Adverse psychological events occurring in the first year after predictive testing for Huntington's disease. The Canadian Collaborative Study Predictive Testing.

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Adverse psychological events occurring in the first year after predictive testing for Huntington's disease

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Abstract

A total of 135 participants in the Canadian predictive testing programme for HD were followed for at least one year in one of four study groups: increased risk (n=37), decreased risk (n=58), uninformative (n=17), or not tested (n=23). Clinical criteria for an adverse event were a suicide attempt or formulation of a suicide plan, psychiatric hospitalisation, depression lasting longer than two months, a marked increase in substance abuse, and the breakdown of important relationships. Quantitative criteria, as measured by changes on the General Severity Index of the Symptom Checklist 90-R and the Beck Depression Inventory, were also used to identify people who had adverse events.

Twenty of the 135 participants (14.8%) had an adverse event. There were no significant differences between those with or without an adverse event with respect to age, sex, marital status, education, psychiatric history, general psychiatric distress, or social supports at baseline. However, evidence for depression was associated with an increased frequency of adverse events ($p<0.04$). The adverse events were similar and seen with equivalent frequency in those receiving an increased risk or decreased risk and persons at risk who did not receive a modification of risk. However, a significant difference was found in the timing of adverse events for the increased and decreased risk groups ($p<0.0002$). In the increased risk group all of the adverse events occurred within 10 days after results whereas, in the decreased risk group, all of the adverse events occurred six months or later after reviewing test results.

These results suggest that people entering into predictive testing with some evidence of clinical depression warrant special vigilance and also suggest that counselling and support should be available for all participants in predictive testing irrespective of the direction of test results.

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Key words: Huntington's disease; predictive testing; adverse events; psychosocial consequences.

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Huntington's disease (HD) is an autosomal dominant genetic disorder characterised by involuntary movements, cognitive decline, and mood disturbance, followed by death usually within 15 to 20 years of onset.[1] Children of subjects affected with HD have an a priori risk of 50% for inheriting the gene and thus developing HD in the future. The fact that it is a late onset disease, usually manifesting in midlife, means that those at risk live a great part of their lives and make many important decisions (concerning career, marriage, and children) with uncertainty regarding their future.

In light of the fact that no cure or effective treatment is available for HD, numerous concerns and cautions were raised before the implementation of predictive testing programmes regarding possible adverse reactions to a modification in risk status.[2–5] The majority of the concerns focused on the possible negative reactions to an increased risk result, including depression, anxiety, marital problems, financial difficulties, and, of course, suicide.[3] Surveys of the at risk population aimed at eliciting their possible responses to a modification of their risk status showed that the vast majority predicted they would feel depression or anxiety or both, and up to 15% indicated that they would be at risk for suicide following an increased risk result.[3 5 6] Despite this focus on potential adverse responses to an increased risk result, several authors[3 4 7] have suggested that adverse reactions might be as much of a concern for those who received a decreased risk. It was suggested that at risk people who have held a long standing belief that they would some day develop HD might experience difficulties in coping with a decreased risk result, as would those who had already made major life decisions based on the belief that some day they would develop the disease.[3 4 7]

The Canadian Collaborative Study of Predictive Testing (CCSPT) for Huntington's Disease (HD) was initiated to evaluate the psychological and social effects of predictive testing. It began as a pilot project in British Columbia in 1986, and expanded to include 14 genetic centres across Canada in 1988. Linkage DNA testing was conducted from the programme's inception to the autumn of 1993 when the direct CAG repeat test became the primary means of informing people of their HD risk status.

At risk subjects participating in the Canadian study undergo an extensive protocol, including counselling sessions before receiving results, and follow up sessions at 10 days, six months, and one year after receiving results.

The fact that the Canadian study is prospective in nature, with a large number of participants and a protocol that includes extensive psychosocial assessment of participants before and after results, provided an opportunity to examine the frequency and nature of adverse reactions in a cohort of at risk subjects, some of whom have received a modification of risk. The present paper examines in detail the short term adverse responses of subjects in the CCSPT who had been followed for at least one year after receiving results.

