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THE HEBREW UNIVERSITY OF JERUSALEM

**ON NOT WANTING TO KNOW AND NOT
WANTING TO INFORM OTHERS: CHOICES
REGARDING PREDICTIVE GENETIC TESTING**

by

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Discussion Paper # 406

September 2005

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Paper appeared in: *Risk Decision and Policy* 9 (2004) 317- 336.

**On Not Wanting to Know and Not Wanting to Inform Others:
Choices Regarding Predictive Genetic Testing**

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Abstract

Recent advancement in genetics testing for late-onset diseases raises fundamental decision dilemmas. The first study surveyed people's willingness to undergo predictive testing to find out about their own predisposition for certain incurable, late-onset diseases. The second study investigated the respondents' willingness to be tested as a function of the base rate of the disease, test diagnosticity, and the availability of treatment for the disease. In addition we surveyed (in the first study) people's willingness to disclose to others personal information about their genetic predisposition. The findings show that people often prefer not to know, as if they are choosing "protective ignorance". Respondents' verbal justifications of their choices were also analyzed. Respondents offered emotional, cognitive-instrumental, and strategic reasons for their preferences. The findings are compared with other issues in behavioral decision theory, including attitudes towards uncertainty and desire for control. The implications of the findings for policies and legislation on genetic tests are also considered.

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A simple test is available that can validly detect carriers of a genetic predisposition to a late-onset, incurable, fatal disease. Would you like to have this particular test done in order to know your genetic status? In general, knowing is considered to be the dominating choice since accurate prediction is a key to making good decisions, taking action, and attaining control over one's own future. But the evidence regarding predictive testing for incurable diseases, such as Huntington's disease, suggests otherwise (Kessler, 1994). Surveys of individuals at high risk show that a sizeable proportion of the respondents prefer not to know.

The two studies reported here are focused on the approach-avoidance conflict with regard to knowing one's own genetic status, and the idea that maintaining uncertainty about one's own predisposition is appealing as a form of self-protection. Another issue is people's (un)willingness to inform others about their genetic predisposition. We focus on genetic testing for two related reasons. First, predictive testing for late onset conditions is becoming a realistic possibility and most probably will continue to grow in importance and use. Indeed, legislation on the use of predictive testing is under way in various countries.¹ Empirical findings on people's intuitions and opinions about predictive testing ought to inform the ethical and legal debates that surround such legislation (Jungermann, 1997). Our second major goal is to shed light on theoretical concepts, such as protective ignorance and preference for uncertainty, and their relevance to behavioral decision theory.

The Dilemma of Predictive Testing

Research in genetics in the last few decades has enabled early detection of late-onset diseases. Predictive genetic testing means that individuals who are carriers of genes for particular diseases can be diagnosed long before they show any symptoms. One late-onset disease that has been subject to psychological research is Huntington's disease, an incurable hereditary disorder characterized by slowly progressing neurological impairment with onset typically between ages 30 and 50. The disorder is characterized by physical and mental deterioration (e.g., dementia, changes in behavior and personality such as aggression and paranoia), socially embarrassing choreiform movements, regressive problems such as incontinence, all leading to extreme dependency. The symptoms worsen until the patient becomes totally incapacitated and bedridden. The disease leads to the patient's death 10 to 20 years after its onset.

Due to the hereditary nature of Huntington's disease, individuals who have a parent affected with the disease are at risk. An offspring (male and female alike) of an affected person has a 50% chance of inheriting the disease-producing gene.² The experience of being at risk for Huntington's disease was described by Wexler (1979) as genetic 'Russian roulette.'

Predictive testing for Huntington's disease has been made possible by recent breakthroughs in genetics research (Gusella et al., 1983; Huntington's Disease Collaborative Research Group, 1993). Presymptomatic (including prenatal) tests for gene carriers yield either a favorable result – the person is not a carrier and will not develop the disease – or an unfavorable result – the person carries the defected gene and will certainly develop the disease in the future. A favorable result may come as a relief, especially to individuals at risk, whereas an unfavorable result causes such agony that Wexler (1979) compared it to “sentencing one to life with a time bomb.”

To Know or Not to Know

The decision on whether or not to undergo a genetic test for a particular condition can be represented in a decision tree, as in Figure 1. The decision is between states of knowledge, that is, whether to maintain the current state of ignorance and uncertainty or to opt for a resolution of the uncertainty.³ The top node represents the person's choice point between resolution (left branch) and ignorance (right branch). Following a choice to have a test done (left branch) two consequences are possible. The person will receive unfavorable news with probability p and favorable news with probability $1-p$. For a person randomly drawn from the population, p is the base rate of the disease in the population, whereas for a person at risk p is 50% (or, if only a grandparent is known to be ill, 25%). Thus, for a person at high risk, the test results alter the a priori probability of developing the disease from 50% to either 0% or 100%. For a person drawn at random, the change is from a base rate of about 1:10000 to either 0% or 100%. Numerous consequences follow from each outcome, with multifaceted implications on emotional, cognitive, social, and financial levels. These consequences are represented in the terminal nodes of this tree.

