits. This suggests that disease related worry is a strong motivator to undergo genetic testing even when benefits are limited. According to the DSM-IV (1994), worry is a key component of anxiety, and is characterized by a tendency to view

ambiguous or uncertain situations as threatening (Butler & Matthews, 1987). Worrying may even be used as a form of arousal control or problem solving strategy (Miceli & Castelfranchi, 2005). Since individuals who view the reduction of uncertainty as an advantage were found to be more positive towards genetic testing it was hypothesised that people high in trait worry, which prefer to avoid uncertainty, would be more positive towards receiving genetic risk information. Accordingly, the present research aimed at assessing the role of trait worry, as opposed to earlier employed measures of disease specific - or state - worry.

Even though uncertainty reduction and increased information search are usually thought of as means of reducing anxiety; there also exists research (see e.g. Pifalo, Hollander, Henderson, DeSalvo, & Gill, 1995) showing that a minority of responders experiences increased anxiety after receiving medical information, and some people might in fact actively avoid receiving such information (Decruyenaere et al., 1993). Findings by Zuuren and Dooper (1999) show that individuals high in monitoring, i.e. seeking information and confronting threats, were more likely to engage in disease detection behaviour, while the effect of blunting, i.e. avoiding information, was unclear. Reviewing the literature on information seeking, uptake of genetic testing, and coping strategies, Case, Andrews, Johnson, and Allard (2005) point to the importance of studying information avoidance. The present study therefore investigates whether wanting to be informed about the existence of genetic conditions within ones family are negatively related to avoidant coping. The employed measure of coping style incorporates a three factor structure of coping, i.e. task-focused, emotion-focused, and avoidant coping, which possibly reflects the underlying dimensions of coping better then earlier to-dimensional taxonomies (e.g. Millller, 1987 monitoring vs. blunting; Folkman & Lazarus, 1980 problem-focused vs. emotion-focused; Endler & Parker, 1990; McWilliams, Cox, & Enns, 2003).

Perceived control is conceptualized as a determinant of behaviour by many health behaviour models (Conner & Norman, 1996; Rutter & Quine, 1994) and research has shown that people who perceive to have control over their lives are more likely to engage in health promoting behaviours (Norman, Bennett, Smith, & Myrphy, 1998). Perceived behavioural control as conceptualized by Ajzen's (1985, 1991) TPB-model is one example of such a control construct. One of the most prominent control constructs is self-efficacy, defined as an individual's belief in her capability to muster the cognitive, motivational and behavioral recourses required to produce given attainments (Bandura, 1997). According to Bandura (1997), self-efficacy (like perceived behavioural control) is a situation-specific, contextualized, and state-like belief in ones competence which is based on personal experience with the behaviour. Specific self-efficacy has been found to predict various health behaviors (Strecher, DeVellis, Becker, & Rosenstock 1986), including intentions to screen for dementia (Galvin, Fu, Nguyen, Glasheen, & Scharff, 2008) and perceived benefits from cancer genetic testing (Manne et al., 2007). Research suggests that repeated experiences of failure or success may develop into a generalized, trait-like belief in ones ability to deal with life in general, which is carried forward into new situations (Wallston, Wallston, Smith, & Dobbins, 1987; Hendy, Lyons, & Breakwell, 2006). Leganger and Kraft (2003) have shown that such general self-efficacy can mediate the relationship between higher socioeconomic status and health behaviours. Taking a genetic test is a novel situation for most individuals, and it is therefore unlikely that many have developed situation specific self-efficacy in this domain. Study two therefore aimed at examining whether general self-efficacy would predict intentions to undergo genetic testing.

The degree to which behavioural choices are influenced by an individual's time perspective or by a consideration for the potential future outcomes of a decision may differ from one person to the next. Strathman, Gleicher, Boninger, and Edwards (1994) have

defined such individual differences in the "Consideration for Future Consequences" (CFC) as "the extent to which people consider the potential distant outcomes of their behaviour and the extent to which they are influenced by these potential outcomes" (Strathman et al., 1994, p. 737). The concept is extensively employed and is primarily assessed by the CFC-scale. CFC has been found to influence and predict a wide variety of phenomena, including health-related behaviors, like decisions concerning HIV screening (Dorr, Krueckeberg, Strathman, & Wood, 1999). Orbell, Perugini, and Rakow (2004) found that individuals who considered immediate rather than distant consequences where more positive towards colorectal cancer screening when it had short term positive and long term negative consequences. The opposite pattern was found for participants high in CFC. In the current thesis it was therefore hypothesized that individuals who consider distant as opposed to immediate consequences when making decisions would be more positive towards learning about their potential genetic risk.

Findings concerning the effect of having more knowledge about genetics and being aware of the possibility of genetic testing are rather inconsistent in previous research. Studies have found knowledge to be both related (Jallinoja & Aro, 2000; Thompson, Valdimarsdottir, Jandorf, & Redd, 2003) and unrelated (Lipkus, Iden, Terrenoire, & Feaganes, 1999; Kinney et al., 2001) to interest in genetic testing. The same is true for familiarity with genetic testing (no relation: Bunn et al., 2002; significant relation: Satia, McRitchie, Kupper, & Halbert, 2006). The present research assessed both knowledge about genetics and familiarity with genetic testing in order to try and clarify the issue.

Disease Characteristics Predicting Interest in Genetic Risk Information

Instead of only investigating test interest for specific diseases, it is important to try to disentangle which disease characteristics motivate test-taking in general. This will make it possible to more accurately estimate the demand for genetic testing services for diseases

where characteristics have changed (e.g. from fatal to non-fatal, or from non-treatable to treatable) and for diseases with given characteristics where testing becomes available for the first time. In addition to examining the influence of the individual characteristics described above, the present research therefore investigates the effect of three disease characteristics, namely treatability, fatality, and penetrance (i.e. the probability of getting ill in case one is a mutation carrier) on interest in receiving genetic risk information.

