REVIEW ARTICLE

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Psychosocial Issues in Cancer Genetics

Current Status and Future Directions

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Diagnostic tests are now available that allow genetic testing for several types of cancer. The aim of genetic counseling and testing for cancer is to educate individuals about cancer risk and cancer prevention, which it is hoped will lead to a reduction in morbidity and mortality. However, at this relatively early stage in the development of genetic counseling and testing programs, information is needed on the psychosocial impact of such programs on both the individual counselee and his/her family. This paper reviews the findings obtained during the past decade on the uptake of genetic testing, reasons for undergoing genetic testing, and the impact of genetic counseling and testing on feelings of distress and guilt. Specific attention is paid to experiences with prophylactic mastectomy and oophorectomy and the effectiveness of the uptake of and satisfaction with these risk-reducing procedures. In addition, the possible impact of genetic testing on insurance, work and future plans is discussed. Suggestions are given for translating research findings into psychosocial services and future research efforts.

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Even before the announcement of the deciphering of the Human Genome in February, 2001 (1, 2), it was common knowledge that having a family history of certain types of cancer is one of the best predictors of developing the disease. However, it was not until the early 1990s that diagnostic tests became available to allow genetic testing for several types of cancer. As cancer genetic counseling and testing programs become more widely available, it is important to assess their psychological and social impact on individuals and families in order to provide the best professional psychosocial support possible (3). This paper reviews the empirical literature on the uptake of genetic testing, the impact of genetic counseling on feelings of distress and guilt, uptake and satisfaction with prophylactic mastectomy and oophorectomy, and the impact of genetic counseling on such psychosocial issues as work, insurance and future planning. The need for psychosocial support and directions for future research are also discussed. Most of the papers reviewed here were studies of individuals counseled for breast and ovarian cancer.

UPTAKE OF GENETIC TESTING IN VARIOUS COUNTRIES

Uptake rates vary between the different disorders for which counseling is requested, and are higher when there are effective ways of treating or preventing the condition. If little can be offered, most people do not want information about their risk status (4). For example, for Huntington's disease, a neurodegenerative disorder for which currently no treatment is available, the uptake of genetic testing is about 15% (5). For colorectal cancer, which can be treated effectively when diagnosed at an early stage, uptake is about 75% (6). The uptake of family members of breast cancer mutation carriers varies between studies and countries, ranging from 27% in France (7), 34% in Israel (8), 38% in pre-symptomatic individuals in The Netherlands (9) and between 43% and 80% (10–14) in the US. These differences in test uptake between countries may reflect cultural differences in attitudes toward genetic testing and risk-reducing surgeries (15), differences in the clinical facilities available for the management of familial cancer (16),

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differences in source of recruitment (e.g. via clinical population or registry population) (17), differing social consequences of genetic testing (4) (for example, for obtaining health and life insurance), and/or differences in study methodology (i.e. sampling strategies, sample composition, response rates, etc.).

RISK PERCEPTION AND REASONS FOR UNDERGOING GENETIC TESTING

In a review of studies investigating risk communication in genetic testing for cancer susceptibility (18), it was concluded that: a) most individuals with a family history of cancer (including those at low to moderate risk) overestimate their personal cancer risk; b) awareness and recall of the risk information of genetic testing is limited; c) decision-making about genetic testing is strongly influenced by perceptions of personal cancer risk and less so by actual risk based on genetic testing; d) perceptions of personal risk of cancer are resistant to standard education and counseling approaches; e) psychological distress and coping processes influence the processing of risk information and subsequent decision-making in genetic testing (e.g. cancer-related distress has been shown to motivate actual use of genetic testing in high-risk populations, whereas generalized distress and depressive symptoms have been associated with lower rates of test use); and (f) family influences play an important role in risk awareness and genetic testing decisions. Thus, it is not so much the objective risk of developing cancer, but the subjective, perceived risk that is most strongly related to test uptake and also to preventive actions such as risk-reducing surgery.

A study among Dutch counselees who attended a family cancer clinic revealed that the three most important motives for undergoing genetic testing were: to obtain certainty (67%), to be able to take preventive actions (61%) and to estimate the risk for their children (47%) (19). Similar results have been reported in several US-based studies (10, 20), although in the study by Lerman et al. (10), general planning for the future/family planning was also identified as a major reason to apply for genetic counseling.