Material and methods

PROCEDURES

Any person 18 years of age or older who had a parent with a confirmed diagnosis of HD and who voluntarily requested predictive testing was eligible for inclusion. All of the participants in the study received pre- and post-test counselling. Subjects who chose to have the predictive test also completed a comprehensive battery of psychosocial questionnaires before and at seven to 10 days, six months, and 12 months after receiving their result. Those who participated in the study but who chose not to have the predictive test completed the questionnaires three times, at entry and at six and 12 months from baseline. Additional details concerning the study protocol have been described elsewhere.[8] It should be noted that during their first assessment session in the study, all participants also completed questionnaires that were designed to obtain demographic, family, and personal psychiatric history data.

DEFINITION OF AN ADVERSE EVENT

Clinical and quantitative criteria were used to identify subjects in the study cohort who had experienced serious problems during their participation in the predictive testing programme. They were defined as having experienced an adverse event if they met the clinical, the quantitative, or both criteria.

The clinical criteria used to define an adverse event were any one of the following: a suicide attempt or the formulation of a suicide plan; psychiatric hospitalisation; depression lasting longer than two months; a marked increase in substance use; and the breakdown of important relationships. These criteria were developed through a two stage process. The geneticists, genetics counsellors, and the psychologist comprising the study team in British Columbia generated an initial list of potential criteria based on their own experiences with study participants, and on information obtained from psychiatric publications. This initial list of criteria was then circulated to members of the study team in each of the 14 participating centres in other parts of Canada. Comments, suggestions, additions, and deletions were reviewed and through a process of consensus building, the final list of criteria was developed. It was agreed that each of the clinical events on the list was serious and should be considered an adverse reaction even if it occurred in the absence of any other criterion.

An adverse event was also considered to have occurred if established quantitative criteria were met with respect to information obtained from two self-report measures: the General Severity Index (GSI) of the Symptom Checklist-90 (R) (SCL90(R)) and the Beck Depression Inventory (BDI). The GSI is an index of psychological adjustment, with scores above 62 reflecting symptomatic clinical distress. It has been shown to be highly sensitive to change in a wide variety of clinical and medical contexts, and has shown high levels of internal consistency and test-retest reliability.[9] The BDI is a 21 item self-report measure of the behavioural manifestations of depression with scores of 10 or greater reflecting at least mild clinical depression. This measure has been shown to be sensitive to varying degrees of depression and to changes in depressive symptomology over time. It has shown high levels of reliability and convergent validity with clinical diagnoses.[10]

To meet quantitative criteria for an adverse psychological event, subjects had to have recorded a measurable change in the scores obtained on both the SCL-90 and the BDI. A change in psychological distress was defined as an increase of more than one standard deviation on the GSI from before results to after results, or from one follow up session to the next. A change in depression was defined as an increase of at least four points on the BDI, but only if the increase raised the person's score above the clinically significant level of 10.

Each person's perceived number of social supports and their level of satisfaction with their social supports were also documented. The Social Support Questionnaire[11] was used for this purpose and was included in the study in order to ascertain any correlation between this variable and psychological responses to a modification in HD risk status.

THE ADVERSE EVENTS QUESTIONNAIRE

Once the clinical criteria defining an adverse event had been developed, a questionnaire was constructed and forwarded to all of the clinicians and counsellors in each of the participating testing centres. In this questionnaire, the clinicians and counsellors were asked to review the records pertaining to each of their clients and indicate whether they had experienced any of the clinical events considered to be a criterion for an adverse event. Clinicians were also asked to indicate when each event occurred (if it was before their client had entered the predictive testing study or at some specific time after receiving their test result), and whether they had information suggesting that the event was or was not directly attributable to the predictive test result. The questionnaire also asked clinicians to report any other incidents that might have been clinically significant, even if the incident did not fall under any of the specified clinical criteria.