The *treatability and/or preventability* of a disease are well established predictors of test taking interest (e.g. Roberts, 2000; Shaw & Bassi, 2001). The *fatality* of a disease is one of its major characteristics, and a prominent indicator of its severity and as such a potentially important predictor of test taking interest. *Penetrance* is the probability of getting ill in case one is a mutation carrier. Penetrance may be low for some diseases (e.g. 10%) and close to certain for others (up to 100%). Increased penetrance has been found to increase test interest (Frost et al., 2001). It is also possible that disease characteristics interact with each other or with individual preferences to influence test taking interest. Findings by Wang, Gonzalez, Janz, Milliron, and Merajver (2007) showed for example that individuals who perceived their own susceptibility to be high but the severity of breast cancer to be low were more likely to pursue genetic testing than all other susceptibility/severity combinations. As mentioned earlier, findings by Croyle and colleagues (1995) point to the possibility that people with a preference for uncertainty might be more interested in genetic testing for diseases with low penetrance.

The duty to warn at risk relatives about their potential genetic risk is also strongly influenced by the characteristics of the disease. It increases as the probability that relatives will be affected increases, the disease becomes more serious, and the disease is preventable or harm is reducible by early detection. Confidentiality guidelines in most countries take this into account and allow breaches of confidentiality only for diseases that meet these criteria

(ASHG, 1998; Godard, Hurlimann, Letendre, Égalité & INHERIT BRCAs, 2006). In terms of the disease characteristics varied in this research it means that test interest and acceptance for confidentiality breaches might be higher for diseases that are highly penetrant, fatal and treatable.

To sum up the present research aims at illuminating the role of individual and disease characteristics in predicting intentions to undergo genetic testing, willingness to receive genetic risk information from relatives, and acceptance of confidentiality breaches by health care personnel in order to be informed about ones genetic risk. The research also aims at comparing the attitudes of affected individuals, i.e. people with hereditary conditions in their family, to the attitudes of the general public on these issues.

Methods

In order to be able to investigate attitudes in large samples and be able to directly compare attitudes across different samples, the research presented in this thesis primarily relied on survey method. Questionnaires describing systematically varied disease scenarios were collected in different samples in Norway and Sweden, including individuals from the general population, patients and students. Individual difference measures were used to investigate the effect of individual predictors on interest in receiving genetic risk information. In order to avoid tiring research participants with long questionnaires, different individual difference measures were employed during separate data collections. This makes it of course impossible to compare samples on the assessed concepts, it allows however for the study of a greater variety of possible predictors.

Participants and Procedure

Paper 1 aimed to investigate predictors for intentions to obtain a genetic test for hereditary diseases with varying characteristics within the general population. A random sample of the Norwegian population (N = 2400) between the ages of 18 to 65 received a questionnaire and one reminder in the mail. The final response rate was 36.4%, resulting in a total of 874 participants, of which 46.2% were male, the mean age was 41.7 (*SD* = 12.8), and 41.0% had university education. Four hypothetical disease scenarios were constructed by systematically varying two disease characteristics: fatality (fatal vs. non-fatal) and penetrance (50% vs. 100% penetrance). All diseases were described as incurable. The design was cross-sectional; only one disease scenario was randomly assigned to each participant. This resulted in four versions of the questionnaire.

Paper 2 investigated under which conditions participants from the general population want to be informed about the existence of hereditary conditions within their family, and

whether they want health care providers to breach confidentiality in order to be informed. The study includes three samples. The first sample is the same Norwegian random sample as in Paper 1 (N = 2400). The second sample is a comparable Swedish sample, randomly drawn from the population between the ages of 18 to 75 (N = 1200). Participants were mailed a questionnaire and three reminders. A total of 665 completed questionnaires were returned, constituting a response rate of 55.4% of which 47.4% were male, the mean age was 44.3 (*SD* = 15.5), and 28.5% had a university education. By varying three disease characteristics (fatality, penetrance, and treatability) four hypothetical scenarios were constructed: 1. Fatal, non-treatable disease with 50% penetrance. 2. Fatal, non-treatable disease with 100% penetrance. 3. Non-fatal, treatable disease with 50% penetrance. 4. Non-fatal, treatable disease with 100% penetrance. One scenario was randomly assigned to each participant.

The third sample is a convenience sample of students from the University of Bergen, Norway (n = 607). Data were collected during lecture breaks in introductory psychology and natural science courses. Response rates were very high, about 90%, 34.3% were male and the mean age was 21.2 (*SD* = 4.6). The same three disease characteristics as in the Swedish random sample were varied, constructing all eight possible disease scenarios. One disease scenario was randomly assigned to each participant.

Paper 3 aimed at investigating attitudes towards the handling of genetic information in an affected group, i.e. people with an increased risk for hereditary cancers, and compared them to the attitudes of the general population. The study includes 2 groups: patients and individuals from the general population. The general population sample consists of the three subsamples described in Paper 2, i.e. the Norwegian random sample, the Swedish random sample and the Norwegian student sample. The patient sample consist of individuals that underwent genetic counselling for suspected hereditary cancers at the University hospitals in Lund or Uppsala (Sweden, N = 408), or the University hospital in Bergen (Norway, N = 414).

Everyone over age 18 who underwent genetic counselling at one of these institutions during 2005 was mailed a questionnaire and one (Norway) / two (Sweden)[1] reminders. Response rates were 73.5% in the Swedish sample and 52.4% in the Norwegian sample, resulting in a total of 517 participants, 15.4% were male, mean age was 48.0 (*SD* = 12.35). The same three disease characteristics as described above, i.e. fatality, penetrance, and treatability, were systematically varied, and all eight possible disease scenarios were constructed. The design was cross-sectional; only one scenario was randomly assigned to each participant.

Measures

Constructs belonging to the TPB (i.e., intentions, behavioural beliefs, subjective norm, and perceived behavioural control) assessed in Paper 1 were measured in accordance with Ajzen's (2002) suggestions.