[Insert Figure 1 about here]

How might one choose? The choice here is among various “states of knowledge,” each carrying a different mix of severe threats and potential benefits with specified probabilities. Within the framework of normative decision theory (e.g., Luce & Raiffa, 1957), choices are made between the two branches by evaluating the expected value of each major branch in this tree and selecting the one that yields a higher value.⁴ Psychologically, the decision to be tested for Huntington's disease presents a dilemma. By choosing to be tested individuals can resolve the uncertainty about their genetic predisposition. But, in the present case, receiving an unfavorable result has little instrumental value in terms of treatment. If they choose not to be tested, individuals can maintain ignorance or uncertainty. While this choice makes it more difficult to make future plans, it also helps avoid the anticipated sharp affective reaction to bad news and leaves room for hope and optimism that one is not inflicted.

Results of Previous Surveys

Do people at risk opt for resolution of their precise genetic status? Surveys of individuals at risk for Huntington's disease were done at health centers in Western Europe and North America, both before and after testing became possible for the first time in the mid-1980s. The surveys focused on the respondents' attitudes towards predictive testing and their actual participation in the testing programs and perception of the benefits and costs of testing.

In various early surveys 50 to 80% of the individuals at risk showed an interest in predictive testing for Huntington disease given the opportunity (Kessler, Field, Worth, & Mosbarger, 1987). Later studies showed that only about 15% actually applied for such testing (Decruyenaere et al., 1995; Meissen et al., 1991; Quaid & Morris, 1993; Tyler, Ball, & Craufurd, 1992). A survey of individuals at risk who elected not to participate in a predictive testing program in the Netherlands (van der Steenstraten et al., 1994) suggested that nonparticipants feared that predictive tests would confirm that they had Huntington's disease, and that they would not be able to handle such a confirmation. In another survey, Tibben et al. (1993) interviewed individuals at risk for Huntington's disease who had applied for presymptomatic testing. They found that the major reason for deciding against being tested was the fear of adverse emotional effects after an unfavorable diagnosis, such as deprivation of hope, life in the role of a patient, obsessive searching for symptoms, and inability to support one's spouse. In contrast, the reasons cited for being tested included the opportunity to make plans for the family and the resolution of uncertainty.

Meissen et al. (1991) asked individuals at risk about their views regarding the future consequences of getting either favorable or unfavorable outcomes. Respondents believed that getting unfavorable test results would cause mainly adverse emotional consequences, such as depression and difficulties with their partner (and children), but also expected benefits, such as forcing them to plan, get more insurance, and focus on their present life. They believed that getting favorable test results would lead to benefits such as reduction of anxiety, family planning and enhancement of family relationships as well as adverse consequences, primarily guilt feelings towards others at risk, especially towards siblings who had received unfavorable news.

Willingness to Inform Others

Suppose you have been told that you are a carrier of the gene for Huntington's disease. Whom (if anyone) would you be interested in or willing to share this information with? A second facet of the present study involves people's willingness to disclose the information to various third parties, ranging from close relations to strangers. A variety of emotional as well as instrumental or strategic reasons affect the choice to disclose information regarding one's

own genetic condition. One could choose to share the information with family members in order to seek support and alert those genetically related to their potential genetic state. One could, alternatively, withhold the unfavorable information so as not to cause anxiety. Hiding the information from third parties, such as members of one's social milieu and potential or actual employers, could serve to protect one's economic interests and social status and to avoid stigmatization.

For instance, Littlewood and Elias (2000) found that a chief reason for avoiding screening for breast cancer among women from various ethnic minorities in London was their fear of "loss of reputation" within the community. Concern about stigmatization is a major sticking point in the debate on legislation, where lawmakers representing the public (consumer) interest are in sharp opposition to representatives of the insurance industry (The Ad Hoc Committee on Genetic Testing /Insurance Issues, 1995; Kolata, 1986).

The dilemmas surrounding the right for privacy of personal genetic information is even more acute vis a vis genetically linked family members. Lehmann et al. (2000) surveyed attitudes regarding one's duty to inform family members about their susceptibility to genetic ovarian-breast cancer. They found that women's attitudes towards the disclosure of genetic information to family members differed depending on whether or not the disease was preventable. But respondents also felt strongly that in both preventable and non-preventable diseases physicians should not seek out and inform at-risk relatives against a patient's wishes. In sum, respondents faced a conflict between family members' need to know and the right for confidentiality of information disclosed within a physician-patient relationship. The disclosure of information raises dilemmas vis a vis close relatives as well as remote third parties.

First Study

The research used a scenario based on Huntington's disease whose characteristics – incurable, late onset, genetically identifiable – present the dilemma of predictive testing in its sharpest form. The results of this specific case appear to apply to a broad range of predictive genetic tests, since the sorts of dilemma raised are general.

Participants were told that the research was being done in collaboration with the human genetics department at the university hospital, that the information in the questionnaires was based on existing medical knowledge, and that recent developments in genetic research allow early detection of various diseases in the general population, even among those who do not have any family history of genetic diseases. Respondents were presented with a description of "disease X" and were told that it roughly corresponds to an actual disease (although the term Huntington's disease did not appear in the questionnaire). They were then told that there is a simple blood test that can identify the carriers of the genetic defect causing this disease.

Our respondents were recruited from the general population. Thus, unlike many previous studies that targeted people at risk, our respondents did not belong to any particular high risk group. Our rationale was that the issue of finding out about one's genetic status (for certain diseases) and the problem of whether to inform others about it are becoming relevant to people at large and not only to those at high risk due to their family history.

We investigated two facets of people's preferences. The first was their interest in obtaining information about their own genetic status (and that of their present or future spouse/partner). The second was their willingness to share unfavorable test results with family members, friends or third parties, such as employers or national data pools. The respondents were also asked to indicate the reasons for their preferences.