STATISTICAL ANALYSES

Chi-square tests (or where applicable the Fisher's exact test) were used to examine the associations between the incidence of adverse

Table 1 Baseline demographic characteristics by risk group

Characteristic	Study group: All participants (n=135) (%)	Increased risk (n=37) (%)	Decreased risk (n=58) (%)	No change in risk (n=17) (%)	Not tested (n=23) (%)	p value
Age, mean [SD]	37.5 [10.3]	35.6 [9.2]	41.0 [11.2]	36.5 [9.8]	32.2 [6.4]	0.002*
Sex						
Male	45 (33)	11 (30)	20 (35)	8 (47)	6 (26)	
Female	90 (67)	26 (70)	38 (65)	9 (53)	17 (74)	0.53
Education (highest grade completed)						
<12	28 (21)	6 (16)	12 (21)	4 (24)	6 (26)	
12	36 (27)	12 (33)	15 (26)	4 (24)	5 (22)	
>12	71 (53)	19 (51)	31 (53)	9 (53)	12 (52)	0.96
Type of employment						
Professional	38 (28)	6 (17)	20 (35)	8 (47)	5 (22)	
Trade or clerical	61 (46)	23 (64)	24 (41)	9 (53)	16 (70)	
None	36 (27)	7 (19)	14 (24)	0 (0)	2 (8)	0.64
Marital status						
Married or equiv	98 (73)	28 (76)	45 (78)	7 (41)	8 (15)	
Single	37 (27)	9 (24)	13 (22)	10 (59)	15 (65)	0.37

Pairwise comparison of group with respect to age
Increased risk *v* decreased risk p=0.09
Increased risk *v* uninformative p=0.96
Increased risk *v* untested p=0.19
Decreased risk *v* uninformative p=0.38
Decreased risk *v* untested p=0.002
Uninformative *v* untested p=0.39

Table 2 Baseline demographic and psychosocial variables in persons having or not having an adverse event

Characteristic	All participants (n = 135)	Study group: No adverse event (n=115)	Adverse event (n=20)	p value
	No (%)	No (%)	No (%)	
Age (y)	37.5 (10.3)	37.9 (10.6)	34.9 (7.8)	0.14
Sex				
Male	45.0 (33.0)	39.0 (34.0)	6.0 (30.0)	0.93
Female	90.0 (67.0)	76.0 (66.0)	14.0 (70.0)	
Marital status				
Single	25.0 (18.0)	22.0 (19.0)	3.0 (15.0)	0.56
Married	98.0 (73.0)	84.0 (73.0)	14.0 (70.0)	
Divorced/widowed	12.0 (9.0)	9.0 (8.0)	3.0 (15.0)	
Education				
<Grade 12	28.0 (21.0)	24.0 (21.0)	4.0 (20.0)	0.39
Grade 12	36.0 (27.0)	33.0 (29.0)	3.0 (15.0)	
>Grade 12	71.0 (52.0)	58.0 (50.0)	13.0 (65.0)	
Psychiatric history				
No history	99.0 (26.0)	87.0 (77.0)	12.0 (60.0)	0.18
Previous problems	34.0 (74.0)	26.0 (23.0)	8.0 (40.0)	
Beck depression I	4.7 (5.3)	4.4 (5.3)	6.6 (5.4)	0.04
Median	3.00	3.00	5.00	0.02
		Mean (SD)	Mean (SD)	
Global severity index	52.4 (10.8)	52.2 (10.7)	53.6 (11.4)	0.61
Social support number	4.2 (2.2)	4.2 (2.2)	3.9 (1.8)	0.41
Satisfaction with social supports	5.4 (0.78)	5.4 (0.81)	5.5 (0.62)	0.56
No of years since finding out about being at risk for HD	12.5 (8.4)	12.6 (8.6)	12.4 (7.6)	0.94

events and sex, marital status, educational level, presence of past psychiatric problems, and group membership in the CCSPT (that is, increased risk, decreased risk, uninformative, or not tested).

T tests or Mann-Whitney U tests were used to assess baseline differences between the "adverse event" and "no adverse event" groups on age, general distress, depression, and social supports.

Results

A total of 135 subjects met the eligibility criteria and were included in the study. Of these, 37 (27.4%) received an increased risk, 58 (43.0%) received a decreased risk, 17 (12.6%) were unable to receive a modification of risk, and 23 (17.0%) elected not to receive a result. The mean age of the cohort was 37.5 years (range 20-68). There were 45 males (33.3%) and 90 females (67%) with 73% of the sample being married. The demographic characteristics of the different risk groups were not different at baseline with regard to sex, education, employment, or marital status (table 1). There were four different visits to the genetic counselling centre during which questionnaires were completed. A total of 116 persons (86%) completed all the follow ups or missed only one. A total of 110 persons completed questionnaires at the one year follow up. Of the 19 (14%) who missed two or more follow ups, the majority were in the group that chose not to be tested (eight or 35% of the total persons in this category) with six in the uninformative group (35%), five in the increased risk group (14%), and one (2%) in the decreased risk group.