Dependent variables.

Intention to undergo genetic testing (Paper1). Participants were asked to imagine that they had a close relative with one of the above mentioned diseases. Following a brief description of the disease participants answered two questions. First: "Would you be interested in taking a genetic test if you had to take the initiative for being tested yourself?" and second: "Would you be interested in taking a genetic test if your physician suggested it?" Answers were given on 7-point semantic differential scales anchored by *very unlikely(1)-very likely(7).* Scores of one and two were coded as not intending to test and scores of six and seven as intending to test.

[1] The different numbers of reminders which were sent to samples in Sweden and Norway throughout the studies presented here are due to the fact that Norwegian ethics committees allow for only one reminder to be sent to research participants while Swedish committees do not pose any limitations. This explains the greater response rates observed in the Swedish samples. Before sending out reminders response rates are comparable in the Swedish and Norwegian samples in all studies. The difference in number of reminders was controlled for by entering this variable into the first block of the regression analysis in all studies. No effect was found in any of the studies.

Desire to be informed (Papers 2 and 3). Participants were asked to imagine that they had a close relative who tested positive for a hereditary disease. Following a description of the disease all participants answered three questions on 7-point scales all anchored by *agree completely(1)–disagree completely(7)*: (1) "I want my relative to tell me that he/she is a carrier of the disease causing mutation." (2) "I want the physician to contact and inform me even if my relative is against it." (3) "I do not want to be informed if one of my close relatives has this hereditary disease." Items one and two were reversed in the analysis. In Paper 2 items one and three were averaged to constitute a measure of "desire to be informed by the relative". In Paper 3 only question one was used as an indicator of "desire to be informed by the relative". Question three was dropped from the analysis because many participants of the patient sample answered it inconsistently, i.e. not noticing that its meaning is opposite to that of question one. Item two was used to indicate "desire to be informed against the relatives' wishes" in both papers. Scores of two or less were coded as not wanting to be informed and scores of six or more were coded as wanting to be informed with or without the relative's consent respectively.

Predictor variables

Attitudes (Paper1). Belief based measures were used to assess both attitude and subjective norm. Anticipated outcomes of genetic testing were mapped by semi structured interviews in a pilot study. Content analysis of the interviews yielded 14 different anticipated outcomes. Five additional outcomes which were to reflect affective outcomes were added to the final questionnaire. Participants judged the probability and the desirability of each anticipated outcome using 7-point semantic differential scales. Corresponding items on both scales were multiplied to constitute belief based measures of attitude. Exploratory factor analysis (varimax rotation) of the multiplied items yielded four meaningful factors, labelled *Negative consequences* ($\alpha = .79$), *Positive consequences* ($\alpha = .66$), *Information* ($\alpha = .68$), and *Future*

effects (α = .56). Items loading on each respective factor were summed to constitute believe based measures of attitude.

Subjective norm (Paper 1). Responses to two questions were given on 7 -point semantic differential scales: (1) "Do you believe that the following people would want you to undergo a genetic test…?" (2) "…will you take these people's opinion into consideration?" Each question was followed by a list of eight persons (husband/wife, children, parents, siblings, grandparents, friends, physician, and the media). All scales were anchored by *no, absolutely not(1)-yes, absolutely(7)*. Corresponding scores on question one and two were multiplied. The resulting eight items displayed high internal consistency (α = .90), and were summed to construct an index of subjective norm.

Perceived behavioural control (Paper1). Four questions were answered using 7-point semantic differential scales: (1) "For me to have a genetic test would be… *very difficult(1)-very easy(7)*", (2) "If I wanted to I would manage to have the test taken.", anchored by *disagree completely(1)-completely agree(7)* (3) "It is up to me whether or not I will have a test like that" *disagree completely(1)-completely agree(7)*, and (4) "How much control do you believe you have over undergoing a test like that?" anchored by *no control(1)-complete control(7)*. Responses to these questions were summed in order to construct an index of perceived behavioural control (α = .54).

Uncertainty avoidance (Papers 1, 2, and 3). The Attitude toward Uncertainty scale (Braithwaite et al., 2002), was used to assess the tendency to avoid uncertainty in the specific context of medical testing. The scale consists of 8 items and responses are given on a 5-point scale ranging from *strongly disagree(1)* to *strongly agree(5)*. The composite measure was computed as the mean of all items (after reversing two items). High scores indicate a preference to avoid uncertainty. Paper 1: α = .88; *M* = 3.28, *SD* = .98; Paper 2: α = .89, *M* = 3.33, *SD* = 1.04; Paper 3: α = 90, *M* = 2.51, *SD* = 1.05

Knowledge about genetics (Paper 2) was measured by seven self-constructed statements about genetics and heritability. Participants indicated whether they believed the statements to be true or false, or whether they were uncertain. Correct (+1), incorrect (-1), and "don't know" (0) responses were summed to constitute a measure of knowledge about genetics (M = 3.64, SD = 2.13). Respondents gave on average 4.23 (SD = 1.83) correct and 0.59 (SD = .78) incorrect answers.

Familiarity with genetic testing[2] *(Papers 2 and 3)* was measured on a single item by having participants indicate how much they had heard about genetic testing for hereditary diseases. The 5-point scale was anchored by *I have never heard of it(1)- I know a bit about it(3)-I am well informed about it(5).* Paper 2: M = 2.55; SD = .82; Paper 3: M = 2.73, SD = 1.02.

Self-efficacy (Paper 2) was assessed using the Generalized Self-efficacy scale (Schwarzer & Jerusalem, 1995), which consists of 10 items measuring the general belief that one can perform a novel or difficult tasks, or cope with adversity in various domains. Responses are given on 4-point scales and summed to constitute a composite measure (α = .88; M = 3.02, SD = .43). High scores indicate high self-efficacy. Self-efficacy was assessed in the Swedish general population sample only.

