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# Decision aids for female BRCA mutation carriers: a scoping review

Sarah A McGarrigle ,[1,2] Geraldine Prizeman,[3] Carol Spillane,[2] Niamh Byrne,[2] Amanda Drury,[4,5] Manria Polus,[5] David Mockler,[6] Elizabeth M Connolly,[2,7] Anne-Marie Brady ,[3] Yvonne P Hanhauser[2]

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A-MB and YPH contributed equally.

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[1]Faculty of Health Sciences, Trinity College Dublin, Dublin, Ireland
[2]Breast Care Department, St James's Hospital, Dublin, Ireland
[3]Trinity Centre for Practice and Healthcare Innovation, School of Nursing & Midwifery, Trinity College Dublin, Dublin, Ireland
[4]School of Nursing, Psychotherapy and Community Health, Dublin City University, Dublin, Ireland
[5]School of Nursing & Midwifery, Trinity College Dublin, Dublin, Ireland
[6]John Stearne Library, Trinity College Dublin, Dublin, Ireland
[7]Department of Surgery, Trinity College Dublin, Dublin, Ireland

**Correspondence to**
Yvonne P Hanhauser;
YHanhauser@stjames.ie

## ABSTRACT

**Objectives** Women who inherit a pathogenic *BRCA1* or *BRCA2* mutation are at substantially higher risk of developing breast and ovarian cancer than average. Several cancer risk management strategies exist to address this increased risk. Decisions about which strategies to choose are complex, personal and multifactorial for these women. Decision aids (DAs) are tools that assist patients in making health-related decisions. The aim of this scoping review was to map evidence relating to the development and testing of patient DAs for cancer unaffected *BRCA* mutation carriers.

**Design** Scoping review conducted according to the Joanna Briggs Institute's (JBI's) scoping review methodological framework.

**Data sources** MEDLINE, EMBASE, CINAHL, Web of Science. No restrictions applied for language or publication date. A manual search was also performed.

**Eligibility criteria for selecting studies** Studies on DAs for cancer risk management designed for or applicable to women with a pathogenic *BRCA1* or *BRCA2* mutation who are unaffected by breast or ovarian cancer.

**Data extraction and synthesis** Data were extracted using a form based on the JBI instrument for extracting details of studies' characteristics and results. Data extraction was performed independently by two reviewers. Extracted data were tabulated.

**Results** 32 evidence sources relating to development or testing of 21 DAs were included. Four DAs were developed exclusively for cancer unaffected BRCA mutation carriers. Of these, two covered all guideline recommended risk management strategies for this population though only one of these was readily available publicly in its full version. All studies investigating DA effectiveness reported a positive effect of the DA under investigation on at least one of the outcomes evaluated, however only six DAs were tested in randomised controlled trials.

**Conclusion** This scoping review has mapped the landscape of the literature relating to developing and testing, DAs applicable to cancer unaffected *BRCA* mutation carriers.

## STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ This study has provided a comprehensive mapping of the literature relating to the features and efficacy testing of existing decision aids for BRCA mutation carriers without a personal history of cancer.
⇒ This scoping review was conducted according to the Joanna Briggs Institute's scoping review methodological framework and was guided by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews checklist.
⇒ Decision aids included in this review were identified by searching four databases, reference lists and the internet, however, it is possible that other relevant decision aids may exist elsewhere in the grey literature.
⇒ A formal independent quality appraisal of included evidence sources was not conducted, however, quality appraisals conducted by authors of included studies were summarised where applicable.

## INTRODUCTION

### Background

*BRCA1* and *BRCA2* are tumour suppressor genes that play an important role in the repair of DNA damage. Women who inherit a pathogenic mutation in the *BRCA1* or *BRCA2* genes are at substantially higher risk of developing breast and ovarian cancer over their lifetime than the average woman. Estimates for lifetime breast cancer risk vary between studies and differ according to mutation location and family history but have been reported to be in the region of 45%–85% for female *BRCA1* mutation carriers and 27%–84% for female *BRCA2* carriers to age 70 overall.[1–13] Furthermore, some studies have reported that *BRCA* mutation carriers born in recent decades, have a substantially higher risk of developing breast cancer than those in earlier birth cohorts.[7 14–16]

Cumulative ovarian cancer risk to age 80 was estimated to be 44% for *BRCA1* mutation carriers and 17% for *BRCA2* mutations carriers in a study using data from a prospective cohort.[1] This represents a significant risk compared with a population average of≤2%.[17]