A total of 20 (14.8%) of the 135 people in the cohort were found to have met the

Table 3 Frequency of adverse events in persons across different study groups

	Study groups				
	Increased risk (n=37)	*Decreased risk (n=58)*	*No change in risk (n=17)*	*Chose not to be tested (n=23)*	
	No (%)	*No (%)*	*No (%)*	*No (%)*	*p value*
Adverse event	7 (18.9)	8 (13.8)	1 (5.9)	4 (17.4)	
No adverse event	30 (81.1)	50 (86.2)	16 (94.1)	19 (82.6)	0.63

identified criteria for an adverse event. Of these, 10 met both the clinical and quantitative criteria, four (20%) met the clinical criteria only, and six (30%) met only the quantitative criteria. Thus, in total, 14 (70.0%) subjects in the "adverse event" group were identified by clinical criteria, either alone or in conjunction with the quantitative criteria. Four of these (29%) reported suicidal ideation to the point of forming a suicide plan, six (43%) reported depression lasting longer than two months, one (7%) experienced a marked increase in substance use, and three (21%) participants underwent a breakdown in significant relationships.

A total of 16 of the 20 persons classified as having an adverse event had changes using quantitative criteria between pre- and post-predictive testing scores that placed them in this category. Two persons at decreased risk, one at increased risk, and one person not tested had changes on the GSI or the BDI between two post-test assessments that was defined a priori as an adverse event.

In order to assess factors that may be associated with the occurrence of an adverse event, the 20 people who experienced problems were compared to the remaining people in the study cohort on a number of demographic and psychosocial variables. As seen in table 2, the "adverse event" and "no adverse event" groups did not differ with respect to age, sex, marital status, education, psychiatric history, general psychiatric distress, social support, or the mean number of years since finding out about being at risk for HD. A significant difference ($p<0.04$) was found, however, when baseline level of depression (as measured by the BDI) was compared. Those in the "adverse event" group had scored higher on the BDI at baseline (mean=6.6) than the "no adverse event" group (mean=4.4). Even though, the analysis did not show statistical significance ($p<0.05$), there was a trend towards a lower age and higher frequency of previous psychiatric problems in the group of persons experiencing an adverse event compared to those who did not.

Table 3 shows the distribution of the occurrence of adverse events across predictive testing study groups. Seven of the 37 (18.9%) people receiving an increased risk result experienced an adverse event, as did eight of the 58 (13.8) receiving a decreased risk, one of the 17 (5.9) unable to receive a modification in their risk status, and four of the 19 (17.4) choosing not to be tested. The differences between the rates of adverse events in each of these four study groups were not found to be statistically significant ($p=0.63$). Moreover, although the numbers are too small to permit statistical comparison, no obvious patterns emerged with respect to the way in which the different types of adverse events were distributed across the four groups (table 4).

In the increased risk group, a total of 13 adverse events were seen in seven persons. Similarly in the decreased risk group, 12 events were seen in eight persons and in the group of persons who chose not to be tested, eight adverse events were seen in four persons.

An examination of the timing of adverse events showed a distinct pattern for both the increased and decreased risk groups. When the entire cohort was examined, it was found that 16 people (80%) experienced adverse events before the six month follow up (eight occurring between test results and the 10 day follow up, and eight occurring between the 10 day and six month follow up). The remaining four adverse events occurred between the six month and one year follow up. Interestingly, when the increased and decreased risk groups were investigated separately, it was found that for all those in the increased risk group (n=7) the adverse events started within 10 days following the results, whereas for all those receiving a decreased risk (n=8) the occurrence of problems occurred six months after receipt of results or later. This finding is significant ($p=0.0002$) using Fisher's exact test.