Method

The respondents were given a questionnaire with two parts. The first included the following general description of a genetic disease: "Disease X is a terminal illness that typically appears between the ages of 35 and 50. The disease causes brain degeneration, and hence progressive deterioration of physical and mental abilities over a period of 10-20 years, until death. The frequency of disease X in the population is 1 in 10,000. The disease is caused by a genetic defect and can be detected before the onset of symptoms by taking a blood sample. Individuals found to be carriers of the gene have a 100% chance of developing the disease. Those found not to be carriers the defective gene will not develop the disease. There is no known treatment that can effectively prevent or cure the disease."

In the first part of the questionnaire, the respondents were presented with a series of questions regarding their interest in obtaining such information about themselves and their partners. First they were asked to indicate their interest in being tested themselves:

"Since the carriers of the gene causing the disease can be identified, is the genetic identification of disease X the sort of information that you would want to have about yourself? Please use the following rating scale:

1-----	2-----	3-----	4-----
would definitely <u>not want</u> to know	would probably <u>not want</u> to know	would probably <u>want</u> to know	would definitely <u>want</u> to know

Next, respondents were asked to explain the reasons for their choice in the space provided, using their own words.

Two subsequent questions elicited their choices with regard to: (1) a hypothetical potential partner; (2) a steady partner / spouse (if they had one). The first one was phrased: "Imagine you had recently met someone whom you consider a potential partner for an extended relationship. Would the genetic identification of disease X be the sort of information that you

would want to have about your new partner?” Next, individuals who were either married or involved in a steady relationship were asked the following question: “Would the genetic identification of disease X be the sort of information that you would want to have about your spouse or steady partner?” Participants rated each preference on a 4-point scale and then provided their explanations.

The second part of the questionnaire investigated respondents’ willingness to share genetic information about themselves with other parties. Specifically, they were told, “Imagine, just for the sake of the present questionnaire, that you have been diagnosed as a carrier of the defective gene that causes disease X. Would you want to inform your steady partner or spouse?

1-----2-----3-----4
 would definitely would probably would probably would definitely
not want them to know not want them to know want them to know want them to know

This question was presented with respect to each of the following parties: (1) your steady partner or spouse; (2) a new (potential) partner you have just met; (3) your brothers and/or sisters; (4) your parents; (5) your best friend; (6) your employer; and (7) a national data pool (government agency). A rating scale was provided for each question, and in addition respondents were asked to provide their reasons for each of their ratings.

The respondents (n = 167) in this study were 22- to 40-year olds who volunteered to participate in a survey on personal decisions. The sample included social-science students at the Hebrew University and employees in a Jerusalem-based organization (Joint-Brookdale Institute).

Results and Discussion

Finding out. The analysis considered the preference ratings as well as the reasons given for them. The ratings on the 4-point scale were collapsed into two categories, “yes” (ratings of 3 or 4) and “no” (ratings of 1 or 2). Table 1 (panel 1a) shows the proportion of respondents who gave “yes” responses. The survey results show that 52% of the respondents were interested in obtaining genetic information about their own status. Regarding the choice to find out about partners, among those who had a steady relationship (n=89), 45% would be interested in getting information about their spouse/ steady partner. The respondents’ interest in finding out about their steady partners was highly correlated with their interest in finding out about themselves ($r = 0.80, p < .001$). Among those who did not have a steady relationship (n=78), 65% would be interested in obtaining this sort of genetic information about a hypothetical new partner. Here also the choice to find out about a new partner was correlated with the choice to find out about one’s own status ($r = 0.48, p < .001$). In sum, about half of the respondents avoided finding out about their own genetic status or that of their significant others, although

less so in the case of hypothetical new partners. The internal consistency among the three preference measures was relatively high (Cronbach's coefficient = 0.76), suggesting that they all tapped similar aspects of the same general personal tendency towards avoiding (or seeking) relevant threatening information.

[Insert Table 1 about here]

A qualitative analysis of the reasons for the preference ratings sheds light on the nature of this avoidance. A list of categories of reasons was prepared, based on pilot research that showed that this procedure could be carried out with high inter-judge reliability. A given explanation could contain none, one or more than one category of reasons. For the analysis one coder coded each explanation for the occurrences of any of the predefined categories. To assess the reliability of coding, a second, independent coder coded a subset of the data. The agreement rate between the two coders was 84%. The differences between them were minor, so none of the substantive conclusions would have changed as a result of their disagreements. This procedure was repeated for each of the ten survey questions.

As Table 2 shows, the most common reason for not wanting to know about one's genetic status was the lack of treatment ("There is nothing I could do, so why should I find out about it," "I cannot control my fate," "Such knowledge would be useless as long as there is no way to cure, treat or prevent the disease.") Reasons in this category were mentioned by 65% of those who did not want to find out and by only 8% of those who wanted to. Another class of reasons involved anxiety, depression, and stress ("I would not want to live in constant fear," "I would want to continue my life as usual and not in the shadow of a ticking bomb.") Such reasons were mentioned by 34% of those who did not want to find out and by 5% of those who did. The most common reason for wanting to find out was future planning (e.g., "I will be better able to plan my life, my career, having children, and taking a trip around the world", "I will get organized, get insurance, prepare for my kids and their future needs," "I will make the most of my life, use best the time I have left.") Such reasons were mentioned by 76% of those who chose to find out and 10% of those who chose not to.