Following a positive genetic test, women diagnosed as *BRCA* gene mutation carriers may be followed up in high-risk programmes for monitoring and management. Management strategies in this setting are aimed at

early detection and/or prevention of the disease. Early detection strategies aim to diagnose breast cancer at an early stage to improve clinical outcomes; these include radiologic surveillance at regular intervals by mammography and MRI. Radiological screening techniques have not been proven to be effective in detecting ovarian cancer at an early stage. Prevention strategies aim to reduce a woman's risk of developing breast or ovarian cancer by means of prophylactic surgery (including risk-reducing bilateral mastectomy and/or bilateral salpingo-oophorectomy (BSO)) or risk-reducing medication (chemoprevention) with drugs such as tamoxifen, anastrozole or raloxifene to reduce breast cancer risk.[18]

For BRCA mutation carriers, decisions about which risk management strategies to choose are complex, personal and multifactorial. Each option has associated risks and anticipated outcomes, which women need to understand to make an informed decision regarding which interventions to choose. Decision aids (DAs) in various formats, have been developed internationally to support decision-making for *BRCA* mutation carriers. Such tools require sophisticated design to effectively support decision-making, communicate risk, and clarify patients' values and preferences.[19] DAs for BRCA mutation carriers have not yet been widely incorporated into routine clinical practice.

### Rationale

In order to better understand the features of existing DAs for this population and to reveal which of these DAs may be appropriate for various populations of *BRCA* mutation carriers a scoping review of existing DAs designed to support decision-making around risk management for female BRCA mutation carriers was conducted.

The overarching goal of this scoping review was to explore the breadth of the literature in this field and to map evidence relevant to cancer risk-management DAs for female *BRCA* mutation carriers without a personal history of cancer. This information may be beneficial for designing new DAs or adapting existing DAs to support decision-making in terms of cancer risk management for female BRCA mutation carriers.

A scoping review can be used to identify, map and discuss certain characteristics in papers or studies.[20] The aim of this review is to summarise the key characteristics (content, features and efficacy) of patient DAs for female BRCA mutation carriers who are as yet cancer unaffected. A scoping review approach can provide a broad overview of the landscape of the literature and is, therefore the most appropriate design for this evidence synthesis.[21]

### Review question

The question that this scoping review aimed to answer is:

What are the characteristics of patient DAs that have been developed to support risk-management decision-making in cancer unaffected female BRCA mutation carriers?

### Objectives

The objectives of this scoping review were:

- To identify and summarise the key features of patient DAs that have been developed for or are applicable to cancer unaffected female BRCA mutation carriers to support decision-making in terms of choosing which cancer risk management options to opt for.
- To map the evidence related to testing of these DAs.

## METHODS

This scoping review was conducted according to the Joanna Briggs Institute's (JBI's) scoping review methodological framework.[20] In addition, the Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews checklist was used for guidance.[22] The published protocol for this scoping review is available here.[23]

### Inclusion criteria

#### Types of participants

This review considered studies on DAs for cancer risk management designed for or applicable to women with a pathogenic *BRCA1* or *BRCA2* mutation who are unaffected by breast or ovarian cancer.

#### Concept

The concept of interest in this scoping review is patient DAs for female BRCA mutation carriers to support decision-making around cancer risk-management options.

In the absence of a universally accepted definition for 'decision aid' we included DAs that were (1) described as such by their developers and/or (2) included in the Ottawa Hospital Research Institute's patient DAs inventory[24] and/or that in the author's judgement could be considered a DA based on the DA definition provided by the International Patient Decision Aids Standards (IPDAS) Collaboration.[25]

#### Context

The context of this review is decision-making supports for female BRCA mutation carriers without a personal history of breast or ovarian cancer. Sources of evidence on cancer risk management patient DAs for BRCA mutation carriers pertaining to any contextual setting were eligible for inclusion.

#### Types of evidence sources

##### *Included*

(1) Studies that describe the development and/or testing of a patient DA suitable for cancer unaffected female BRCA mutation carriers to support decision-making in terms of choosing which cancer risk management options to opt for; (2) standalone DAs applicable to this population (ie, those that are available publicly but whose development has not necessarily been reported in a journal article); and (3) systematic reviews of the above-mentioned evidence sources.

