Title: Sociodemographic, psychosocial and clinical factors associated with uptake of genetic counseling for hereditary cancer: a systematic review

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ABSTRACT

Evidence suggests that a significant proportion of individuals referred to cancer genetic counselling (GC) do not attend, and thus may not be engaged in adequate cancer risk management. We aimed to review the literature to better understand barriers to accessing GC and how they may be overcome. We conducted a systematic literature search for articles examining factors influencing cancer GC uptake as well as motivators and barriers to GC attendance. Factors were categorised as sociodemographic, psychosocial or clinical. The literature search identified 1,413 citations, 35 of which met the inclusion criteria. GC uptake ranged from 19% to 88%. With the exceptions of education level, socioeconomic status, cancer-specific distress, personal cancer diagnosis and actual and perceived risk of cancer, support was lacking for most sociodemographic, clinical and psychosocial factors as predictors of GC uptake. Cost and logistical barriers, emotional concerns, family concerns and low perceived personal relevance were reported as important considerations for those declining GC. We conclude that there is poor understanding of GC and a lack of decision support among those referred to GC. Research into ways of providing education and support to referred individuals will be important as the scope and availability of genetic counselling and testing broaden.

KEY WORDS

Barriers; Genetic counselling; Hereditary cancer; Systematic review; Uptake

INTRODUCTION

An estimated 10-24% of cancers are caused by a mutation in a known cancer susceptibility gene (1-3), with mutation carriers at significantly higher risk of cancer compared to the general population (4-6). Evidence-based strategies, including cancer screening, risk-reducing medication and risk-reducing surgery, are effective in reducing cancer-related mortality and morbidity in hereditary cancer syndromes (4-6). As such, identification of individuals at increased risk of cancer by genetic testing is important in order to provide adequate risk management to at-risk individuals and their relatives, as well as exclude non-carriers from intense cancer screening and unnecessary surgery.

Genetic counselling (GC) affords individuals at potentially increased risk of cancer the opportunity to make an informed decision regarding genetic testing and risk management. GC involves discussion of risk management options, including cancer screening and risk reduction. Emotional and decisional support are also provided, along with assistance with family communication about genetic risk. GC is considered essential for individuals undergoing genetic testing and has been shown to improve cancer risk perception without causing long-term psychological distress (7, 8).

Despite the benefits of both GC and genetic testing for hereditary cancer, genetic testing uptake among eligible individuals remains at about 50% (9, 10). There are fewer studies on uptake of GC amongst individuals who are referred to, or recommended to attend, GC as those declining to attend are difficult to recruit to research. There are also concerns that while those declining genetic testing may have made an informed decision through pre-test GC, individuals who decline GC may not have received sufficient information or support in the

decision-making process (11). In addition, genetics awareness and knowledge remain sub-optimal in the community (12, 13) and among health professionals as demonstrated by low rates referral for individuals eligible for counselling and/or testing (14-17). Utilization of genetics services is also lower in disadvantaged groups, such as those of low socioeconomic status and ethnic minorities (18, 19).

The aim of this review was to identify the factors and barriers associated with cancer GC uptake, in order to better understand the decision-making process and identify unmet information and support needs among individuals referred to GC.

METHODS

Literature search strategy

A literature search was conducted using the PubMed, Embase, PsychINFO and CINAHL databases. Search terms used were a combination of the following: genetic counselling, genetic risk assessment, cancer, neoplasm, uptake, attendance, participation, utilization and barriers, with a final search date of May 2015. The search was limited to original peer-reviewed articles published in English after 1994. This date was chosen as the *BRCA1* gene was isolated in 1994 and clinical genetic testing was offered from this point in time onwards. Additional relevant publications were identified by examining the reference lists of all articles accepted for inclusion and correspondence from field experts.

Inclusion and exclusion criteria

Quantitative and qualitative studies were eligible for inclusion if they addressed differences between those who attended GC and those who declined GC. Studies of GC withdrawal were

also included if withdrawal was prior to receiving personalized risk information. Articles grouping individuals who declined GC with those who declined genetic testing were included if the decliners group consisted mainly of counselling decliners. A small number of studies addressing barriers to GC uptake among decliners exclusively were identified and included as the additional data regarding reasons for non-uptake of GC were deemed relevant to the aims of this review to understand the barriers to attendance and identify future improvements to practice. Studies addressing any cancer type and female and/or male participants were included.