IMPACT OF GENETIC COUNSELING ON PSYCHOLOGICAL DISTRESS AND FEELINGS OF GUILT

Genetic testing may result in many individuals being burdened by the knowledge that they have an increased probability of developing a life-threatening disease long before its likely onset, and options for managing this risk are currently limited (3). At the same time, genetic testing can be a relief to those individuals who are found not to have inherited the specific mutation associated with cancer in their family. What have we really learned about the individual psychological impact of genetic counseling for cancer during the past few years?

Marteau & Croyle (1998) (4) concluded from their review that the psychological impact depends more on pre-test expectations (e.g. if expectations are different from test results, the impact is higher), mood (pre-test mood is positively correlated with post-test mood) and social support (when absent, the impact of testing is higher) than on the results of the test itself. Adverse reactions to genetic testing are uncommon when testing is provided within a program that separates the offer of testing from the actual taking of a biological sample for the test, and that provides clear information and emotional support both before and after testing (4). Broadstock et al. (21) concluded that, on average, of the 15 papers they reviewed, none reported levels of increased distress (general and situational distress, anxiety and depression) in carriers or non-carriers at any point during the 12 months after testing. Both carriers and non-carriers showed decreased distress after testing; this was greater and more rapid in non-carriers. The DNA test result was rarely predictive of distress more than one month after testing. In about half of the 27 analyses included in the review, the best predictor of distress was pre-test emotional state. The main conclusion drawn from their review was that those undergoing predictive genetic testing do not experience adverse psychological consequences. This conclusion was supported by the results of a meta-analysis (22) in which it was shown that genetic counseling of women with a family history of breast cancer reduced anxiety levels and, in addition, improved the accuracy of perceived risk. However, because the results of most studies have been based on highly self-selected groups (e.g. study participants are usually Caucasian, highly-educated individuals, living in the US), the results may not be representative for all high-risk individuals.

In the genetic literature, there is considerable speculation about the existence of so-called 'survivor's guilt' (i.e. feelings of guilt that one has not (yet) developed cancer, while other relatives have) among persons who are found not to have gene mutations associated with heightened risk of disease (3, 23, 24). This speculation continues despite the fact that there is little empirical evidence to suggest that being informed that one does not have the mutation increases feelings of guilt. Kash et al. (25) reported that feelings of guilt were pervasive among women at high risk of developing breast cancer, but it is not clear from their study for what percentage of women this was the case. They reported that (for some cases) this guilt was related to feelings of inadequacy in helping other relatives who had had cancer. Other women felt guilty because they may have passed a gene on to their offspring. Many women felt guilty about their 'preoccupation' with the possibility of developing breast cancer in the future, given that they were currently healthy. In a study of relatives of ovarian cancer patients

(26), 25% of the women indicated that they would feel guilty if they were to test negative for a hereditary breast-ovarian cancer susceptibility gene. However, since this situation was hypothetical, it is not clear whether these feelings of guilt would actually be experienced.

EFFECTIVENESS OF RISK-REDUCING SURGERY

Women at high risk of developing breast/ovarian cancer may choose to undergo risk-reducing surgery, such as prophylactic mastectomy and/or oophorectomy. Several studies have investigated the efficacy of bilateral prophylactic mastectomy in women with a family history of breast cancer. Hartmann et al. (27) and Meijers-Heijboer et al. (28) reported a significant reduction in risk of breast cancer after bilateral prophylactic mastectomy among BRCA1/2 mutation carriers. The estimated risk of breast cancer was reduced by 85% to 100%. Rebbeck et al. (29, 30) and Kauff et al. (31) recently reported that, in women with BRCA1/2 mutations, bilateral prophylactic oophorectomy reduced the risk of ovarian cancer by 90% to 95%, and the risk of breast cancer by 50% to 70% (29, 32, 31). Despite this risk reduction for ovarian cancer, the occurrence of peritoneal carcinomatosis after prophylactic oophorectomy surgery is still a possibility (32, 33).