*Excluded*

This review did not include case reports, non-systematic reviews, protocols, letters, posters or conference abstracts. Studies that described patient DAs aimed solely at BRCA mutation carriers with a personal history of breast or ovarian cancer were excluded. Patient DAs that focused on interventions that do not manage or reduce cancer risk (such as genetic testing, breast reconstruction or hormone replacement therapy) were also excluded.

### Search strategy

A three-step search strategy was used. First, an initial limited search of the databases MEDLINE (Ovid) and EMBASE was conducted. This initial search was followed by an analysis of the text words contained in the title and abstract of retrieved papers, and of the index terms used to describe the articles. A second search using identified keywords and index terms was then be undertaken across all included databases (MEDLINE, EMBASE, CINAHL, Web of Science) (online supplemental appendix 1). Databases were searched from inception to 6 October 2020. No restrictions were applied for language or publication date. The reference lists of reports and articles selected for inclusion in the review were also searched for additional sources. Finally, a manual search of the internet using Google Scholar and The Ottawa Hospital Research Institute Decision Aid Library Inventory (decisionaid.ohri.ca) was conducted on 9 March 2022.

### Evidence source selection

Search results were uploaded to EndNote X8 (Clarivate Analytics, PA, USA) and duplicate records were removed. Retrieved studies were initially screened for inclusion by title and abstract by two review authors independently using the web-based Covidence screening tool (Veritas Health Innovation, Melbourne, Australia). Disagreements were resolved by discussion. Full-text papers and reports were retrieved for potentially relevant studies. For these studies, Covidence software was again used to assess and document studies for inclusion and exclusion according to the inclusion criteria. Studies for inclusion were selected independently by two review authors. Disagreements were resolved by discussion. In cases of no consensus, final resolution was achieved by involving a third review author as arbiter.

### Data extraction

Data were extracted from included articles and other evidence sources using a data extraction form developed by the reviewers, pilot tested and modified in an iterative process to produce the final version (online supplemental appendix 2). The design of this instrument is based on the JBI instrument for extracting details of the studies characteristics and results. Data extraction was performed independently by two reviewers. Disagreements between the reviewers were resolved through discussion. Extracted data were tabulated.

### Patient and public involvement

Patients and public were not formally involved in the development of this scoping review protocol; however, the research questions were informed by the author team's extensive clinical experience working with BRCA mutation carriers.

### Deviations from the protocol

The data extraction template was amended from that published with the protocol to include additional fields to capture pertinent data identified during pilot testing (online supplemental appendix 2).

## RESULTS

### Evidence source inclusion

A total of 1007 articles were retrieved through database searching. An additional 1647 records were identified through searching other sources including reference lists of included studies (n=5), the Patient Decision Aids Inventory maintained by The Ottawa Hospital Research Institute (n=12) and a manual internet search of Google Scholar performed on 9 March 2022 (n=1630). Following exclusion of duplicates and irrelevant records; 32 studies/evidence sources were included in the scoping review. The screening and selection process is depicted in figure 1.[26]

Of the included articles/evidence sources; 15 solely described DA development or presented a developed DA,[27–41] 10 reported testing of a previously developed DA(s)[42–51] and 6 articles reported both development and testing of a DA.[52–57] In addition, one systematic review of DAs developed for the population of interest was included.[58]

Within the above-mentioned evidence sources, 21 DAs that met the inclusion criteria were identified. However, of these, there appeared to be some overlap between two pairs of DAs; those reported in Tiller *et al*[55] and C. f. G. E. N. Health[31] with the latter DA based on work reported in the former and those reported by van Roosmalen *et al*[56 57] whereby the later study incorporated the former DA as part of a wider decision-making intervention. There may also be some overlap between the DAs described by van Roosmalen *et al*[56 57] and Unic *et al*[41] that were developed by the same author teams, though the extent of overlap is difficult to gauge as the full DAs are not publicly available.

### Review findings

#### Characteristics of included evidence sources

An overview of the included evidence sources is shown in online supplemental tables 1 and 2.