Worry (Paper 2) was assessed using the Penn State Worry Questionnaire (Meyer, Miller, Metzger, & Borkovec, 1990), which is a 16-item self-report measure of trait worry. Responses were given on 5-point scales and summed to constitute a composite score (α = 92; M = 2.42, SD = .78). High scores indicate greater worry. Worry was measured only in the Swedish general population sample.

Coping Style (Paper 3). Coping style was measured using the 30-item Coping Style Questionnaire, CSQ-30 (Joseph, Williams, & Yule, 1992) which assesses subjects' style of coping with a specific event on three dimensions: task focused-, emotion focused-, and

[2] This construct is labeled "Knowledge about genetic testing" in the published version of paper 2. One of the reviewers of paper 3 suggested however that it more correctly should be labeled "Familiarity with genetic testing". For consistency this label is used throughout the thesis.

avoidant coping. Responses are given on 4-point scales and three sum scores are computed to constitute measures of task focused- (α = .66; *M* = 26.86, *SD* = 3.65), emotion focused- (α = .60; *M* = 24.24, *SD* = 3.72), and avoidant (α = .58; *M* = 19.05, *SD* = 3.46) coping respectively. High scores indicate the presence of the given coping style. Coping style was assessed in the patient sample only.

Consideration for Future Consequences, CFC (Paper 3). This was assed with the 12-item CFC-questionnaire (Strathman et al., 1994). Responses are given on 5-point scales and summed to constitute a composite measure (α = .75; *M* = 39.08, *SD* = 3.67). High scores indicate a tendency to consider distant rather than immediate consequences of behavioural decisions. CFC was only measured in the patient sample.

Statistics

Two-way ANOVA was used to compare mean scores of the intention to test take a genetic test for the four different disease scenarios of Paper 1. One-way ANOVA (Bonferroni) and independent sample t-tests were used to compare mean scores of the desire to be informed for the different diseases in Paper 2. In Paper 3 one-way ANOVA (Bonferroni) and Mann-Whitney U tests were used to compare the desire to be informed for the different scenarios. Wherever multiple comparisons where made, a significance level of $p < .001$ was applied. Regression analysis was used to check for a possible interaction effect between uncertainty avoidance and penetrance.

To analyze the influence of demographic and other individual difference variables, block wise multiple regression analyses were performed. Regression analysis was used to check for a possible interaction effect between uncertainty avoidance and penetrance.

Results

The intention to obtain a genetic test was relatively high in the Norwegian general population, and across all samples the desire to be informed about the existence of a hereditary disease within one's family was also quite high. So was the acceptance of confidentiality breaches. Among the predictors the most important disease characteristic was the treatability of the disease, and the most important individual predictor was uncertainty avoidance. Patients were found to be even more positive towards being informed by a relative and towards breaches of confidentiality then the general population.

Paper 1

The study aimed at investigating predictors for the intention to obtain a genetic test for hereditary diseases with varying characteristics within the general population. Intentions to be tested were relatively high across all disease scenarios, varying between 40 and 63%. Intentions were greater for highly penetrant diseases, and when testing was suggested by the physician. Fatality did not influence test taking intentions. The most important individual predictor was uncertainty avoidance. The extended TPB model predicted intentions to undergo genetic testing moderately well, explaining about 11% of the variance (10% when the test is suggested by the physician). The attitude factors labelled *Negative consequences* and *information* were significant predictors. So was subjective norm. The interaction term for penetrance and uncertainty avoidance was insignificant.

Paper 2

This paper investigated whether individuals from the general population wanted to be informed about the existence of hereditary diseases within their family, and under which conditions they wanted health care providers to breach confidentiality in order to be informed.

The Norwegian general population sample, the Swedish general population sample, and the student sample were analyzed separately. The desire to be informed was quite high in all samples and for all diseases. Between 52 and 83% of participants stated that they wanted to be informed by their relative, depending on sample and disease characteristics. Only between 0 and 13% stated that they did not want to be informed. The proportions of participants wanting to be informed against their relatives' wishes were significantly lower, but still quite high, ranging from 18 to 54%. Between 11 and 36% of participants opposed breaches of confidentiality. The desire to be informed both with and without the relatives consent was greater for treatable compared to non-treatable conditions, and for fatal compared to non-fatal diseases in cases where the relative did not consent. Penetrance did not influence the desire to be informed. The disease that fulfils all ASHG (1998) criteria as to when confidentiality breaches might be permissible displayed the greatest support for confidentiality breaches, i.e. the disease that is highly penetrant, fatal and treatable. Individual characteristics which predicted the desire to be informed by the relative were first and foremost uncertainty avoidance, but also female gender, younger age, and having or planning for children. Wanting to be informed against the relatives' wishes was predicted by uncertainty avoidance, Swedish nationality, higher age, by less knowledge about genetics, and by less self-reported familiarity with genetic testing. Self-efficacy and worry did not predict the desire to be informed.

Paper 3

The purpose of this study was to investigate attitudes towards the handling of genetic information in an affected group, i.e. people with an increased risk for hereditary cancers, and compare those to attitudes of the general public. In this study the Norwegian general population sample, the Swedish general population sample, and the student sample were combined to constitute the general population sample. The desire to be informed by a relative

was quite high, ranging between 65 and 82% in the general population, and even higher between 83 and 94% in the patient sample depending on the disease scenario. The proportions of participants not wanting to be informed by their relative were relatively small, between 3 and 10% in the general population and 0 and 8% in the patient sample. Main effects were found for sample and treatability, i.e. patients were more interested in being informed than the general population, and the desire to be informed was greater for treatable than non-treatable diseases. The interaction term was also significant showing that the difference in desire to be informed found between the general population and patients was greater for non-treatable compared to treatable diseases.