Although the reduction of the cancer risk by prophylactic surgery is obviously important, both prophylactic mastectomy and oophorectomy are mutilating and irreversible interventions which may affect body image and sexuality. An alternative option for the management of breast and ovarian cancer risk is frequent surveillance. However, periodic breast screening has its limitations. The interpretation of mammograms in pre-menopausal women is less reliable, and the frequently repeated controls may be experienced as stressful. In the case of ovarian cancer surveillance, the currently available screening tests have relatively poor sensitivity and specificity and, again, the stress experienced when undergoing gynecological controls can be substantial. Little is currently known about the efficacy of surveillance programs in these high-risk women (34, 35).

UPTAKE OF RISK-REDUCING SURGERY

The percentage of women at high risk of developing breast/ovarian cancer who actually undergo prophylactic surgical procedures varies per country and hospital. This variability reflects the cultural context in which physicians and patients make decisions (36, 37). For example, a cross-cultural study on women's attitudes toward preventive strategies showed that British women are more in favor of prophylactic mastectomy and oophorectomy than French women (15).

To our knowledge, there are very few reports on the proportion of women who are carriers of a BRCA1/2 mutation, who actually have undergone prophylactic surgery (Table 1). In the US, Lerman et al. (38) studied 29 female BRCA1/2 carriers eligible for bilateral prophylactic mastectomy and found that only one of these women (3%) actually underwent this surgical procedure within the 12 months following BRCA1/2 testing. Of the 39 asymptomatic mutation carriers eligible for prophylactic oophorectomy, 5 women (13%) underwent this intervention within the one-year follow-up period. Of the female carriers who did not undergo prophylactic oophorectomy, only 21% reported surveillance of the ovaries.

In a larger US study by Lynch et al. (39), 19% of the 142 BRCA1/2 mutation-positive women had undergone bilateral prophylactic mastectomy, and 35% had undergone bilateral prophylactic oophorectomy.

In a Dutch study (9) of 68 female carriers of a BRCA1/2 mutation, 51% ($n = 35$) opted for prophylactic bilateral mastectomy. In addition, 29 (64%) of 45 asymptomatic mutation carriers, aged 35 years and older, opted for this procedure; the majority (83%) underwent the surgery within 9 months after DNA test disclosure. A relatively older age (40–54 years vs. < 40 years) was found to be a significant predictor of opting to undergo prophylactic oophorectomy.

In a UK study by Hatcher et al. (40), 154 women with an increased risk of developing breast cancer were offered bilateral prophylactic mastectomy. In contrast to the other studies reviewed, most of the women did not know whether they were carriers of a BRCA-gene mutation or not. However, all of these women had a sufficient number of high-risk factors to justify the option of bilateral prophylactic mastectomy. Of these women, 11 (7%) deferred making a decision, 79 (51%) chose to undergo the surgery and 64 (42%) declined. These percentages of uptake of prophylactic mastectomy are similar to those reported in the Dutch study (that included unaffected BRCA1/2 carriers only), but much higher than those reported in US studies (including BRCA1/2 carriers).

SATISFACTION AND REGRET FOLLOWING PROPHYLACTIC MASTECTOMY

What is the impact of risk-reducing surgery, such as prophylactic mastectomy, on quality of life? To what extent do women who undergo this operation feel relief or regret? Do worries about developing breast cancer decrease and, if so, does that weigh against possible changes in body image and sexuality? The results of studies addressing these issues are summarized in Table 2.

In 1995 Stefanek et al. (41) reported on 14 women at increased risk of breast cancer who underwent bilateral prophylactic mastectomy. Levels of depression were within the non-patient normative range. All 14 women were satisfied with their decision. In addition, 12 of these women declared they would recommend preventive surgery to other women at comparable risk, while the other 2 indicated that this was a decision that was too personal to recommend. Of

Table 1

Uptake of prophylactic mastectomy and oophorectomy

Reference	Country	Study population	Uptake of preventive option	Time-frame
Lerman et al. (38)	US	n = 29/39 unaffected BRCA1/2 carriers	3% mastectomy 13% oophorectomy 21% ovarian screening	Within 12 months after BRCA1/2 test
Lynch et al. (39)	US	n = 142 BRCA1/2 carriers	19% bilateral mastectomy. 35% bilateral oophorectomy	Unclear
Meijers-Heijboer et al. (9)	The Netherlands	n = 68/45 unaffected BRCA1/2 carriers, eligible for surgery	51% mastectomy (35/68) 64% oophorectomy (29/45)	Median follow-up = 26 months (range 16–62 months), 83% within 9 months' follow-up
Hatcher et al. (40)	UK	n = 154 at increased risk of developing breast cancer, participating in psychological study	51% mastectomy	Unclear

the 11 women who opted for bilateral breast reconstruction, 3 were dissatisfied because of the cosmetic results that turned out 'worse than expected', and/or because of tissue rejection and subsequent infections.