#### Target populations

Of the 21 included DAs; 8 were developed exclusively for known BRCA mutation carriers.[27 28 30 40 52–54 57] A further DA was aimed at women undergoing genetic testing for germline BRCA 1/2 mutations but whose genetic test results were not necessarily known.[56] 11 DAs were targeted at mixed groups of women at increased risk of developing

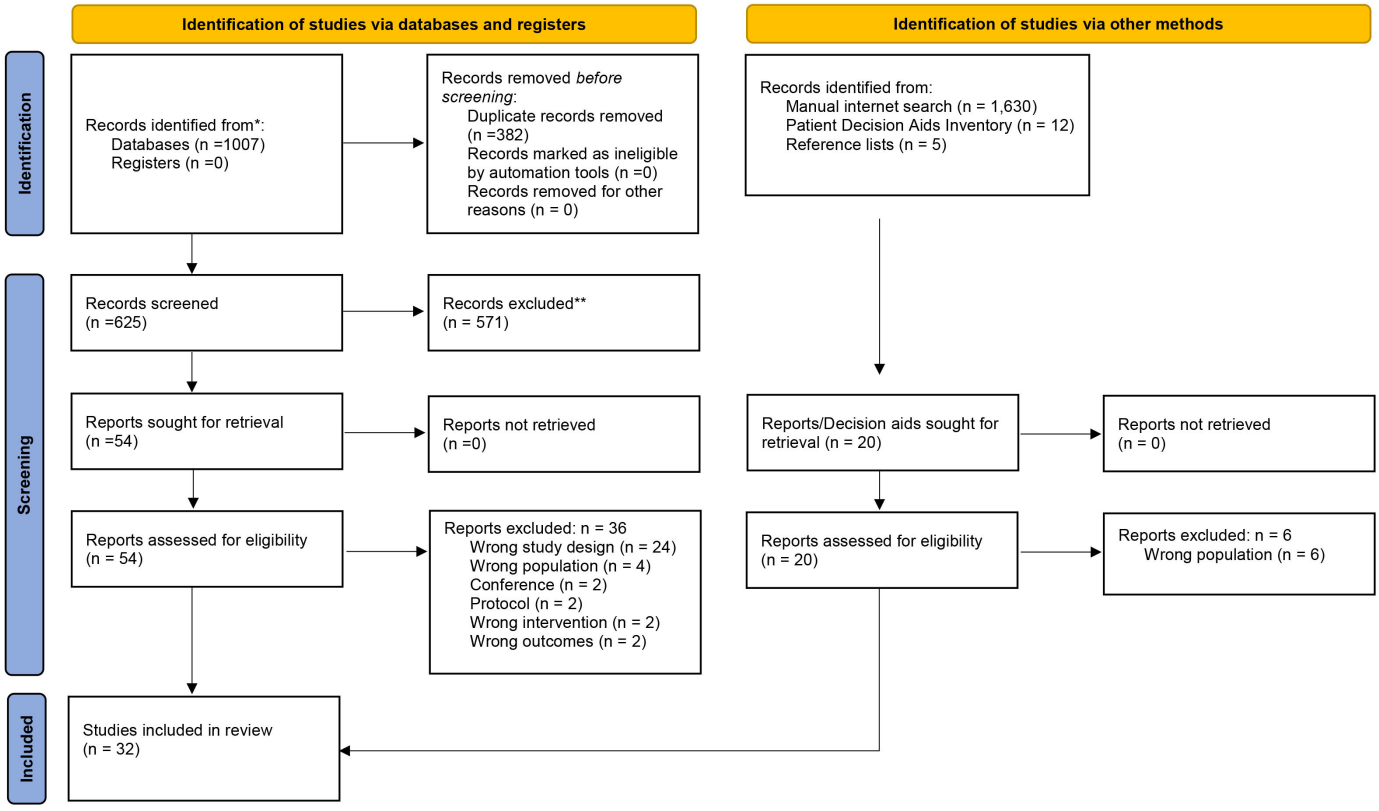


**Figure 1** Preferred Reporting Items for Systematic Reviews and Meta-Analyses flow diagram detailing search results and evidence source selection and inclusion process. Adapted from Page *et al*.[26]

breast or ovarian cancer.[31–39 41 55] In addition, one DA was aimed at women across the spectrum of breast cancer risk including those with a known BRCA mutation.[29] Five of the identified DAs were targeted specifically at women without a personal history of breast or ovarian cancer 'previvors'.[28 40 41 53 54] Three DAs were targeted at those unaffected by breast cancer (but not necessarily unaffected by ovarian cancer)[29 37 38] and six DAs were aimed at women unaffected by ovarian cancer (but not necessarily unaffected by breast cancer).[30 31 35 39 52 55] Five DAs were targeted at women either affected or unaffected by breast cancer.[27 32 36 56 57] For two DAs the target population in terms of cancer affected status was not reported or unclear.[33 34]