The desire to be informed without the relatives consent was significantly lower, ranging between 25 and 46% in the general population and between 58 and 75% in the patient sample. Proportions of participants opposing confidentiality breaches ranged between 22 and 36% in the general population and between 7 and 27% in the patient sample. There was a main effect for sample, with patients being more positive towards confidentiality breaches, and a significant interaction effect for sample and treatability, indicating that the difference found between patients and general population is greater for non-treatable diseases.

While the general populations desire to be informed both with and without the relatives consent was greater for treatable compared to non-treatable diseases, the patient samples desire to be informed was completely unaffected by the characteristics of the disease. Individual predictors for the desire to be informed by the relative were uncertainty avoidance, female gender, and having or planning for children in the general population. In the patient sample the only predictor was uncertainty avoidance. The desire to be informed against the relatives' wishes was predicted by uncertainty avoidance, Swedish nationality, higher age, and by less self-reported familiarity with genetic testing in the general population; and by uncertainty avoidance and Swedish nationality in the patient sample.

Discussion

Approximately half of the Norwegian general population sample wanted to obtain a genetic test even for an incurable disease. A clear majority of both the patient sample and the overall general population sample wanted to be informed by a relative about the existence of a hereditary condition within their family, and patients preferred so to an even higher degree than the general population. In fact, there is a ceiling effect in the patient sample, illustrated by mean scores well above 6 (maximum score being 7) for all disease scenarios. Willingness to be informed against the relatives' wishes was significantly lower, but still quite high, with up to half of the participants of the overall general population sample, and up to ¾ of the patients, supporting breaches of confidentiality.

Findings clearly show that interest in receiving genetic risk information is quite high in all samples and for all disease scenarios. These findings are in line with other research showing high interest in genetic testing (e.g. Bosompra et al., 2001; Satia et al., 2006). The fact that patients report a greater desire to be informed about disease causing mutations than the general population is hardly surprising. After all, this sample was selected on the basis of having received genetic counselling in the past, i.e. they had already demonstrated their desire to learn about hereditary conditions in their family. However, *the extent* of positivity towards receiving genetic risk information expressed among patients might be considered somewhat surprising. It may be illustrative of the experience responders had with receiving such genetic risk information in the past: The vast majority does not show signs of regretting having learned about their genetic risk, and states that they would choose to do so again in the future. Together with the possible health advantages of knowing ones carrier status, these findings might be interpreted as support for the common practice of encouraging patients to inform at risk family members.

It is also interesting to note that patients, much more clearly than the general population, support confidentiality breaches. This is true despite the fact that many of them have been in the position of the patient who has to disclose information to family members and whose confidentiality would be breached, in the described situation. Possibly, this finding may be interpreted as an illustration of the fact that most patients understand the importance of, and perceive it to be their duty to inform family members about, their carrier status (d'Agincourt-Canning, 2001; Wilson et al., 2004).

The proportions of participants explicitly stating that they do *not* want to receive genetic risk information are small. In the Norwegian general population sample no more then ¼ of participants did not want to obtain a genetic test, at least not for an incurable disease. The proportions of participants that did not want to be informed about their genetic risk by a relative were even smaller, maximally 13% in the overall general population sample, and maximally 8% among patients depending on the characteristics of the disease. Proportions of participants opposing confidentiality breaches reached 36% in the overall general population sample, and 27% among patients.

Again, together with the advantages of knowing about an increased health risk, findings support the practice of encouraging patients to inform at risk family. However, although the numbers of participants that do *not* want to know their genetic risk are small, they are not insignificant. These findings are in line with other research showing that there are individuals who do not want to learn about an increased genetic risk. A review by Gaff et al., (2007) found that some patients had difficulties communicating with relatives, due to adverse reactions by some family members. Kenen, Ardern-Jones and Eeles (2004) also found that some individuals actively tried to avoid receiving genetic risk information from their relatives, e.g. by hanging up the phone or refusing to answer questions. If one is to take the right *not* to know seriously, it is important to try and find ways of not spreading information to

individuals who do not welcome it. At the very least it might be time to encourage individuals to discuss the issue with their family and inform others about whether and under which condition they want or do not want to be informed about hereditary diseases within their family.

Disease Characteristics Predicting Interest in Genetic Risk Information

Different disease characteristics had some influence on the participants' willingness to learn about ones genetic risk status. In the Norwegian general population sample intentions to obtain a genetic test were greater for diseases with high penetrance compared to diseases with low penetrance. This difference in test taking intentions was however no longer significant when the test was suggested by a physician. Even though survey research has demonstrated this difference (e.g. Frost et al., 2001), the present findings may suggest that it might not generalize to settings where patients rely on their physician's opinions to guide their decision making concerning genetic testing. Whether or not the disease was described as fatal did not influence intentions to be tested. It is important to note however that all diseases in the Norwegian general population sample were described as incurable, and that fatality might influence test taking intentions when the disease is treatable or preventable. (Note that since all diseases were described as incurable in this sample, there are no data on the possible effect of treatability.)

When it comes to being informed about the existence of a hereditary condition within the family, there was an effect of treatability, with the general population being more interested in being informed about treatable than non treatable diseases both with and without their relatives consent. This is in line with other research showing greater interest in genetic testing for treatable diseases (e.g. Roberts, 2000; Shaw & Bassi, 2001). In the Norwegian student sample there was also an effect of fatality, with students showing greater desire to be

informed against the relatives' whishes for fatal diseases. Patients' desire to be informed was very high and completely unaffected by characteristics of the disease. This can possibly be explained by patients being influenced by the disease they have experience with, i.e. hereditary cancer (which most of the time is treatable), rather than by the hypothetical scenario described in the survey. The ceiling effect of the dependent variable among patients is another possible explanation. Treatability also diminished the difference found between the patients and the general populations desire to be informed with and without the relatives consent. This means that there is more agreement between the samples on the importance of being informed when diseases can be treated then when they can not. This is in line with guidelines described above in countries that allow for confidentiality breaches under certain circumstances (ASHG, 1998; Godard et al., 2006). Findings are also in line with an important point made by Evans and Burke (2008), that the more useful genetic information becomes the more it challenges the concept of genetic exceptionalism, and furthermore that the *special* protection of genetic information is important only for diseases that are not treatable.