Borgen et al. (42) studied regrets after bilateral prophylactic mastectomy in a volunteer sample (i.e. readers of several lay journals) of 370 women who had undergone this procedure, on average, 15 years earlier. Breast reconstruction was performed in 75% of all cases. Five percent (n = 21) of the women expressed regrets over the prophylactic mastectomy. These regrets were most likely to occur when the discussion about prophylactic mastectomy was initiated by the physician rather than by the woman herself. These results should be viewed with some caution owing to possible sampling bias. Participants in the study were recruited through lay journals (e.g. a publication of the American Association of Retired Persons). It is often the case that individuals who respond to surveys publicized via the lay press often have extreme views. In this case, those women who responded to the survey may have been either

Table 2

Experiences with prophylactic mastectomy

Reference	Study population	Study design	Experiences
Stefanek et al. (41)	US, n = 164 ♀♀ at risk, n = 14 having bilateral prophylactic mastectomy	Questionnaire Descriptive study Follow-up 9 months after mastectomy, (range 6–30 months)	Satisfied with decision to have bilateral prophylactic mastectomy Mixed feelings about satisfaction with breast reconstruction
Borgen et al. (42)	US, n = 370 ♀♀ of volunteer population	Questionnaire Descriptive study—on average 15 years after mastectomy	5% expressed regrets, Regrets correlate with initiation of discussion about previous surgery by clinician rather than woman herself
Hopwood et al. (43)	UK, n = 52 ♀♀ questionnaire/n = 45 ♀♀ interview (6 carriers, 3 with breast cancer)	Questionnaire & interview Follow-up study in 1st 3 years following surgery, most data relate to 1st follow-up (10 months after mastectomy)	Frequency of minor changes in: Sexual attractiveness (55%) Feeling less physically attractive (53%), Self-consciousness about appearance (53%), Decreased satisfaction with body (47%) Feeling less feminine (35%)
Frost et al. (44)	US, n = 572 ♀♀ at risk, who underwent bilateral prophylactic mastectomy between 1960 and 1993	Questionnaire Descriptive study, on average14.5 years after mastectomy	70% satisfied with procedure, 11% neutral, 19% dissatisfied 74% diminished level of emotional concern about developing breast cancer No change or favorable effects in level of: emotional stability (91%), stress (86%), self-esteem (82%), sexual relationships (77%), feelings of femininity (75%), satisfaction with body appearance (64%)
Hatcher et al. (40)	UK, n = 143 ♀♀ at increased risk, n = 79 mastectomy n = 64 no mastectomy	Questionnaire & interview Follow-up study: preoperatively, 6 and 18 months postoperatively	Psychological morbidity declined in the surgery group, but not in the no-surgery group Level of sexual discomfort and degree of sexual pleasure unchanged over time in both groups

very satisfied or very dissatisfied with their preventive mastectomy.

Hopwood et al. (43) followed 49 women during the first three years after bilateral prophylactic mastectomy. For the majority of these women there was no evidence of significant mental health or body-image problems. Of all women, 21% reported no negative change in body image during the first year following surgery. Minor changes were reported in sexual attractiveness (55% of the respondents), feeling less physically attractive (53%), change in self-consciousness about appearance (53%), decreased satisfaction with body (47%), and feeling less feminine (35%). A minority (approximately 15%) reported more serious psychological problems or body-image concerns, which were usually related to surgical complications.