## DA development methods

The IPDAS include 'a systematic development process' as a quality criterion for patient DAs.[59] DA development methods were reported (fully or partially) for 15 of the included DAs. Methodology used during DA development process varied but frequently involved a review of the literature and/or clinical guidelines in the field,[27 28 30 37–40 52–55] a needs assessment with targeted end users,[27 29 30 40 53–55 55] prototype development,[27 29 30 39 40 53–55] acceptability and usability testing followed by refinement based on end user and/or clinician feedback.[27 30 39 41 53 54] In the case of DAs that incorporated a cancer risk estimate calculator or algorithm, modelling approaches such as Markov or Monte Carlo modelling were used.[28 52] In one case, existing risk prediction models were incorporated into the DA.[29] The DA development process was often overseen by a steering committee or working group.[29 30 37 38 40 53–55]

## Risk management options addressed

An overview of the risk management options addressed in each DA is shown in online supplemental table 1 and depicted in figure 2. Five DAs included both breast and ovarian cancer risk management options.[28 40 53 56 57] 10 DAs focused on breast cancer risk management options.[27 29 32 33 36–38 41 52 54] Many of these also briefly mentioned ovarian cancer risk management options[27 36 54] and several included BSO but focused on this intervention from a breast cancer risk management rather than an ovarian cancer risk management perspective.[29 33 52] Five DAs addressed ovarian cancer risk management options only.[30 31 34 39 55] Eight DAs included[27 29 33 52–54] or focused solely[37 38] on chemoprevention (risk reducing medication). Of these one DA was targeted exclusively at premenopausal[37] and one DA exclusively at postmenopausal women[38] based on the different risk-reducing medications recommended for each of these groups. A further two DAs mentioned chemoprevention briefly, however this option was not a focus of these DAs.[32 36]

## Presentation of risks and benefits

The IPDAS quality criteria framework for patient DAs outlines several quality criteria for presenting probabilities

| Decision Aid | Developed exclusively for *BRCA* mutation carriers | Developed exclusively for BC & OC unaffected women 'previvors' | Risk management options addressed | | | | | Full DA readily available publicly |
|---|---|---|---|---|---|---|---|---|
| | | | Surveillance/Screening | | RRM | BSO | Chemoprevention (for BC ) | |
| | | | Breast | Ovarian | | | | |
| Kaufman 2003 | ✓ | × | ✓ | ✓** | ✓ | ✓** | ✓ | ×* |
| Armstrong 2005 | ✓ | × | ✓ | × | ✓ | ✓*** | ✓ | ×* |
| Jabaley 2020 | ✓ | ✓ | ✓ | ✓ | ✓ | ✓ | ✓ | ✓ |
| Kurian 2012 | ✓ | ✓ | ✓ | × | ✓ | ✓ | × | ✓ |
| Collins 2016 | × | ? | ✓ | × | ✓ | ✓*** | ✓ | ✓ |
| Harmsen 2018 | ✓ | ? | × | × | × | ✓ | × | ✓ |
| Centre for Genetics Education, NSW Health (2012 update) Breast | × | ? | ✓** | × | ✓ | × | ✓** | ✓ |
| Centre for Genetics Education NSW Health (2017) Ovarian | × | ? | × | × | × | ✓ | × | ✓ |
| Healthwise staff a (2020 update) Breast | × | ? | ✓ | × | ✓ | ✓*** | ✓ | ✓ |
| Healthwise staff b (2020 update) Ovarian | × | ? | × | ✓** | × | ✓ | × | ✓ |
| Mayo Clinic Staff (2020 update) Ovarian | × | ? | × | ✓** | ✓** | ✓ | × | ✓ |
| Mayo Clinic Staff (2021 update) Breast | × | × | ✓** | × | ✓ | ✓** | ✓** | ✓ |
| Metcalfe 2007 | ✓ | ✓ | ✓ | × | ✓ | ✓ | ✓ | ×* |
| NICE 2017 (Pre-menopausal) | × | ? | × | × | × | × | ✓ | ✓ |
| NICE 2017 (Post-menopausal) | × | ? | × | × | × | × | ✓ | ✓ |
| TILLER 2003 | × | ? | × | ✓ | × | ✓ | × | ×* |
| VANROOSMALEN BJC 2004a | ✓# | × | ✓ | ✓ | ✓ | ✓ | × | ×* |
| VANROOSMALEN JCO 2004b | ✓ | × | ✓ | ✓ | ✓ | ✓ | × | ×* |
| Witt 2014 | × | ? | × | × | × | ✓ | × | ×* |
| Kautz-Freimuth 2021 | ✓ | ✓ | ✓ | × | ✓ | ✓ | × | ×* |
| Unic 1998 | × | ✓ | ✓ | × | ✓ | × | × | ×* |