Individual Characteristics Predicting Interest in Genetic Risk Information

Uncertainty avoidance was by far the most important individual predictor of intentions to obtain a genetic test and of the desire to be informed with and without the relatives consent. In all samples this variable had by far the strongest correlation of all predictor variables with the dependent variables. These findings are in line with previous findings by Braithwaite et al. (2002) who found only one variable to predict test taking interest better than uncertainty avoidance, namely a direct measure of attitude, which was not employed in the present research. Findings clearly demonstrate that the desire to reduce uncertainty may be an important motivator for individuals wanting to learn their genetic risk status. In order to facilitate patients' decision making and secure informed choice concerning genetic testing it is

therefore important to inform them about the remaining uncertainties, e.g. tests which are less then 100% accurate, incomplete penetrance or residual risks for individuals testing negative. In light of the problems lay people in general have regarding understanding and interpreting probabilities and risk information, this is a challenging aspect of providing health information and genetic counselling.

It was expected that there would be an interaction effect between uncertainty avoidance and penetrance, i.e. participants high in uncertainty avoidance preferred to receive genetic risk information when the disease was high in penetrance, and participants low in uncertainty avoidance preferred this information when the disease was low in penetrance. This would have replicated findings by Croyle et al. (1995) showing that women who disliked uncertainty decreased their interest in genetic testing after being informed about its remaining uncertainties, while women preferring uncertainty increased their interest. No such interaction effect was found in any of the samples. Possibly participants did not understand the impact of penetrance on the degree of certainty of a genetic test result. This would explain why penetrance was not a more important predictor, even though uncertainty avoidance in itself seems to be very important to the participants.

The intention to undergo genetic testing was assessed in the Norwegian general population sample, and intention was tried to be explained by an extended version of Ajzen's (1985, 1991) Theory of panned behaviour. This extended version included measures of both cognitive and affective behavioural beliefs (i.e. outcome expectations), which fell into four factors labelled *Negative consequences, Positive consequences, Information,* and *Future effects*, as well as measures of subjective norm and perceived behavioural control. The employed TPB-model predicted intentions to undergo genetic testing only moderately well. The belief based attitudinal measures of the TPB contribute somewhat to the variance

explained in intentions. The factor *Negative consequences* is a significant predictor of test taking intentions when the participant has to initiate testing. It also predicts intentions when the physician suggests testing, but only until uncertainty avoidance is entered into the final model. This implies that participants who perceive the consequences of genetic testing to be less negative have stronger intentions to obtain a test. Furthermore *Information* is a significant predictor when the physician suggests testing, until uncertainty avoidance is entered into the model. In other words, participants who expect to receive more information, and who value this information positively, are more likely to intend to obtain a genetic test. It is worth noticing that *Negative consequences* is the only attitudinal factor which is comprised of mainly affective outcome expectations. Since the factor is a predictor of intention, findings point to the possible importance of including affective outcome expectations into the attitudinal measures of the TPB. This is in line with earlier quoted findings (Parker et al., 1992; Richard et. al, 1998; Abraham & Sheeran, 2004) showing that the inclusion of affective outcomes can increase the models predictive power. Findings are also in line with Lawton, Conner and McEachan (2009) who, using direct measures of attitude, found that affective attitudes where a stronger predictor than cognitive attitudes for 9 out of 14 health related behaviours.

The employed measure of uncertainty avoidance construes the concept as an attitudinal measure. Since it is such a strong predictor of intention one wonders why it was not mentioned in the pilot interviews conducted to map salient belief based attitudes for the TPB-questionnaire. Possibly the reduction of uncertainty, together with affective outcomes belongs to a subset of behavioural beliefs which are not readily accessed by the commonly employed method of asking for advantages and disadvantages of a given behaviour (Conner & Armitage, 1998). Since these non-cognitive beliefs are potentially important predictors of

intentions and behaviour, future research should address the question of how to elicit such beliefs among informants.

The TPB component subjective norm also contributes somewhat to the amount of variance explained in both measures of intention; however this effect disappears when uncertainty avoidance is entered into the model. It is somewhat surprising that subjective norm does not have a stronger influence on interest in test taking in our sample, especially since the results of a genetic test have a direct impact on family members. However the possibility of having genetic tests is rather new and participants might not be aware of how significant others feel about them having a genetic test. This would mean that there still does not exist any social pressure or subjective norm in this domain, and while filling in the questionnaire, participants simply assume that friends and family feel the same way they do. This would explain both the very high internal consistency of the measure ($\alpha = .90$) and why it does not influence test taking interest in this study.

The TPBs perceived behavioural control component did not predict test taking intentions. According to Ajzen (1991) perceived behavioural control is a construct which is useful for predicting intentions especially in situations where participants feel that the behaviour is difficult to control. Examples of these kinds of behaviours are quitting to smoke, exercising regularly or refraining from having unsafe sex. These are behaviours that need to be performed continuously in order to be beneficial, whilst having a genetic test is a one-time behaviour. Hence obtaining a genetic test might be considered a behaviour that is easy to control, which might be the reason that perceived behavioural control did not predict testing intentions. Therefore increasing the availability of genetic testing services might not increase test uptake.

The desire to be informed by a relative about the existence of a hereditary condition within the family was assessed both in the general population in Norway and Sweden as well as among patients from both countries. As discussed earlier, the main predictor of the desire to be informed was uncertainty avoidance in all samples. In the general population other predictors were female gender, younger age, and having or planning for children. These effects were small however. In the patient sample uncertainty avoidance was the only significant predictor, explaining only minimal amounts of variance. This is probably due to the ceiling effect of the dependent variable among patients.