Fairly similar types of reasons were used by the respondents to explain their choice of whether or not to find out about the genetic status of their potential or steady partners. Two reason categories that did not occur in explanations about own status stand out here. First, some people indicated that genetic information could influence their decision to maintain or abandon relations with a newly acquainted partner (reasons for not wanting to find out: "I would not want this to be a criterion in choosing a partner," "I love my partner and I would not want this information to influence me,"; reasons for wanting to find out: "I would not start a relationship with someone who has this gene"). The second observation is that respondents justified (or rationalized) not wanting

to find out about their significant others' genetic status by saying that there was no reason to "single out" this particular disease given the sheer number of other risks that their partners might be exposed to during their lifetime ("Life is unpredictable anyway, even a partner with 'good' genes could die tomorrow in a road accident," "You take risks in life," "So many other bad things can happen.")

[Insert Table 2 about here]

Informing others. The second part of the research investigated people's willingness to share unfavorable information with third parties. Table 1 (panel 1b) shows the percentage of respondents who said they would be willing to share information with a third party. Whereas a large majority would disclose information to steady and new partners, siblings, parents, and friends (61 to 92%), few would disclose it to remote parties (14 and 10%, for national data pools and employers, respectively). The means of the seven ratings of willingness to disclose information (Table 1b) differed greatly in their absolute levels, but the ratings also exhibited internal consistency (Cronbach's coefficient = 0.70). We conclude that, one can speak of a common, general tendency to disclose personal genetic information, although the strength of this tendency is graded, depending on the third party.

Table 3 shows the frequencies of the various types of reasons that were mentioned. The chief reason for sharing information with one's partner was distaste for secrecy ("It's dishonest to hide such information", "I would not want lie to my partner," "We must tell each other everything and cope with it together") and future planning ("So we can plan whatever remains of our future together"). The major category of reasons for not informing potential new partners was fear of desertion. The reasons for informing siblings and parents included mostly the need for support ("So I will have someone to talk to", "So they will know what is happening to me...I will need a good deal of support"), and also the potential medical benefit to the genetically-related family members ("So they can be tested as well"). In contrast, the main reason against informing parents and siblings was to refrain from placing the burden on others ("Why worry them," "So they don't suffer").

The main reason for not sharing information with an employer was fear of stigmatization and discrimination, which might damage one's position in the job market ("This would be a fatal stigma. Who would want to hire me?" "This will lead to discrimination against me, even provide a reason for firing me.") This reason was mentioned by 51% of the respondents. The main reason against placing the information in a government agency data pool (64%) was concern for privacy. The fear of losing control over the information was often mentioned along with the risk of it falling in the hands of someone who could use it against one (e.g., "This is a fatal label, who would want me?" "This would be an invasion of privacy," "This is private

information, it is none of their business”). Of those few who were willing to share genetic personal information with a government agency, most cited benefit for research as their main reason (71%).

[Insert Table 3 about here]

In sum, the reasons for or against disclosing unfavorable test results to family members included a myriad of emotional and strategic considerations, even conflicting ones, such as seeking support on the one hand, and wishing not to burden others, on the other. In contrast, the reasons mentioned in response to the questions about remote third parties (national data pool and employer) were strategic, involved impression management and damage control, and geared at not suffering any social stigmatization or economic harm as a result of the new information.

Finding out vs disclosing to others. There was little evidence in these data for correspondence between the willingness to find out genetic information and the willingness to disclose personal genetic information to others. The correlations across the two sets of measures were fairly small. For instance, the correlation between wanting to find out the genetic status of one’s steady partner and the intention to disclose personal information to one’s (steady) partner was 0.14 ($n=91$, $p>.1$); in the same vein, the correlation between wanting to find out a new partner’s status and the intention to disclose information to a new partner was 0.07 ($n=78$, $p>.1$). Indeed, the content analyses suggest that different considerations underlie the answers to the two types of questions. In explaining the option to disclose information to others, respondents frequently used instrumental and strategic terms, such as seeking support and impression management (e.g., so as not to harm one’s future job prospects). In considering the option to find out for themselves, respondents mentioned mainly emotional reactions, such as fear and anxiety, and utilitarian issues, such as the importance of planning.

Study 2

In Study 1, the survey of attitudes towards being tested found that respondents’ interest in finding out their genetic predisposition for an incurable late-onset disease was far below 100%. The content analysis of the reasons indicated that the apparent lack of control over the disease was a decisive factor in reducing people’s willingness to find out about it. What role did other characteristics of the disease play – most notably, its rarity and the high diagnosticity of the gene? Other diseases vary along those parameters. For example, women with the susceptibility genes for ovarian-breast cancer have about a 10-40% chance of contracting ovarian cancer and about a 60-80% chance of contracting breast cancer over their lifetime (Ford et al., 1998), and the incidence of these types of cancer is much higher than that of Huntington's disease. The content analysis of the previous study revealed that reasons involving statistical prediction factors, namely base rate and

diagnosticity, were hardly ever mentioned (e.g., base rate was mentioned by less than 5% of the respondents). Nevertheless, these variables might still have an effect on respondents' ratings of their willingness to be tested if they are manipulated in an experimental fashion. In Study 2 we systematically varied the description of disease X from Study 1 to test these factors.