**Figure 2** Overview of target populations and risk management options addressed in each decision aid (DA). *May be made available through contacting authors but not readily accessible in public domain. **Option mentioned but not a main focus of DA. *** BSO included as a BC risk management option in DA. #Women being tested for a BRCA mutation but not necessarily confirmed BRCA mutation carriers. BC, breast cancer; BSO, bilateral salpingo-oophrectomy; RRM, risk-reducing mastectomy.

of outcomes including the use of multiple methods to view probabilities (words, numbers, visual diagrams).[59] Among the DAs reviewed here, where included, various approaches were used to present baseline cancer risks and cancer risk reductions associated with the different options. Commonly, a text description of risks (and risk reductions) was included,[27 29–39 41 53–56] often with a visual presentation by means of bar charts,[27 28 53 57] pie charts,[30] shaded icon arrays[29 30 33 37–39 54] or other graphical presentations.[29 31 52] Other benefits and harms (or side-effects) of the various options were typically portrayed using text descriptions[27 29–41 53–56] and in some cases photographs and videos.[41 56]

### Values clarification approaches

According to the IPDAS patient DA quality criteria framework, DAs should include 'methods for clarifying and expressing patients' values' to enable patients to consider what matters most to them.[59] 13 of 21 DAs included an activity that enabled end users to work through their values and feelings in relation to the risk management options presented. Various values clarification approaches were used such as rating or scoring statements or attributes relating to the benefits and harms of the risk management option(s) in question based on how important they are to the user.[27 30 33 34 37–39 53–55] Several DAs included a space for users to write additional thoughts or concerns that they have.[30 33 34 37–40 53 54] In some cases, users are asked to rank statements in order of importance in an attempt to clarify which values matter most to them.[30] In some cases, more complex approaches to values clarification were used such as time trade-off methods[41 57] or model-based approaches.[27]

### DA recommendation for which option(s) the patient should choose

The majority of included DAs did not provide a recommendation for which option(s) the patient should choose. One DA provided a recommendation for which option the patient should choose based on their answers to values clarification statements by stating that 'If you mainly 'agree' with these three statements, removal of the fallopian tubes and ovaries is the best option for you. If you mainly 'disagree' with these three statements, initial removal of the fallopian tubes and removal of the ovaries at a later date is the best option for you'.[30] In addition, one DA implies, but does not explicitly recommend, which option the patient should choose by indicating that during the 'decision task' activity, the highest preference score indicates the risk management option that is most consistent with the values and preferences the woman entered in the decision task.[27]

### DA formats and availability

The most common format of the DAs was paper-based, typically in the form of a booklet or brochure (online supplemental table 1). Some of these booklets were provided with an accompanying videotape containing informational material.[41 56] Other paper-based formats included pdf formats available online or binders containing printed material.[52] The second most common DA format was web-based. Web-based DAs were usually interactive to some degree with some web-based DAs enabling a large degree of individualisation particularly in terms of presenting personalised cancer risk estimates based on user inputted data.[28 29] Some web-based DAs were also available as printable pdf versions.[33 34] One DA was in the format of a CD-ROM.[27] Only 12 DAs[28–38 53] were available in full in the public domain without requirement to contact the developers for access (figure 2).

### Year of DA development or update

The identified DAs span a time period of greater than 20 years in terms of their year of development or last update. More than half of the included DAs (n=12), however, were developed and/or updated in the past 10 years[29–31 33–40 53] with six of these developed/updated in the past 5 years.[33–36 40 53] For several DAs the date of last update was not readily apparent. For DAs whose development was reported in journal articles, the development year was recorded as the year of article publication unless a more recent update was available publicly in which case the later year was reported (online supplemental table 1). For publicly available DAs (whose development was not necessarily reported in journal articles), the development/update year was recorded as year of update or last review stated on the DA when this was reported (online supplemental table 1).