When it comes to being informed against the relatives' wishes, again the main predictor was uncertainty avoidance. In the general population additional predictors were Swedish nationality, higher age, less knowledge about genetics, and less self-reported familiarity with genetic testing. These effects were also small. Among patients the only predictor besides uncertainty avoidance was Swedish nationality.

Results indicate that increasing people's knowledge about genetics and their familiarity with genetic testing might reduce their acceptance of confidentiality breaches. Together with the finding that uncertainty avoidance motivates genetic testing, the fact that knowledge about testing decreases interest indicates that participants may overestimate the degree of certainty that genetic testing can provide. Again this illustrates the importance of informing the general public about the remaining uncertainties of genetic testing. Possibly this might also reduce their acceptance of confidentiality breaches. However looking at the degree of positivity towards confidentiality breaches among the participants most familiar with genetic testing, i.e. patients, this is far from certain.

The fact that Swedish participants, both general population and patients, were more positive towards confidentiality breaches than participants in Norway can possibly be explained by Sweden being the most liberal country in the EU and one of the most liberal

countries in the world as far as access to documents is concerned (Österdahl, 1998). Sweden has the world's oldest freedom of information law, and views on public access of information as a human right (Davis, 1999).

Other individual predictors that were assessed but did not predict the desire to be informed either with or without the relatives consent were Self-efficacy and worry in the general population of Sweden, and Coping style and Consideration for Future Consequences (CFC) among patients in Sweden and Norway. Because Coping style and CFC were assessed in the patient sample only, and because this sample displayed a ceiling effect in the dependent variable it is difficult to conclude that these variables are *not* related to interest in receiving genetic risk information. Future research, using samples with greater diversity of responses, will have to illuminate this question.

The fact that Self-efficacy did not predict desire to be informed by a relative is in line with findings described in Paper 1, showing that perceived behavioural control did not predict intentions to undergo genetic testing. As discussed earlier, possibly having a genetic test, or receiving genetic risk information is not viewed as difficult to control or to perform by participants. This might of course be due to the fact that the situation is hypothetical for most participants from the general population.

The finding that trait worry was unrelated to desire to be informed might indicate that the desire to reduce medical uncertainty which seems to motivate genetic testing is not related to a general tendency to worry.

Research Limitations

Ecological validity.

One limitation of this research is the rather low response rates especially in the general population samples. Inspection of the sample demographics shows that higher education and

female gender are overrepresented amongst the participants. Furthermore it is unfortunate that the samples also differ from each other in demographic characteristics and in survey administration, e.g. number of reminders received. By comparing participants who responded immediately to participants who responded after receiving a reminder on the depended variables (i.e. intention to undergo a genetic test and desire to be informed) the possible effect of the number of reminders was examined. Also demographic variables, as well as the number of reminders received before answering, were entered into the first block of the regression analyses in order to control for their effect on the dependent variables. Findings revealed that these variables wield minimal to no influence. Group differences are therefore *un*likely to be caused by differences in sample demographics or survey administration. This implies that the findings probably can be generalized even though the samples' demographic characteristics might not be entirely representative of their respective populations. How non-responders might have responded and how they differ from participants is a recurring problem of all survey research, including the current work, regardless the size of the response rates. However while the proportions of respondents stating that they want to receive genetic risk information may be affected by the response rates, there is less reason to assume that the pattern showing different levels of interest for different diseases would be very much affected, that individual predictors would be different, or that there would be a higher degree of consensus regarding the issue of confidentiality.

The use of individuals who have received genetic counselling to represent the at-risk population is another problem regarding the ecological validity of the current research. This group has been selected on the basis of their previous interest in genetic information and may thus be a special sub-sample not entirely representative of the at-risk population in general. Ethical and legal considerations make it difficult, if not impossible, to recruit a representative at-risk sample in a different manner, and for the time being one will have to live with this

shortcoming in order to be able to make the comparison between general population and at-risk individuals. It is also worth noticing that even though the entire patient sample had received genetic counselling, not everybody chose to go ahead with genetic testing. The overwhelming positivity amongst these patients is not self-evident and hence interesting to note, although a more representative at-risk sample or the entire at-risk population might display somewhat less positive attitudes.

Hypothetical disease scenarios have been employed throughout the research presented in this thesis. To what extent the respondents perceived the scenario descriptions as realistic, and hence responded in accordance with their "real" attitudes and reactions, is therefore uncertain. The use of hypothetical scenarios also implies that attitudes and intentions were measured instead of actual genetic testing behaviour. This does of course imply some limitations to the generalizeability of the findings as research shows that the relationship of attitudes and intentions with behaviour are often only moderate (Glasman & Albarracín, 2006; Cooke & French, 2008). Several factors have been shown to influence the attitude-behaviour relation. For example are attitudes a stronger predictor of behaviour when they are based on consistent or one-sided information and on direct experience with the behaviour (see e.g. Glasman & Albarracín, 2006). In study one participants where confronted with both positive and negative outcomes of genetic testing, and in all three studies presented here only the patient sample had direct experience with receiving genetic risk information. Therefore one might be somewhat pessimistic about how representative the assessed attitudes and intentions are of actual test taking behaviour. The often found discrepancy between high self-reported intentions and much lower actual test uptake (Binedell & Soldan, 1997) may certainly apply to this research as well. One can therefore speculate that actual test taking behaviour will be lower in a real life setting, and future research will have to examine the relations between predictors of test taking intentions with actual testing behaviour. However, as discussed

above, while the proportions of supporters and opposers of receiving genetic risk information might be affected by the limitations of this research, predictors are less likely to be affected.

It should also be noted that the use of hypothetical scenarios has advantages. Only the use of diseases with systematically varied features makes it possible to study the effect of disease characteristics and their possible interaction with individual differences. Furthermore, using non-specific diseases ensures that the participants' decisions are influenced by relevant disease characteristics like treatability, rather than by less relevant characteristics like the familiarity of any specific disease. One might even argue that the result's generalizeability increases by the use of hypothetical diseases. This is because it becomes possible to infer the relative degree of test interest for different conditions depending on how these conditions rank on the three characteristics that have been manipulated, i.e. fatality, penetrance and treatability.