In a larger study (44), 572 women with a family history of breast cancer who had elected to undergo bilateral prophylactic mastectomy between 1960 and 1993 completed a questionnaire, on average 14.5 years later. Of all these women, 70% were satisfied with the procedure, 11% neutral and 19% dissatisfied. Of the women who were not satisfied with the prophylactic mastectomy, the most frequently reported adverse symptoms or complications included: implant concerns, adverse body image, and insufficient information or support. Most women (74%) reported decreased emotional concern about developing breast cancer. No change, or favorable effects were reported for the levels of: emotional stability (91%), stress (86%), self-esteem (82%), sexual relationships (77%), feelings of femininity (75%) and satisfaction with body appearance (64%). The majority (67%) indicated that they definitely or probably would choose the procedure again, 15% were unsure, and 18% would probably or definitely not. The women who were (very) satisfied usually cited peace of mind, good health since the operation or no problems with the procedure, satisfaction with body image and risk reduction or enhanced detection of cancer. Women who were (very) dissatisfied with prophylactic mastectomy usually reported adverse symptoms or complications including implant concerns, adverse body image and insufficient information or support.

In the prospective study by Hatcher et al. (40) (see above), the 79 women who opted for bilateral prophylactic mastectomy reported a significant decrease in psychological morbidity assessed by the General Health Questionnaire 18 months after surgery. This was not the case for the 64 women who declined surgery. In contrast to Hopwood et al. (43), the level of sexual discomfort and degree of sexual pleasure did not change significantly over time in either of the two groups.

In summary, the negative psychosocial impact of bilateral prophylactic mastectomy seems to be small: levels of distress are the same or decreased after surgery, and most women reported being satisfied with their decision, despite the dissatisfaction of some with the reconstruction (see Table 2).

SATISFACTION AND REGRET FOLLOWING PROPHYLACTIC OOPHORECTOMY

In counseling women about the options of prophylactic oophorectomy or ovarian cancer surveillance, little is known about how women choose either option and how they later feel about their decision (45). Presently, only data from cross-sectional, mostly qualitative studies with relatively small sample sizes are available (see Table 3).

In an Australian study, Meiser et al. (46) interviewed 14 women who had undergone prophylactic oophorectomy 4 months to 7 years prior to the interview. All of these women had a family history consistent with a dominantly inherited susceptibility to ovarian cancer. At the time of prophylactic oophorectomy, 8 women were found to be carriers of BRCA1/2 mutations. Almost all of the women (13/14) were satisfied with their decision to undergo this preventive surgery. The oophorectomy decreased their anxiety about developing ovarian cancer. Premenopausal women reported unmet information needs about the effects of surgical menopause and the link between hormone replacement therapy and breast cancer.

Hallowell (47) conducted interviews with 23 women who had undergone prophylactic bilateral oophorectomy to manage their inherited risk of ovarian cancer. She found that the women experienced the benefits of risk reduction as outweighing the costs of surgery. All of the women reported feeling an overwhelming sense of relief following oophorectomy. However, with one exception, all reported experiencing either physical problems (e.g. postoperative complications) or emotional problems (i.e. mood swings, loss of libido, altered body image) following surgery. Most of the women claimed that these problems were exacerbated by a lack of knowledge about oophorectomy and its after effects. Women reported that they would have liked more information about ovarian function and menopause, hormone replacement therapy, surgical procedures, convalescence and the risk of inheriting a genetic mutation and developing cancer. Again, in this UK study, one of the conclusions was that there is an unmet need for information about the physical and emotional after effects of oophorectomy.

In a US study, Swisher et al. (45) compared 30 women who had undergone prophylactic oophorectomy (median age 47 years) with 30 women who had undergone ovarian cancer surveillance (median age 43 years). Almost half of the surgery group would have liked more information prior to the surgery. This comprised information about hormone replacement therapy, and general information about the procedure and the physical effects of the loss of their ovaries. Two women (7%) expressed regrets about their decision. One woman had regrets because she experienced