Method

The disease description presented to respondents was similar to the one presented in the previous study. As before, no specific name of any real disease was ever mentioned. Three factors were varied experimentally in the description of disease X in order to test their effects on respondents' willingness to undergo testing: (1) the availability of treatment, (2) the diagnosticity of the test, and (3) the base rate of the disease in the population. For the treatment factor, respondents were told either that “there is no known treatment that can effectively prevent or cure the disease” or that “there is a treatment that can effectively prevent and cure the disease.” The base rate quoted was either low (1:10,000) or high (1:50). The diagnosticity of a test was presented in terms of conditional probabilities. Thus the probability of developing a disease if the person is not a carrier was 0 in all conditions; the probability of developing the disease if the person is a carrier was 100% in one condition and 60% in the other.

In sum, eight conditions created by varying three orthogonal factors: treatment (incurable or curable), diagnosticity (100% or 60%), and base rate (1:10,000 or 1:50). The participants (N = 120) were social science and humanities undergraduate students at the Hebrew University who volunteered to fill out the questionnaire. The questionnaires were randomly assigned to 15 respondents in each of the eight conditions.

Results

Two analyses were conducted on the same data, the first treating the data as binary (yes/no) decisions and the second using the 4-point rating scale. The results of the two closely agreed. Ratings were grouped into two response categories, “yes” (ratings 3 or 4) and “no” (ratings 1 or 2). Table 4 also shows the proportion of respondents who gave yes responses (rating 3 or 4). The proportion of respondents who were willing to be tested ranged from 33 to 47% in the no-cure condition and from 80% to 93% in the cure condition. A log-linear analysis of respondents' willingness to be tested was conducted, with treatment, base rate, and diagnosticity as factors (N=120). The only significant effect was treatment effectiveness, $\chi^2(1) = 22.2, p < .001$, while the other factors and interactions were not significant, $\chi^2 < 1$.

[Insert Table 4 about here]

A three-way analysis of variance on interest in being tested (4-point rating scale) was conducted with treatment, base rate, and diagnosticity as between-subjects factors (see means in

Table 4). We found a significant effect of treatment, $F(1,117)=39.4$, $p < .001$, but no effects of base rate and diagnosticity; none of the interactions was significant, $F < 1$ for all other effects. Inspection of effect sizes (in units of standard deviation (sd) Cohen, 1962) is instructive.⁵ The effect size for treatment was large, 1.90 sd, whereas the effect sizes for base rate and diagnosticity were virtually nil, 0.04 and 0.01 sd, respectively.

Discussion

Only the treatment factor had an effect on respondents' willingness to be tested. When no treatment was available more than half of the participants opted for not finding out, a proportion similar to that seen in the first study. The availability of treatment increased greatly individuals' interest in being tested, consistent with the finding of another study that involved samples from the general population (Shiloh, Ben-Sinai, & Keinan, 1999). The diagnosticity of the test or the base rate of the disease had no effect on this interest. The respondents' unwillingness to find out about their predisposition for an incurable condition seems stable across the range of parameters used here. In one previous survey women (not at risk) were told to imagine they were at 50% risk for Huntington's disease. About half of them said that they would undergo testing and a similar percentage indicated that they would consider prenatal testing under these conditions (Decruyenaere et al., 1993). It seems that the various base rates shown in our study (1:10000, 1:50), and the one used in the survey above (1:2), did not change fundamentally the nature of the decision.

This result is consistent with our finding based on the content analysis of the reasons collected in the first study that the statistical prediction factors had hardly any bearing on the decisions. It appears that the affective response to the threat of the disease, the vividness of the deterioration, and the lack of control, have far greater impact than the more pallid dimensions namely, base rate and validity which are harder to evaluate as factors in the decision. Loewenstein, Weber, Hsee, & Welch, (2001) suggest in the context of their framework of risk-as-feelings that in evaluating choices, strong emotional response tends to supersede more cognitive responses of dimensions, such as probability. Similarly, in support of their "affect heuristic" Slovic, Finucane, Peters, & MacGregor (2002) bring evidence that choice aspects which elicit an affective response (e.g., fear, pain, and pleasure) carry more weight in the final decision than aspects that are harder to evaluate due to absence of context or lack of a standard for comparison.

General Discussion

We asked respondents about their willingness to undergo predictive genetic testing for an incurable, late-onset "disease X" whose characteristics were similar to those of Huntington's disease. A central finding of the present studies is that nearly half the respondents preferred not

to find out their own genetic predisposition to this disease. We note that in normal practice predictive genetic testing for various diseases is typically offered in the context of other risk factors, such as family history, or under special circumstances, such as pregnancy. Our questionnaire was hypothetical and presented to people who were not at increased risk (cf. Welkenhuysen, Evers-Kiebooms, & van den Berghe, 1997). Nevertheless the responses of our respondents (who were not at risk for any genetic disease) were generally consistent with those obtained in previous surveys of populations at increased risk (due to family history) for Huntington's disease (Decruyenaere et al., 1995; van der Steenstraten et al., 1994). We suggest that the study of people who are not at risk is useful since genetic screening is becoming more prevalent. In a different vein, the experimental investigation of people at large regarding the choice between resolution and ignorance, could shed insight on basic issues in risky choice, attitudes toward uncertainty, and information seeking, as we shall see below.