Studies were excluded if they only included individuals who had attended GC, or the decliners group was not an appropriate comparison group. Studies were also excluded if they measured GC uptake without analysing factors influencing uptake, assessed uptake of group GC or information sessions, did not adequately describe the referral status of the participants or compared different GC interventions/models of care.

Data extraction and quality assessment

Data extracted included the study characteristics as outlined in Table 1 and the independent variables and association (or lack thereof) with uptake and reasons for and against GC uptake. Variables measured by fewer than three studies were not included in the analysis.

Data quality was assessed using the following pre-defined quality criteria developed by the authors: clearly stated research aims, clearly defined study population, well described study design and data collection methods, clearly defined, validated and consistently implemented independent variables (if not validated then carefully designed and clearly described) , clearly defined and consistently implemented outcome variables and well described statistical

analysis with controlling for potential confounders. The data extraction and quality assessment was conducted by one reviewer (A.W.) and any study for which inclusion or data quality was unclear was discussed with up to two additional reviewers (S.S. and B.M).

[Insert Figure 1 about here]

RESULTS

Study characteristics

Thirty-five studies met the inclusion criteria and were included in the review (Figure 1). The details of the included studies are summarized in Table 1. Uptake was measured in different settings: standard referral practice (n=16), invitation to GC through a research study based on personal or familial risk factors (n=12) and invitation to GC based on the results of research genetic testing (n=7). GC uptake among referred individuals ranged from 19% to 88% across studies. A variety of research methodologies were used, including prospective cohort studies, cross-sectional studies, database/medical record review and qualitative methods.

[Insert Table 1 about here]

The results of the data quality assessment are outlined in Table S1. Few studies failed to meet one or more of the quality criteria and none of the studies meeting the inclusion criteria were deemed ineligible for inclusion on the basis of the quality assessment. Studies not taking cofounders into account in the analysis were generally unable to do so based on sample size.

Systematic review

A wide range of factors were measured in relation to GC uptake, which were categorized as either sociodemographic, psychosocial or clinical. Tables 2, 3 and 4 provide summaries of the associations observed across studies for each category. Each table is divided to show, from top to bottom: (i) factors where studies consistently indicate either a clear positive or negative association; (ii) factors where studies show a mixture of either positive *or* negative associations in combination with non-significant results, a situation that may arise if the effect sizes are moderate and sample sizes are limited; and (iii) factors where there is no consistent pattern of association. Studies with conflicting results for different measures of the same factor, or measures grouped under the same factor, are listed in each relevant table column.

Sociodemographic factors

The associations between sociodemographic factors and GC uptake for each study are summarized in Table 2 and described below.

Education level

Higher level of education was a significant predictor of GC uptake in six studies (20-25); one study observed a positive trend approaching significance (19); and no significant effect of education was observed in seven studies (26-32).

Socioeconomic status (SES)

Higher income was a significant predictor of uptake in two studies (24, 27), but was not associated in three (19, 23, 32). Being employed was significantly associated with uptake in two studies (22, 25), but was not associated in another five studies (20, 23, 27, 30, 31).

Higher social class (based on occupation) and lower social deprivation (based on postcode) were significantly associated with GC uptake (14, 33). Having health insurance was not associated with GC uptake (15, 20, 24).

Marital status

Being married was a significant predictor of GC uptake in three studies (24-26), and was not significantly associated in eight (19, 23, 28-33).

Age

Younger age was a significant predictor of *BRCA1/2* GC uptake in six studies (20, 24, 28, 31, 33, 34), and Lynch syndrome GC uptake in one study (35) and older age was a significant predictor of uptake in three studies of familial melanoma, *BRCA1/2* and Lynch syndrome (14, 30, 36).

Sex

Women were significantly more likely to attend GC than men in one study (38), with six others finding no significant association (14, 15, 25, 26, 34, 36).