Table 3

Experiences with prophylactic oophorectomy

Reference	Study population	Study-design	Experiences
Meiser et al. (46)	Australia, n = 14 ♀♀ Family history of breast and/or ovarian cancer 8/14 are carriers of BRCA1/2, 8/14 are affected	Interviews 4 months–7 years after surgery	All but one were satisfied with decision to undergo oophorectomy Surgery decreased anxiety about developing ovarian cancer Postmenopausal women reported no negative impact on their libido Premenopausal women reported unmet need for information about effects of surgical menopause and link between HRT and breast cancer.
Hallowell (47)	UK, n = 23 ♀♀ with preventive oophorectomy High risk (i.e. > 2 relatives with ovarian cancer) Premenopausal No history of cancer Mean age at surgery 38.8 years	Interviews on average 5.5 years after surgery (range: 0.5–25 years)	Benefits outweigh the costs of risk-reducing surgery Most would have liked more information about physical and emotional effects prior to/following surgery More information was needed about ovarian function and menopause, HRT, surgical procedures, convalescence, risk of inheriting a genetic mutation and developing cancer
Swisher et al. (45)	US, n = 60 ♀♀ n = 30 preventive oophorectomy n = 30 screening	Telephone interview on average 33 months after surgery (range: 1–84)	2/30 (7%) of the oophorectomy group were dissatisfied with their decision, 93% were satisfied and reported no regrets 15/30 (50%) of the surveillance group expressed dissatisfaction or some regret or reservations about their choice
Fry et al. (48)	UK, n = 57 ♀♀ n = 29 preventive oophorectomy n = 28 screening	Questionnaire, 1–5 years after surgery	Surgical group poorer functioning on SF-36 Surgical group more menopausal symptoms No differences for cancer worries and sexuality Premenopausal ♀♀ more vulnerable to distress after oophorectomy than postmenopausal ♀♀
Hollenstein et al. (49)	The Netherlands, n = 29 ♀♀ n = 16 preventive oophorectomy n = 13 screening	Questionnaire, 1–5 years after surgery	Surgery group more satisfied with preventive choice than screening group Surgery group fewer cancer-related worries, less general distress (MHI-5), fewer intrusive thoughts about cancer in the family (IES)

memory loss and an inability to concentrate as a result of the anesthesia, and one woman had regrets when she realized that the surgery could not prevent her from developing peritoneal cancer, which had happened to her sister. However, most women undergoing prophylactic oophorectomy had no regrets about their decision. Also, most of the women (90%) who opted for screening were (for the moment) satisfied with their decision, although half of these women expressed some reservations about their choice and would consider prophylactic oophorectomy in the future.

In a study by Fry et al. (48), 29 women who had undergone prophylactic oophorectomy 1 to five years earlier were compared with 28 high-risk women who had opted for ovarian screening. No significant differences were found between the groups in terms of cancer worry or sexual functioning. However, the women who underwent surgery reported greater interference with work and social activities owing to physical or emotional problems, and more physical problems such as aches and pains, weight gain and headaches. The women who were premenopausal at the time of the oophorectomy had, on average, higher scores on the General Health Questionnaire (indicating higher levels of psychological distress) and took longer to recover postoperatively (5.4 months for the premenopausal women vs. 3.6 months for the postmenopausal women).

Hollenstein et al. (49) reported on 29 women at risk of ovarian cancer, of whom 16 had undergone prophylactic oophorectomy and 13 had opted for a surveillance program. The women who had the risk-reducing surgery were significantly more satisfied with their choice than those who underwent screening only. On average, the surgery group reported fewer cancer-related worries, and less general and specific distress as compared to the screening group.

In summary, most studies on prophylactic oophorectomy indicate that the majority of high-risk women who undergo preventive surgery are satisfied with their decision (Table 3). Although prophylactic oophorectomy brings with it some physical and psychological problems, for the majority of women, the benefits of the operation outweigh the costs. Furthermore, there is a widely expressed need for informa-

tion both pre- and post-surgery about the physical, sexual and emotional consequences of the procedure and about hormone replacement therapy and its relationship with breast cancer.

Again, it should be emphasized that the results discussed here are based on small studies with probably highly selective samples, and the associations observed have yet to be examined prospectively. In particular, there is a need for longitudinal studies to evaluate the extent to which oophorectomy allays patients' fear of cancer over a longer period of time.