Protective Ignorance: Descriptive and Normative Issues

Individuals sometimes choose to protect themselves against knowing about outcomes that are both threatening and uncontrollable. People's preference not to find out, that is, to maintain the uncertainty about one's genetic status, can be labeled "protective ignorance." Indeed, the respondents' reasons for choosing not to find out agree with the interpretation that uncertainty shields the decision maker from anticipated anxiety, stress and depression.

The concept of protective ignorance – a "veil of uncertainty" about the outcome – seems an adaptive approach vis a vis outcomes that are threatening and uncontrollable at the same time. Such a perception is reflected also in common ethical practices involving predictive genetic testing. At Leiden University in the Netherlands, parents are refused the right to submit a minor child to predictive testing for Huntington's disease (de Wert, 1992). Prenatal testing (at Leiden) for Huntington's disease in an unborn child is limited to parents who intend to abort the pregnancy in case of a positive test result. De Wert (1992) explains such restrictive testing policies on philosophical and psychological grounds. First, testing done by the parents infringes on the child's moral right not to know. Thus the child's right not to know supersedes his or her parent's right to know. Second, knowing with certainty that a child will develop a fatal disease could have detrimental effects on the child's upbringing and family relationships (Bloch & Hayden, 1990). Knowing with certainty might destroy valid hopes, leaving room only for illusory hopes. In contrast, under the shadow of uncertainty, parents can remain hopeful. The probabilistic information allows parents to foster a certain degree of unrealistic optimism, with its attendant benefits for mental well-being (Taylor & Brown, 1986).

Such a preference for ignorance may not be evoked in the case of genetic diseases that can be treated to some degree. For instance, the level of interest in being tested is about 90% among

people at risk for hereditary kidney disease (Sujansky et al., 1990) and 70 - 90% in surveys of women's attitudes tests to breast and ovarian cancer (Croyle & Lerman, 1993; Lerman, Seay, Balshem, & Audrain, 1995; Shiloh, Petel, Papa, & Goldman, 1998). In a recent study, Shiloh, Ben-Sinai, and Keinan (1999) found that people's willingness to be tested depended on the availability of effective treatment for the disease tested. Indeed, in our Study 2 the availability of treatment largely boosted the interest in being tested.

The preference for ignorance or uncertainty is in sharp contrast with a normative dictate that underlies decision theory, namely that one ought to use all the information relevant to one's subjective beliefs, decisions, and actions (see discussion in Ullmann-Margalit, 2000). Abundance of evidence both from informal observation and experimental studies shows – what almost needs no proof – namely, that people both seek and highly appreciate certainty. At the cognitive-instrumental level, uncertainty hinders planning and limits one's ability to exert control over one's future. At an emotional level, uncertainty causes a sense of discomfort and apprehension (Lazarus & Folkman, 1984).

So how does a tendency to remain in the “shadow of uncertainty” help? A key idea here is that people tend to enhance the contrast between possibility and certainty (e.g., Lopes, 1987) – a phenomenon best demonstrated (within expected utility theory) in the Allais paradox (1979) and Tversky and Kahneman's (1981) “certainty effect.” The lure of certainty is so high that individuals are willing to pay a disproportionate premium to reduce a given risk to zero. The steep transition from possibility to certainty is also captured in the probability weighting function (Gonzalez & Wu, 1999; Tversky & Kahneman, 1981).

The enhanced contrast between possibility and certainty implies that a probabilistic outcome is highly discounted relative to the certain outcome. Considering now predictive genetic testing for threatening diseases, respondents presumably tend to compare the uncertain knowledge state (ignorance) with their knowledge state following the receipt of unfavorable test results. Maintaining the uncertainty about their status provides a sense of protection since the probabilistic event is disproportionately discounted compared with the certain knowledge.⁶ The enhanced contrast between possibility and certainty forms in our view the foundation for the hospital practices mentioned above with respect to prenatal testing and testing children. Our findings – the preference ratings as well as verbal justifications – are consistent with the view that one can hide in the “shadow of uncertainty” for defensive reasons.

Prospective vs Retrospective Benefits of Knowing

Looking ahead, a substantial proportion of people prefer not to know their genetic status with respect to an incurable disease. But, is protective ignorance a “good thing to have” in retrospect? That is, would people be better off knowing or not knowing? The question is not easy to answer.

There are preliminary findings suggesting that people who are tested are better off than those who choose not to be tested, even if they receive unfavorable results (Lerman et al., 1998; Sieff, Dawes, & Loewenstein, 1999; Wiggins et al., 1992). It should be noted that the samples in these studies were subject to self-selection biases (see critical discussion in Sieff et al., 1999). Nevertheless, such findings pose a challenge to the idea of protective ignorance.

How do we reconcile the notion of protective ignorance with the possibility that getting information about one's genetic predisposition might be beneficial to the recipient, even if the information is unfavorable? Kahneman and Snell (1990) suggest that making a decision requires one to forecast what one's emotional reactions of pleasure or pain will be when one experiences a future outcome. While under some conditions people's predictions are fairly accurate, there are situations where people systematically mis-estimate the experienced utility of an outcome. In particular, people tend to underestimate their ability to adapt to new situations (Kahneman & Snell, 1990).

Such prediction-experience gaps can explain the prevalence of protective ignorance and the finding that knowing one's predisposition is advantageous over not knowing. In making a forecast of his or her emotional reaction to unfavorable test results, a person may fail to appreciate the effect of family support, or the therapeutic potential of counseling on his or her well-being. For example, in the study by Wiggins et al. (1992) the psychological benefit was observed after a year, during which participants received consistent psychological support. The prediction-experience gap seems a reasonable explanation for the difference between the prospective and retrospective appreciations of the benefit of knowing. The potential gap between the two perspectives is intriguing and awaits further treatment.