Ethnicity

African-American ethnicity was strongly associated with lower uptake in one study (24). In another, Ashkenazi Jewish women were significantly more likely to attend GC compared to women of European, African-American and native American ancestry, although this effect was no longer statistically significant when controlling for education (21). Non-English speaking individuals were more likely to attend for all forms of familial colorectal cancer

(86% compared to 58% for English-speaking); however the sample size was small and statistical significance was not assessed (15).

Parenthood

One study found the number of offspring had a significant positive association with continuation of counselling (29), and another observed a trend toward higher uptake with parenthood (36). Having daughters was a significant predictor of GC uptake in one study (33); while another found that uptake was significantly lower among women who had daughters (22).

Summary

There were no sociodemographic factors displaying a clear positive or negative association with GC uptake. There was a pattern suggestive of a positive association for education level, SES and marital status, which may indicate that studies demonstrating no association were underpowered to detect differences between those accepting and declining GC. However, many of the associations observed were only significant at the bivariate level, either because confounders were not taken into account in the analysis or the association was no longer significant in multivariate analysis. In addition, SES measures differed across studies, with the effect of different measures within studies not always consistent (24, 27). Individuals attending GC were also more likely to be married in one study measuring household income, suggesting that marital status may have acted as a confounder (24). There was no consistent pattern of association observed for age, sex, ethnicity and parenthood. There was also no observable difference in associations for any socio-demographic factors according to the reasons for referral or the cancer status of study participants.

[Insert Table 2 about here]

Psychosocial factors

Table 3 provides a summary of the psychosocial factors measured in relation to GC uptake. Measures used varied across studies, with some studies incorporating multiple measures of the same factor.

Interest in genetic testing

Baseline interest in genetic testing was high (range 60%-74%) and was only measured for GC uptake in a research setting (19, 21, 26, 31). Research participants were significantly more likely to have expressed an interest in genetic testing in two studies (26, 31).

Cancer-specific distress

Five studies found a positive association between cancer-specific distress and GC uptake using the Impact of Events Scale (IES) (26, 30, 32), the Cancer Worry Scale (28) or purposively designed items (24). No difference in cancer-specific distress was observed between attenders and decliners in five studies, three using the IES (27, 28, 31), one using the Cancer Worry Scale (22) and another measuring “cancer-related concern” (19).

Perceived risk

A higher perceived risk of cancer was significantly associated with *BRCA1/2* (21, 22, 24) and melanoma (26) GC uptake in four studies. In the melanoma study, perceived risk was reported as a summary item that included perceived risk of developing a melanoma and that of having a mutation (26). One study found no association between perceived risk and GC

uptake, but did not specify whether perceived risk of cancer or of carrying a *BRCA1/2* mutation was measured (27).

A significant positive association between GC uptake and perceived risk of a *BRCA1/2* or *CDKN2A* mutation was observed in two studies (21, 26) and two studies observed a positive trend between GC uptake and *BRCA1/2* mutation risk (23, 31).

Knowledge

Men with lower knowledge scores were significantly less likely to attend GC (30).

Significantly lower knowledge scores regarding the genetics of cancer were observed among African-American women who declined GC, compared to those receiving GC, but no effect of general breast cancer knowledge was observed (32).

Perceived benefits and limitations

Despite GC being a process distinct from genetic testing, studies reported on the impact of perceived benefits and limitations of genetic counselling *and* testing, or of genetic testing only, on GC uptake. Out of six studies, greater endorsement of benefits was significantly associated with GC uptake for *BRCA1/2* in two studies (24, 31) and one study reported a trend towards significance in the same direction for melanoma (26).

Women who perceived fewer limitations of *BRCA1/2* genetic testing were significantly more likely to attend GC in two studies (19, 32); while another study observed a trend towards significance in the same direction (27). In contrast, one study found that women attending GC were more likely to have concerns regarding anxiety arising from genetic testing (24).

Anxiety, depression and general distress

The Hospital Anxiety and Depression Scale (HADS) was the most commonly used measure of general distress, but scores did not differ significantly between GC attenders and decliners (26, 29, 31). Other measures of general distress were the Mental Health Index-5 (28), the Profile of Mood States (27) and the Minnesota Multiphasic Personality Inventory-2 (MMPI2) (29). Only one study observed any association, with GC withdrawers having a more depressive affect compared to attenders (MMPI2), despite no difference observed in HADS scores in the same cohort (29).