Discussion

Previous research has concluded that, for the most part, people derive more psychological benefits than harm from receiving a modification of their HD risk status through predictive testing, regardless of the direction of the test result.[12-17] This may in part indicate that persons choosing to participate in predictive

Table 4 Frequency and type of adverse events by study group

	Study groups			
	Increased risk	*Decreased risk*	*No change in risk*	*Chose not to be tested*
	No (%)	*No (%)*	*No (%)*	*No (%)*
No of persons with adverse event	7 (18.9)	8 (13.8)	1 (5.9)	4 (17.4)
No of adverse events	13	12	1	8
	Type of adverse event			
Suicide plan	1	1	1	1
Psychiatric hospitalisation	1	0	0	0
Depression (>2 months)	4	3	0	2
Relationship breakdown	2	0	0	1
Substance abuse	0	1	0	1
Quantitative criteria	5	7	0	3

testing are self-selected,[17] and perhaps less psychologically vulnerable than the general at risk population. This finding strongly reinforces the importance of autonomy and removal of undue influence by others as a factor in promoting a more favourable outcome in predictive testing. However, a small proportion of persons who receive test results do experience significant psychological distress.[7 13] A precise estimate of the frequency and type of adverse psychological events in a prospectively followed cohort has not been reported.

Here we report in detail on adverse events occurring during the first year after receiving results of predictive testing. Of the 135 participants, 20 (15%) experienced adverse events, as defined by clinical or quantitative criteria or both. No differences in age, gender composition, education, or marital status at baseline were found between the group of people who experienced an adverse event and the group who did not. Also, no group differences were found at baseline regarding global psychological distress or the number of, or satisfaction with, social supports. However, the "adverse event" group was found to have higher baseline depression scores than the "no adverse event" group. Although the mean on the BDI (6.6) for this group was still well below 10 (the criterion for mild clinical depression), the higher mean suggests that this group may be more vulnerable upon entry into predictive testing than are those who do not subsequently experience adverse events.

Further investigation found that 19 (14.1%) of the 135 people in the study scored greater than 10 (range=10 to 33) on their initial BDI (the "depressed" group). Of these 19, six (31.6%) had an adverse event during their one year follow up. By comparison, of the 116 people in the study who scored less than 10 on the BDI (the "not depressed" group), only 14 (12.1%) had an adverse event during follow up. The difference in the rates of adverse events between the "depressed" and "not depressed" groups is approaching statistical significance ($p=0.06$) and suggests that those who exhibit clinical symptoms of depression at entry into predictive testing (as measured by the BDI) may be at greater risk of adverse events than those who score in the normal range. It also suggests that the BDI, which is very rapidly completed (less than five minutes), may be a useful tool for identifying persons who are likely to have an adverse event and, therefore, are perhaps in need of additional clinical vigilance and support.

The likelihood of an adverse event was not associated with the direction of risk modification. Adverse events occurred in persons receiving both increased risk and decreased risk results. Thus, the emotional difficulties were not limited only to those who received an increased risk result.[3 4] People who received a decreased risk result experienced adjustment problems in the present study with a frequency similar to those who received an increased risk result.[7 14] We have previously reported[7] that the people most likely to experience difficulties adjusting to a decreased risk result are those who entered the programme believing they would receive an increased risk result, those who have made irreversible decisions, and those susceptible to survivor guilt. This study focused on the direct participants in predictive testing. The impact of predictive testing on family members has not fully been explored. However, preliminary results suggest that an increased risk result may result in significantly increased marital distress compared to couples receiving a decreased risk.[18]

Those receiving increased risk did not differ with regards to type of adverse event experienced compared to persons receiving a decreased risk result. However, the timing of adverse events differed significantly between the groups. Seven people who received an increased risk result experienced the onset of difficulties within 10 days following the disclosure of results. In contrast, eight participants with a decreased risk experienced their adverse event at or after six months from the time of receiving their test result. These findings suggest, therefore, that the timing, but not the nature or rate, of adverse events may be influenced by the direction of risk modification.

A similar pattern has been noted previously[14] where feelings of pessimism were evident after increased risk results and declined at the six month follow up, whereas pessimism declined sharply immediately following disclosure of a decreased risk result, only to emerge again for some at a later time. These findings suggest that clinical follow up for predictive testing for HD should not be limited to those who receive an increased risk result. The present study reinforces the need for long term clinical monitoring and support after a modification of risk status for HD. The present study only examined the occurrence of adverse events during the first year following results. However, it is possible that some people at increased risk may begin to experience a greater frequency of adverse events closer to their expected age of disease onset.