Strategic Aspects of Informing Others

Access to genetic information raises strategic issues in addition to emotional ones. Revealing or sharing personal information with others affects how one will be perceived and treated by others. In our survey, the respondents' willingness to share information with various parties depended on the type of the relationship. Whereas a majority of the respondents would inform close friends and family, most of them expressed a clear preference not to share genetic information with third parties, such as employers and government agencies. Their stated reasons indicated concern for privacy and fear of stigmatization and discrimination.

Indeed, there is real cause for concern that personal genetic information would be used to discriminate against genetically disadvantaged individuals in the job market or in the market for insurance and bank loans. The moral basis for such discrimination is in question and has recently been debated in various countries, with proposals for legislation being discussed in some of them (The Ad Hoc Committee on Genetic Testing /Insurance Issues, 1995; Kolata,

1986). For instance, the recent Genetic Information Act passed in the Israeli parliament in 2000 severely limits the access of third parties to personal genetic test results.⁷ It also grants individuals the right not to be tested for certain incurable conditions, and follows the practice of not allowing parents to test their children for incurable conditions.

Final Remarks

The present work considered two types of dilemmas in the context of genetic testing. The first, and main one, was individuals' interest in finding out about their own genetic predispositions. The second involved people's willingness to inform others about their own status. Respondents' justifications of their choices were different for each dilemma. The limited willingness to inform certain third parties was justified mostly in strategic terms, including impression management, and the need not to harm one's future prospects. In contrast, the justification for the choice not to know was often cast in emotional terms, namely the anticipation of adverse emotional reactions to unfavorable test results, thereby leading to protective ignorance.

Nevertheless, one could imagine certain links between the two dilemmas. Knowing one's genetic predispositions could create a moral dilemma as to whether one ought to disclose the information and to whom (Ullmann-Margalit, 2000) -- for instance, at what point, if any, should one inform one's partners/significant others. This dilemma is particularly acute with respect to a newly introduced individual whom one considers to be a potential future partner.

The bases for the presumed benefits of protective ignorance remain intriguing and merit further work in the domain of genetic testing and also in connection with other phenomena, such as feedback seeking. Current choices could be affected by expectations regarding the availability of post-choice feedback (Josephs, Larrick, Steele, & Nisbett, 1992; Zeelenberg, Beattie, van der Pligt, & de Vries, 1996). Such findings suggest that protective ignorance may play a role in information search and feedback seeking and that its significance perhaps goes beyond its current application in the domain of genetic testing.

Footnotes

¹ Legislation considered in the United Kingdom (“Insurers to DNA test for genetic illnesses,” Daily Telegraph, March 20, 2000) and the United States (“Clinton proposes legislation to bolster privacy protections,” Wall Street Journal Europe, May 2, 2000).

² The gene for Huntington's disease is, in technical language, autosomal dominant, namely, it is not sex-linked and is certain to produce the disease if it is inherited from just one parent.

³ The common technical distinction between risk and uncertainty is not strictly followed here. This is because for some types of disease the probabilities are precisely known, whereas for others they are not.

⁴ The decision tree is presented only for illustrative purposes. Standard decision theory applies the expected value calculation to this decision. The application of such calculations is beyond the scope of the present research.

⁵ Effect size is measured by the difference between the marginal means divided by the pooled standard deviation. We follow Cohen's (1962) advice and treat effect sizes larger than 0.8 as high and those smaller than 0.2 as low.

⁶ One might argue, contrary to our position, that choosing the status quo (in Figure 1) indicates a preference for certainty (rather than uncertainty) since the outcome, ignorance, is guaranteed with certainty; thus, it is guaranteed with certainty is that one will remain uncertain about one's genetic status. The paradox arises since the outcomes are knowledge states, rather than being goods, wealth states, or health states. Consider however an individual who chooses to undergo diagnosis of an easy-to-cure illness. Such a choice is likely to be viewed as a preference for resolution (certainty), rather than for uncertainty. Therefore we argue that the knowledge state denoted as status quo would be aptly labeled “preference for uncertainty”.

⁷ See Knesset site (in Hebrew)