Summary

A clear association with GC uptake was not observed for any of the psychosocial factors, although many displayed a pattern suggestive of a positive association. Perceived risk of cancer was most consistently associated with GC uptake, with two of six studies finding no association, but still reporting a positive trend. One of these studies performed no significance test (20) and the other had a sample size of 36, suggesting it may have been underpowered (27). A similar pattern was observed for cancer-specific distress, with studies finding no association having small sample sizes or including small numbers of GC decliners. There was also an overrepresentation of studies with cancer unaffected (or assumed unaffected) participants finding an association between perceived risk of cancer and cancer-specific distress and GC uptake.

The evidence for perceived risk of a mutation, intent to have genetic testing, knowledge and benefits of genetic testing was less convincing. There was also no notable difference between the sample sizes of studies that found an association and those that did not. A clear shortcoming of the available literature regarding psychosocial factors and GC uptake is

overrepresentation of hereditary breast/ovarian cancer cohorts and homogeneity of samples with regard to SES, ethnicity and sex. In addition, retrospective data collection in some studies means that participant responses may not be reflective of the time of actual decision-making.

[Insert Table 3 about here]

Clinical factors

Clinical factors were inconsistently measured across studies and the associations observed between clinical factors and GC uptake are summarized in Table 4.

Risk of carrying a mutation

Among known *BRCA1/2* mutation-carrying families, first-degree relatives of mutation carriers or probands (38, 39) were significantly more likely to attend GC, as were people at 50% risk or higher of carrying a *CDKN2A* mutation (26). Pre-test risk of a p-16 mutation came close to predicting counselling uptake (36). Two studies using risk assessment algorithms to estimate participants' risk of carrying a *BRCA1/2* mutation found that higher estimated risk of a mutation was positively associated with GC uptake (23, 24). Data on the impact of mutation risk on GC uptake for Lynch syndrome is notably lacking.

Personal history of cancer

Of the nine studies measuring the influence of a cancer diagnosis on GC uptake, seven were conducted in female-only cohorts affected by breast and/or gynaecological cancers (Table 4).

There was some evidence of an association, with cancer-affected individuals more likely to attend GC in five studies (26, 28, 29, 38, 39).

Family history of cancer

Having a family history of *BRCA1/2* or Lynch syndrome related cancers was significantly associated with GC uptake in only five studies (16, 20, 29, 30, 33). However, first-degree relatives of cancer-affected individuals were more likely to attend than distant relatives (39) and a higher number of relatives who died of melanoma was strongly associated with GC uptake (26), indicating that multiple aspects of family history may influence GC uptake.

Tumour characteristics

Tumour stage, histological subtype, tumour detection method, node status and histopathology were inconsistently measured and, for the most part, not associated with GC uptake (16, 19, 27, 31, 33, 37). The exceptions to this were: significantly higher uptake among women with a smaller tumour size in one study (33); and significantly higher uptake among women with serous ovarian cancer or stage III/IV cancer compared with other histological subtypes or stage I/II cancer in another (16).

Referral characteristics

Discussion of genetics referral or genetic testing with a health care provider was significantly associated with GC uptake in two studies (24, 40). An extended period between cancer treatment and referral led to lower GC uptake in one study (35); however there was no significant association in two other studies (27, 37). Breast cancer patients who indicated their referral was poorly timed in relation to their cancer treatment were significantly less

likely to attend GC in one study (31), though referral timing was not associated with uptake in another study (16).

Treatment characteristics

The association between treatment and GC uptake was only measured in *BRCA1/2* cohorts. Women undergoing chemotherapy were more likely to attend GC than those who did not have chemotherapy in one study (33). There was no evidence of an association between GC uptake and surgical treatment, chemotherapy, radiotherapy, hormone therapy or the time since diagnosis/stage of treatment in four studies (19, 27, 31, 33).