The results of the present study showed that psychological difficulties were experienced by some people across all categories of risk status, including those who chose not to be tested. The finding that participants who elected not to be tested experienced a similar rate of significant psychological distress may at first appear surprising. However, it could simply indicate that predictive testing is not associated with any significant increase in adverse events beyond that expected in the general at risk population. This would assume, however, that the group of persons choosing to be tested are similar in psychological profile to those choosing not to be tested. There is evidence to suggest that those who choose to be tested may be a self-selected group of people, confident in their ability to cope with the test results, well educated, and with more social supports than is seen in the general population.[17] If this is so, then the frequency of adverse events following predictive testing might begin to increase, particularly if a less self-selected group of people with less social support and a higher frequency of depression at baseline present for testing.

It is therefore not certain that the group of people who chose not to be tested, even though they participated in the research programme, are representative of the general at risk population who never make contact with predictive testing programmes. It is also possible that people who choose to participate in the research but not in the testing do so because they feel they will derive some psychological benefit from the counselling sessions. These persons may feel psychologically vulnerable and believe that participation in the research project will be one way to gain access to support. Certainly it has previously been suggested[5 19 20] that some people are not confident of their ability to cope with an increased risk result and will choose not to be tested,[5 21-23] but may participate in research studies. Therefore, the similar frequency of adverse events in the untested group compared to the increased and decreased risk groups could still be evidence for an increased adverse event rate after receiving results in predictive testing.

Alternatively, the similar rates of frequency and intensity of adverse events across the risk categories may simply represent base rates of psychological difficulties over a one year period in persons at risk for HD. The point prevalence of self-reported depressive symptoms has been stated to be from between 13% to 20% within the general population and the incidence rate of clinical non-bipolar depression to be as high as almost 8% per year.[24] Given these figures, the incidence rate of 15% for the types of difficulties evident in our sample may correspond to that of the general population, or the population of those living at risk for HD over a given year. Owing to the correlational design of the present study, the occurrence of adverse events cannot be related in a causal way to the experience of predictive testing. This paper reports on the incidence, not the causes, of adverse psychological events during one year of implementation of a predictive testing programme

However, based on the present research and past findings,[12-14] it can be concluded that the frequency and intensity of psychological difficulties occurring during predictive testing programmes appear to be less than was feared before their implementation. With current levels of psychological support, catastrophic reactions to predictive testing for HD have not occurred in the first year after results. No suicide attempts were made by any of the participants, and the formulation of a suicide plan was quite infrequent, with one participant in each result category reporting that they had made such a plan. It is evident, however, that certain people will experience psychological stress and will need support. Those who are experiencing some depressive symptoms before testing might be particularly vulnerable to adverse reactions and should receive particular attention in counselling.

It must be noted that the present investigation used objective and clinical measures of the occurrence of an adverse event. Standardised measures of psychological distress and the clinical judgments of counsellors formed the basis by which people were classified as having experienced an adverse event. Future investigations must also take into account the subjective accounts of the predictive testing participants. It needs to be determined whether other people would categorise their experience as negative, even though the levels of distress did not reach the clinical levels as defined. In a previous study, however,[22] only a very small proportion (0.9% in the decreased risk group and 4.5% in the increased risk group) of predictive testing participants reported that predictive testing had decreased their quality of life. The majority of persons (68.7% in the increased risk group and 50.9% in the decreased risk group) noted that predictive testing had no effect on the quality of their lives. However, 26.9% of those receiving an increased risk and 48.1% of those receiving a decreased risk reported that the quality of their life had improved after predictive testing. These data, together with the results presented here, suggest that adverse events in the first year after predictive testing do occur, but that for the majority of people predictive testing either has no effect or may have a positive effect on their quality of life. Clearly, long term assessment and a better understanding of how predictive testing might improve quality of life would be helpful so that appropriate support and measures are in place to favour such an outcome.

Future research should also strive to gain subjective accounts from the participants regarding their perception of the process and the attributions they make regarding their emotional difficulties. Such qualitative research would provide valuable information about the experience of receiving a modification of risk for HD.

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