<http://www.knesset.gov.il/asp/hebframe.asp?content=/laws/heb/laws.asp?type=2>

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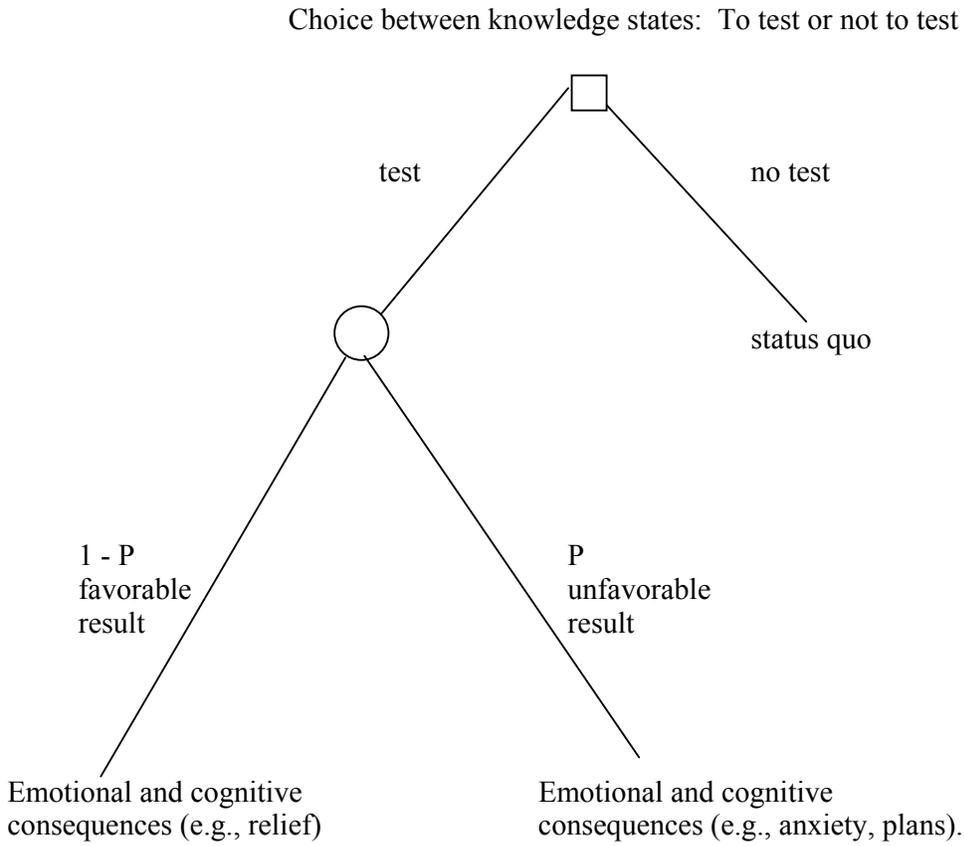


Figure 1. Depiction of the decision tree for the choice to know. Only some of the immediate psychological consequences of each outcome are illustrated in the terminal nodes.

Table 1

Results from Study 1.(1a) Interest in Finding out about Own and Partner's Genetic Status

	<u>% respondents saying "Yes"</u>	<u>Mean rating</u>
Self	52%	2.55
Your steady partner/spouse	45%	2.48
A potential partner you recently met	65%	2.82

(1b) Willingness to Inform Third Parties about Unfavorable Genetic Status

<u>Other Party</u>	<u>% respondents saying "Yes"</u>	<u>Mean rating</u>
Steady partner/spouse	92%	3.56
A new partner you met	81%	3.29
Brothers and sisters	75%	3.12
Parents	61%	2.87
Best friend	63%	2.74
Employer	10%	1.68
National data pool	14%	1.60

Table 2

Study 1: Explanations of the Choice (not) to Know: Frequencies (%) of Various Reason Categories as a Function of the Choice to Find Out.

	<u>Reasons (%) mentioned by those who</u>	
	chose to know	chose not to know
<u>About self</u>		
Lack of treatment	8	65
Anxiety, stress, depression	5	34
Future planning	76	10
<u>Steady partner/spouse</u>		
Lack of treatment	0	26
Anxiety, stress, depression	2	19
Future planning	57	35
“This is not the only risk in life”	0	11
<u>New Partner</u>		
Lack of treatment	3	13
Anxiety, stress, depression	2	7
Future planning	76	31
“This is not the only risk in life”	0	28

Note: Entries in the table show the percentages of respondents who mentioned each reason category. Since respondents could mention one or more reasons (or none) the numbers do not add to 100%. Reason categories that occurred only infrequently are not presented in this table.

Table 3: Study 1: Explanations of the Choice (not) to Inform: Frequencies (%) of Various Reason Categories as a Function of Willingness to Inform.

	Reasons of those <u>willing</u> to inform	Reasons of those <u>not willing</u> to inform
<u>Steady partner/spouse</u>		
Against secrecy	59	21
Future planning	27	7
Gain support	10	7
<u>New Partner</u>		
Against secrecy	68	3
Future planning	25	13
No commitment yet to partner	1	45
Fear of being left	0	23
<u>Siblings</u>		
Against secrecy	18	0
Gain support	43	0
Potential benefit to siblings (be tested)	23	7
Afraid to burden them	4	90
<u>Parents</u>		
Against secrecy	16	0
Gain support	48	0
Potential benefit to parents (be tested)	9	2
Afraid to burden them	13	98
<u>Best Friend</u>		
Against secrecy	20	0
Gain support	72	3
Afraid to burden them	1	16
Need to maintain own privacy	3	36
<u>Employer</u>		
Fear of stigmatization & discrimination	18	51
Need to maintain own privacy	0	29
<u>National pool</u>		
Fear of stigmatization & discrimination	0	18
Maintain privacy & not lose control over information	12	64
Promote research	71	2

Note: Entries in the table show the percentages of respondents who mentioned each reason category. Since respondents could mention one or more reasons (or else none) the numbers do not add to 100%. Reason categories that occurred only infrequently are not presented in this table.

Table 4

Study 2: Interest in Predictive Testing as a Function of the Experimental Factors

Treatment	Base rate	Diagnosis- ticity	Preference	
			Mean rating	% yes
Incurable	1:10,000	100%	2.5	47%
		60%	2.3	47%
	1:50	100%	2.1	33%
		60%	2.5	40%
Incurable	1:10,000	100%	3.3	80%
		60%	3.3	87%
	1:50	100%	3.5	93%
		60%	3.3	87%