Summary

Objective risk of carrying a mutation displayed the most consistent association with GC uptake in this review. Associations observed for both personal and family cancer history were also suggestive of an association with GC uptake. A number of the studies finding no association also had fewer than 100 participants and/or enrolled participants based on a significant personal or family history of cancer, and thus may have been statistically underpowered to detect differences. The evidence for tumour and treatment characteristics is less consistent. Referral characteristics were infrequently measured and results were variable, with some evidence of an association between GC uptake and discussion of referral with the referring clinician. The majority of these studies were performed in cancer affected or mixed cohorts with no differences in associations observed regarding the cancer status or referral method for any of the clinical factors presented.

[Insert Table 4 about here]

Genetic counselling attendance

Some studies attempted to ascertain the reasons behind the decision to attend or not attend GC. The findings of these studies are described below; with particular attention paid to reasons for non-attendance.

Motivators for attendance

Motivators for GC attendance commonly endorsed or stated by attenders were: to obtain information for family members (11, 20, 31, 36, 41), to learn about their own risk of developing cancer (20, 31, 36), to have genetic testing (22, 36, 42) and/or to obtain a risk management plan (31, 36, 42). Other reasons given included: to help scientific research (22, 26, 36); to find out risks for offspring (28, 31, 36, 41); or being urged/encouraged to attend by relatives (20, 28, 42). Convenience also appeared to facilitate attendance at clinics, with insurance coverage, shorter distances to travel, available transport and convenient appointment times stated as additional reasons for attendance (11, 20).

Barriers to attendance

A variety of cost and logistical barriers were endorsed or stated across 15 studies. The financial cost of genetic testing and concerns around future insurability or financial discrimination were often reported (17, 26, 28, 35, 43-45). The time and travel required to attend an appointment were also stated as barriers (21, 26, 35, 37, 42, 43, 46, 47). Other logistical barriers that deterred people from attending were: GC not being recommended by a health professional (11, 20); organizational barriers (47, 48); or simply being unaware of the service or their eligibility for GC (11, 42, 47).

Existing emotional issues and concerns regarding the potential emotional impact of GC were reported as barriers to attendance in 13 studies. Participants reported emotional concerns such as anticipated worry or anxiety regarding the results of genetic testing (17, 26, 28, 31, 44, 46) and difficulty coping with the knowledge of an increased risk or need for screening (43, 44, 48). GC decliners also cited non-attendance due to feeling overwhelmed at the time of referral, either by their own cancer diagnosis and treatment, a relative's cancer diagnosis or other unrelated health problems or life stressors (11, 21, 25, 41, 43, 45).

Low perceived personal relevance was another barrier to attendance at GC, reported across 13 studies. Some participants simply stated they did not feel GC was relevant for them or they were not interested (21, 26, 28, 31, 44, 46, 48). Participants in five studies declined GC based on the expectation that it would not change their risk perception or behaviour (17, 21, 26, 43, 48) or because they did not have relatives for whom the information would be useful (31, 37). Others did not attend because they had decided *a priori* not to have genetic testing and precluded themselves from GC (26, 42, 45, 48). In two qualitative studies, women diagnosed with breast cancer and individuals at risk for Li-Fraumeni syndrome who received a personalised letter recommending they attend GC misinterpreted the letter as providing general information about GC rather than being an individual recommendation based on their personal circumstances (41, 45). Frequent reporting of low personal relevance is of particular concern in this context, given participants in these studies were eligible for genetic counselling, and often testing, based on their personal or family history of cancer.

Small numbers of decliners in 10 studies reported family-related concerns as contributing to their decision. These included discouragement or objection from family members (9, 11, 17,

31, 44, 46, 48); concerns about the possible emotional impact of GC on family members (11, 26, 28, 43, 48); the potential negative impact on family relationships (26, 28); and concerns regarding communication of genetic test results to family members (26, 48). Some had declined GC based on not wanting to involve their family members in the GC process when this was a requirement for testing (31, 48).