Furthermore, it is in fact impossible to ask someone the question of whether they want to be informed about the existence of a hereditary disease within their family for any specific disease without already making the essence of that information known to the person (Wertz & Fletcher, 1991). On a general or hypothetical level, however, it is possible to ask if, and under which conditions, someone would be interested in knowing whether they are at risk. If one is to take patients' right of choice seriously, this is, in fact, the only way to grant this right.

Measurement issues.

The employed measures of individual differences vary in their degree of specificity. While some of them constitute broad personality measures, like e.g. general self-efficacy, others are much more specific attitudinal measures, like the measure of uncertainty avoidance. According to Ajzen's (2002) principle of compatibility, in order to find strong correlations between variables, all constructs need to be defined in terms of the same elements regarding specificity, context, and time. While having a genetic test is a rather specific behavior,

generalized Self-efficacy and trait worry are very general measures. This is another possible explanation for why the results did not show a relation between these variables and the desire to be informed about genetic conditions within ones family, while more specific measures like disease related worry (Cameron, 2003) and specific Self-efficacy (Strecher et al., 1986) have been found to be associated with health related behaviours. The compatibility principle may also partly explain why the employed measure of uncertainty avoidance was such a strong predictor, compared to all other variables. The relation between uncertainty avoidance (as measured by the Attitude towards Uncertainty-scale; Braithwaite et al., 2002) and test taking intention as well as the desire to be informed in the current research is interesting to observe and in line with findings by others (e.g. Decruyenaere et al., 1993; Croyle et al., 1995). However, it will be important in the future to study how this measure relates to more general trait measures of uncertainty avoidance and interest in genetic risk information.

The ceiling effect observed in the patients desire to be informed is problematic and a clear limitation of this research. This methodological problem makes it difficult, if not impossible, to make any conclusive inferences about the possible predictive value of the measures employed in this study, i.e. Consideration for future consequences and Coping style. Future research may work to construct a more nuanced and sensitive dependent variable to be able to further examine the role of these concepts in predicting interest in receiving genetic risk information.

Another limitation of the thesis is that samples to a certain extend have been "reused" in different papers. While this is cost efficient, it results in smaller sample size and, may be more importantly, one also misses the opportunity of replicating findings which could have increased confidence in the conclusions. It is however interesting to note that participants from samples who differ in sample selection method and demographic characteristics, like e.g. student sample and randomly drawn population samples, still are very similar in their

responses to whether and under which conditions they want to receive genetic risk information. This clearly supports the generalizeability of the findings and conclusions.

It may also be argued that it is also somewhat unfortunate that all studies have used the same method of data collection, i.e. questionnaires using hypothetical scenarios. Therefore findings may have been influenced by common method variance (Campbell & Fiske, 1959), which may artificially inflate the correlations between variables assessed by the same method. This may weaken the certainty with which conclusions can be drawn from this research. The advantage of using the same method was however the possibility to directly compare affected and non-affected groups.

Conclusions

Whether knowing that one has an increased genetic risk really is a health advantage depends on whether or not it actually leads to necessary behaviour change. This is of course an empirical question beyond the scope of this thesis. Let it be mentioned, however, that research indicates that it might, at least for some people lead to these changes (see e.g. Sanderson, Humphries, Hubbart, Hughes, Jarvis, & Wardle, 2008; Claassen, Henneman, Kindt, Marteau, & Timmermans, 2010). Possible negative psychological impacts of knowing that one is a mutation carrier has also not been a subject of this thesis; research seems to indicate however that there do not seem to be long term negative emotional consequences of testing positive (see e.g. Meiser et al., 2008; Green et al., 2009; Kaphingst & McBride 2010). Knowing that one is *not* a mutation carrier (which will be the case for the majority of people undergoing genetic testing) is of course an indisputable advantage. It brings relief from possible worry and frees one from undergoing possibly harmful screening procedures.

The present thesis found participants to be interested in receiving genetic risk information and they displayed a relatively high acceptance of confidentiality breaches.

Patients were even more positive towards receiving information than the general population. The main individual predictor in all samples was uncertainty avoidance; the most important disease characteristic was treatability. Findings support the common practice of encouraging patients to inform relevant family members about their increased genetic risk. Still there are some participants both in the general population and among patients who state that they do not want to receive such information. If one is to take their right of *not* knowing seriously, it will be necessary to find ways to protect them from this unwanted information. Findings also showed that it will be important to inform the public about the remaining uncertainties of genetic testing in order to facilitate informed choice and to avoid possible misconceptions about the degree of certainty genetic testing can provide.

Furthermore, findings point to the possible importance of including affective outcome expectations into the attitudinal measures of the TPB. The present research also found that asking informants about the possible advantages and disadvantages of genetic testing did not elicit all belief based attitudes which predicted intentions in the TPB-model. Future research might find a better way to elicit less assessable attitudes like e.g. affective outcomes which might further increase the predictive power of the model.

Finally, current legislations in both Norway and Sweden specifically forbid physicians to breach confidentiality in order to inform at risk relatives. The majority of foreign jurisdictions (World Medical Association, World Health Organization, Council of Europe, Nuffield Council on Bioethics, Health Council of the Netherlands, Privacy Commissioner of Australia) are taking a different stance. While maintaining that confidentiality must be ensured and protected, they allow limited disclosure of genetic test results without the patients consent in cases where the harm to at-risk relatives is grave and imminent, and the information could result in effective intervention (ASHG, 1998; Godard et al., 2006). The findings of the current thesis show that the attitudes of both the general population and

certainly of the people concerned, i.e. patients with an increased risk for hereditary conditions, seem to support the later stance, which allows for confidentiality breaches under certain conditions.

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