IMPACT OF GENETIC COUNSELING ON INSURANCE, WORK AND FUTURE PLANS

Genetic counseling may have significant consequences in the areas of insurance, work and family planning (50). Most concerns about discrimination against carriers of genetic conditions revolve around insurance coverage (51). A common concern is that genetic counseling may be made mandatory for some insurance schemes (52) and that this may result in limited coverage or outright denial of insurance. In a US study (n = 384) (53), it was reported that approximately one-quarter of patients eligible for BRCA testing may decline because of concerns about cost, confidentiality and discrimination. However, these concerns may not reflect the actual situation in that no experiences with discrimination were reported. To our knowledge, no systematic studies of discriminatory practices have been carried out in the area of cancer genetics in European countries. Incidental problems with insurance companies in The Netherlands relating to a different hereditary syndrome (i.e. hypercholesterolemia) may indicate that problems with cancer counseling might be expected in the future (54). In this study, 37% of those who wanted to obtain a (life-) insurance, indicated having experienced problems, despite the fact that, in most cases, the amount requested to be insured was low, and their disease highly treatable.

With regard to work discrimination, Billings and colleagues (52) reported a range of problems experienced by asymptomatic individuals with a variety of genetic health conditions unrelated to cancer, including difficulty in obtaining employment, early termination, being passed over for promotion and mandatory job transfer. Lapham et al. (55) reported that 13% of a group of 332 members of a genetic support group (including 101 different genetic disorders) believed they were denied or released from a job; fear of genetic discrimination resulted in 17% of the respondents not revealing information to employers. To our knowledge, no information about work discrimination, specifically related to cancer genetics, has been reported in the literature.

Finally, in the area of family planning, Kash and colleagues (25) reported that many women counseled in a familial cancer clinic in New York and who were found to be at increased risk of developing breast cancer postponed marriage and/or decided not to have children. These women reported that they were certain that they would eventually develop breast cancer and would die of their disease, and thus chose to avoid the long-term commitments associated with marriage and child-bearing. However, the authors did not report the actual percentages of women who decided to postpone marriage or decided not to have children.

PSYCHOLOGICAL SUPPORT NEEDS

It is estimated that the percentage of individuals needing professional psychosocial support for genetic counseling ranges from 13 to 41 (25, 56–60). In some countries, family cancer clinics offer professional psychosocial support to asymptomatic women from HBOC families (61, 62). However, more data are needed to determine: 1) how much and what type of information and support (both pre- and post-testing) are required for those offered genetic testing and how this can be most efficiently provided and 2) how to achieve better understanding of genetic testing and its results, and how one can best facilitate risk-reducing behavior without high levels of emotional distress or false reassurance (4).

The literature provides some suggestions for recognizing those who might be most vulnerable to psychosocial problems as a result of genetic counseling (Table 4). First, support should be offered to counselees who may encounter problems in contacting their family members; they should anticipate that information about hereditary cancer is not always desired or appreciated. Recognition and support of the counselee who is the 'family messenger' and who is the 'first utilizer' of predictive testing and preventive surgery in the family is important, especially when this counselee feels that he/she has to set a 'good example' to the rest of the family (63).

Earlier studies on the diagnosis of (breast) cancer have shown that those individuals who experienced high levels of distress before diagnosis are at risk of experiencing high levels of distress after diagnosis (64, 65). This was also reported for couples: early difficulties and distress in couples facing breast cancer predicted later problems (66). Similarly, distress or depressive symptoms experienced before genetic testing are predictive of levels of distress after counseling (21, 67).

Results of a study on daughters of breast cancer patients suggest that the experience of cancer within the family, especially during adolescence (11–20 years), may have a significant impact on levels of distress (68). It was found that adolescent daughters of women with a poor survival prognosis were at greatest risk for adverse emotional outcome. These daughters were more likely to report long-term life-span changes and role changes with their mothers (68, 69). Erblich et al. (70) stress the importance of

Table 4

Professional psychosocial support needs in cancer genetics

Support might be needed in the following situations
When there are problems in contacting family members ('the messenger', 'first utilizer')
When a high level of distress before genetic counseling is observed
When a negative experience of cancer in the family occurred during adolescence (11–20 years)
When women experienced both the care-giving and death of their mother from cancer
When a relative recently died of cancer
When the counselee was recently diagnosed with (a recurrence of) cancer
When the counselee declines receipt of the genetic test results
When the genetic test result is not in accordance with the anticipated result
When risk-reducing surgery (e.g. mastectomy, oophorectomy, colectomy) is considered

care-giving by women whose mothers were dying of breast cancer. Care-giving was found to be predictive of cancer-related distress and depressive symptoms later in life. These results suggest that individuals who come for genetic counseling for cancer and who cared for and/or lost a parent during adolescence may be more vulnerable to psychological distress and may request additional psychosocial support during the genetic counseling process.