Summary

Studies used a variety of methods to elicit reasons for attendance/non-attendance at GC, including open-ended questions and closed-ended lists of possible reasons, which participants were asked to endorse. Thus, the data regarding reasons for attendance and non-attendance are a combination of both qualitative and quantitative data. A number of studies asked participants about their motivations retrospectively, leaving the findings of these studies open to recall bias. Also, only one of these studies focussed on individuals unaffected by cancer, making it difficult to determine whether the barriers to attendance are different for individuals with and without a prior cancer diagnosis. Despite these limitations and the variation in methods, findings were fairly consistent across studies with cost and logistical concerns, emotional concerns, perceived personal relevance and family-based concerns quoted or endorsed as reasons for non-attendance by a range of participants across different hereditary cancer syndromes.

DISCUSSION

Evidence regarding sociodemographic factors as predictors of GC uptake was inconsistent, reflective of research regarding predictors of genetic testing uptake (49). This makes drawing conclusions regarding the utility of sociodemographic factors as predictors of GC uptake

difficult, though it does highlight potential areas of disparity. Of note, there was evidence of higher education and SES among GC attenders in this review. This association has also been observed in studies comparing GC attenders to the general population (50, 51), which may indicate some disparity in access to genetic services. This finding was not necessarily surprising given health literacy, commonly defined as the “capacity to acquire, understand and use information in ways which promote and maintain good health” (52), in the community is sub-optimal and lower socioeconomic groups are disproportionately affected by low health literacy (53, 54).

While no consistent association between sex or ethnicity and GC uptake was observed in this review, these results are not consistent with the wider literature and may warrant closer scrutiny. Male non-participation in positive health behaviours is a widely recognized phenomenon (55). Thus, the lack of association between sex and GC uptake is more likely related to the number of studies including only female participants, and under-representation of males in mixed cohorts. There is also evidence in the wider literature of lower knowledge and uptake of genetic services in ethnic minority groups (18, 56). The lack of ethnic diversity in the included studies may have contributed to the findings of this review.

Associations observed for the psychosocial factors included in this review are consistent with theoretical models of health behaviour previously used in the cancer setting. For example, disease-specific distress is accepted as influencing health behaviours, whereas general distress is not. Perceived risk has also been used to predict a variety of health behaviours, with variable results. However, positive associations between perceived risk and other cancer-protective behaviours have been observed (57), consistent with these results for GC

uptake. The associations observed between GC uptake and personal/family history of cancer and objective risk are also in keeping with current knowledge, given their influence on perceived risk and cancer-specific distress (58). However, while an association between personal history of cancer and GC uptake was observed in mixed cohorts, unaffected cohorts were overrepresented among studies where GC uptake was associated with cancer-specific distress and perceived risk of cancer. This may suggest that cancer affected and unaffected individuals' decisions regarding GC uptake are influenced by different motivating factors. Perhaps unaffected individuals require higher levels of cancer concern to engage with these services compared to individuals already affected by cancer, who may more readily perceive GC as personally relevant and be more likely to receive a referral through their existing engagement with the healthcare system.

Finally, perceived benefits and limitations had strong support as predictors of genetic test *intention* in a recent systematic review (49); however evidence for an association with actual GC uptake in this review was weaker. Possible explanations for this are use of perceived benefits and limitations of genetic testing to predict GC uptake, and/or intentions being inconsistent predictors of actual health behaviour (59), which was also reflected in the findings of this review.

Practical insights may be better gleaned from the reasons for attendance and non-attendance at GC. Concerns about the cost of GC and testing were raised, even in countries where GC (and genetic testing for eligible individuals) is offered for free through a national health system. The potential implications of genetic testing on insurance were also of concern to individuals considering GC, despite most of the countries in which the studies were based

having legislation or industry standards to protect against genetic discrimination. These findings add to the mounting evidence of inequity in access to genetics services in the community (51, 56), which in this case may be partly overcome by increasing awareness of the actual costs and availability of GC.

Concerns about family members also play an important and seemingly complex role in decisions about GC, acting as both a motivating factor and a barrier to GC uptake. Personal decisions regarding GC may be complicated by differing opinions among family members, as demonstrated by discordant GC uptake within families (34). While much research has been done on family communication regarding genetic test results (60), less is known about how the family influences the pursuit of genetic information in these earlier decision-making stages. The diverse inputs contributing to GC decision-making speak to the complexity of the decision, with individuals balancing their own informational and emotional needs with those of their family, all while navigating what can be a complex and costly healthcare system.

Decisional and emotional conflict are consistently reported as important issues facing counselees, yet the decisional and emotional support provided by GC was not explored or mentioned as a factor in the decision to attend. Individuals who did not have an opportunity to discuss their referral were less likely to attend and some individuals had declined GC based on not wanting genetic testing, or a low perceived personal relevance despite being at increased risk. This is further evidence of the poor public awareness of GC and its goals reflected in other findings of this review and reported elsewhere (12, 13). Given health professionals also lack awareness of GC, evidenced by sub-optimal referral practices (15, 16), it is likely that both patients and referring health professionals lack the knowledge and

confidence to initiate discussions regarding GC. However, the importance of appropriate referral and decision support cannot be overstated given the potential for poor health outcomes among high-risk individuals with inadequate cancer screening. In addition there is evidence of negative psychosocial outcomes among those unaware of their referral to GC (61), and lower decisional satisfaction among decliners compared to attenders (62). This suggests that individuals may be making decisions inconsistent with their personal values and underscores the need for more research into decision-making about genetic counselling.

Strengths and limitations of the review

This review provides an overview of a range of factors that may influence actual GC uptake for hereditary cancer. We feel this targeted approach is a strength of the review given that intent has not proved to be a reliable predictor of actual health behaviour (19, 21, 59), and the cohorts and outcomes of GC differ across sectors. There was a stronger focus on reasons for GC non-attendance as identification of barriers to GC uptake was viewed as important for identifying areas for improvement to practice.

It is possible that nuances of individual hereditary cancer syndromes may influence GC uptake, in which case overrepresentation of hereditary breast/ovarian cancer cohorts in this review would make the results less generalizable. Also affecting the generalizability of the results is the limited ethnic diversity and underrepresentation of males in study cohorts and international variation in health care costs, GC service delivery and legislation around genetic discrimination and insurance. Uptake rates may also have been influenced by the variable time period allowed for decision-making across studies, which was often not explicitly stated. Other limitations common to studies included in this review that may have influenced the

findings include: limited sample sizes, different GC models of care, differences in measures used and operationalization of measures. There is also the possibility of selection bias in studies recruiting from existing research cohorts, or where GC was offered within the context of the research study. An additional limitation of the review is the reliance on a single coder for study selection and critical appraisal, which may impact on the reliability of the results.

Future studies aiming to assess factors associated with GC uptake would benefit from utilising validated measures, accounting for possible confounders in the analyses and considering the consistency of measures used across studies to enable comparisons across cohorts. Prospective studies of GC uptake would also add value to the existing literature by reducing the impact of selection and recall bias. Future research should also consider the representativeness of study samples and aim to include individuals from culturally and socioeconomically diverse backgrounds.

Important directions for future research identified by this review include research regarding GC uptake for hereditary cancer syndromes other than hereditary breast/ovarian cancer, investigation of the decision support needs of individuals for whom GC is recommended and the development of decision support interventions to target this group. Attempts have been made by genetics health professionals to improve access to services through implementation of alternate service delivery models, including telephone GC and telegenetics, with positive outcomes.^(63, 64) Continuation of this work is important, as is seeking out new ways to break down institutional barriers to access, including cost, logistics and appropriate referral. Improving awareness and understanding of GC in the community and among non-genetics health care professionals, particularly those who are likely to refer to GC, is also important as

the transition to genomic technologies continues and demand increases. Further research into effective health messaging and education in this context is strongly recommended.

CONCLUSION

This review demonstrates that there is room for improvement in GC uptake. The review allows a better understanding of the sociodemographic, clinical and psychosocial factors that influence GC uptake. This, combined with the reasons behind the decision to attend GC, may have the potential to improve health outcomes among those at potentially increased risk of cancer by minimizing barriers to accessing genetic services. One area for improvement identified by this review is increasing awareness of GC in the community and among health professionals to empower both groups to initiate discussions about GC. How best to educate health professionals and potential counselees, as well as methods of providing decision support to those considering GC, are important areas for future research.

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