Another vulnerable group is those counselees who recently lost a close relative to cancer. They may postpone genetic counseling in order to gain time to cope with their loss and grief. Conversely, the recent occurrence of cancer or metastases in the counselee may lead to the wish to accelerate the predictive testing procedure (71). Among these latter counselees it is important to allow sufficient time for an informed decision to be made with regard to the genetic testing and/or preventive actions.

Special attention should be paid to those counselees who come for genetic testing but decline receipt of the test results; these persons may be at risk for depression. Lerman et al. (72) found that, in BRCA1/2-linked families, persons with high levels of cancer-related distress who decline genetic testing may be at risk for depression. These families may benefit from psychosocial support even if they prefer not to be tested.

Special care is needed in dealing with those families in which clear expectations exist about (non)carriership. Grosfeld et al. (73) found that, if the test outcome differs from what is anticipated, families have more difficulty in adapting. Anticipation is often based on assumptions of resemblance with an affected relative; physical resemblance as well as similarity in personality of a child with the affected parent may play an important role (74).

Finally, as indicated earlier, psychosocial support should be offered to those who have made a decision about undergoing risk-reducing surgery. Pre- as well as post-surgery support can help individuals in making a well-balanced decision about undergoing surgery, facilitate the adaptation to the operation and prevent feelings of regret.

DIRECTIONS FOR FUTURE RESEARCH

Since hereditary cancer syndromes are both an individual and a family issue, future studies should focus on the psychological adjustment of the individual in the context of the family in order fully to understand the adjustment to genetic counseling and testing for cancer syndromes (75). There is a need for valid and reliable questionnaires that assess the impact of an event such as cancer on family relationships. Although there are questionnaires that focus on the 'nuclear' family (father, mother and children), there is still a lack of measures of the relationship between the extended family members. Since every family differs in number, cohesiveness and importance of relationships, a standardized questionnaire may be a less optimal way to assess changes in family functioning than by using an interview.

An important topic for study concerns the various roles people play in the counseling process (63) and the developmental stage of family members (76). The psychosocial impact of genetic counseling and testing probably varies significantly from one developmental stage to another (e.g. a family with young children versus a retired couple). In addition, adjustment to the frequent occurrence of cancer in the family may also depend on the number of family resources and type of family-coping strategies (77).

Future studies should preferably use a prospective, longitudinal study design when determining the psychosocial effects of genetic counseling and testing. Psychosocial assessment before, and at several time-points during and after the counseling and testing procedure, can provide information about the changes in, for example, risk perception, knowledge, cancer worries and distress. Presently, few data are available on the impact of genetic counseling and testing in the long term (two or more years after disclosure of the genetic test results). Therefore, long-term follow-up studies are needed.

To assess distress in the members of families with hereditary cancer syndromes, we recommend the use of situation-specific questionnaires such as the Impact of Event Scale (IES) (78), or the cancer worries scale of Lerman et al. (26). More general measures of distress such as the State-Trait Anxiety Inventory (STAI) (79) and the Center for Epidemiologic Studies-Depression (CES-D) (80) do not appear to detect problems that are specific to cancer genetics.

To date, there is a paucity of literature addressing the psychosocial consequences of genetic counseling and DNA testing for those high-risk individuals that receive an *inconclusive* DNA test result. Since the most important reason to undergo genetic testing is 'to obtain certainty', it might be hypothesized that this group of individuals may be

particularly vulnerable to psychosocial difficulties. The psychosocial impact of genetic counseling and testing in these families merits further study.

More information is also needed on the impact of genetic counseling and testing on employment opportunities, job discrimination, insurance coverage and financial matters such as the ability to obtain long-term loans (e.g. home mortgages). Because such effects can depend on macro-level social and economic factors, studies in this area should preferably be of a cross-cultural nature.

Finally, as indicated earlier, research is needed to determine how much and what type of information and professional psychosocial support are required by genetic counselees, and how these can be provided most efficiently (4, 81). Since the provision of genetic information may not necessarily increase an individual's motivation to change his or her behavior (81, 82), effective interventions following the provision of risk information should be developed to maximize appropriate preventive actions and to minimize adverse psychosocial reactions.

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