## Intended moment(s) of use of DAs

In the majority of cases, DAs were intended to be self-administered by patients at home.[27 30–34 37 38 40 41 52 54–56] Five DAs were designed to be used collaboratively with a clinician.[28 29 37 38 53] For five DAs, developers specified that the DA was intended to be used by the patient at home in addition to a consultation with a healthcare professional.[27 30 40 41 54] One DA included a shared decision-making intervention that was interview administered by a researcher.[57] For three DAs the intended moment of use of the DA was unclear or not explicitly reported though these appeared to be suitable for self-administration by patients at home.[35 36 39]

### Patient and public involvement

There was some degree of patient and public involvement (PPI) in development of the majority (14 of 21) of included DAs (online supplemental table 1). PPI commonly entailed a needs assessment with target end users of the DA by means of focus groups or interviews.[27 29 30 39 40 53–55] Target end user representatives frequently contributed to DA development through reviewing the DA prototype and/or subsequent DA versions and providing feedback to facilitate DA refinement.[27 29 30 40 53] In some cases, DA development was led by a steering group containing patient representatives.[37 38 55] In some cases, patients and their families featured in the DA informational material through featuring on videos or providing quotes about

their personal experiences.[27 41 56] For the remaining DAs PPI was either not reported[28 33–36 52 57] or where PPI was reported, the nature of patient involvement was not specified.[31 32]

### Adherence to quality criteria

In this scoping review, a formal quality appraisal of included DAs was not performed as per guidance on conducting scoping reviews.[20] However, a recent full systematic review on this topic evaluated the quality of DAs for preventive treatment alternatives for BRCA 1/2 mutation carriers. In this review, the authors reported that only 9 of the 20 DAs included in their review (19 of which are included in the current scoping review) met fundamental quality criteria of the IPDAS Collaboration (IPDASi V.4.0).[58]

### Testing and effectiveness of DAs

The IPDAS recommend that patient DAs are field tested with users (patients and practitioners) to evaluate whether the DA is acceptable, balanced in terms of information and is understood by those with limited reading skills. This framework also recommends DA efficacy testing in terms of determining whether the DA improves the match between the chosen option and the features that matter most to the informed patient.[59]

11 of the 21 included DAs had been tested for efficacy in 15 primary studies. Study designs included seven randomised controlled trials (RCTs), one non-RCT, two single group pretest/post-test studies, four pilot studies. One study that compared responsiveness of several instruments used to evaluate DA effectiveness, using two DAs for BRCA mutation carriers, was also included, however this study did not report results in terms of effectiveness of these two DAs themselves.[49] In addition, one systematic review synthesised evidence on effectiveness of four of the included DAs.[43] Outcomes evaluated typically included decision related outcomes and/or information related outcomes. In some cases, outcomes on actual preventive choice and other health related outcomes were evaluated. Pilot studies commonly evaluated DAs in terms of usability, feasibility or acceptability. A description of individual effectiveness studies and their findings is shown in online supplemental table 2. All of the included studies reported a positive effect of the DA under investigation on at least one of the outcomes evaluated. However, negative effects of DAs were also found at some time points. For example, Hooker *et al* reported increased distress among DA users compared with the control group at 1-month postrandomisation.[42] Indeed, timing appears to be relevant with some studies reporting differential effects of DAs on outcomes in the short term versus longer term.[42 48 57]

The included systematic review reported that BRCA mutation carriers using a DA had less decisional conflict, were more likely to reach a decision and were more satisfied with their decision, however, the authors noted that overall risk of bias was high or serious in all but one of the studies evaluated.[58]

## DISCUSSION

This scoping review has mapped evidence relevant to cancer risk-management DAs that are applicable to female BRCA mutation carriers without a personal history of cancer. Specifically, we have identified and described the features of cancer risk-management DAs for this population and reported on the efficacy testing of these DAs where this has been conducted.

Two other systematic reviews on this topic have been published by Krassuski *et al*[43 58] as well as a further study that incorporated a survey of existing DAs.[40]

Krassuski *et al* conducted a structural analysis and quality assessment of DAs for BRCA mutation carriers (with or without breast/ovarian cancer) and examined their applicability to the German context.[58] In this study they identified 20 patient DAs of which nine met fundamental IPDAS quality criteria. The authors reported that some DAs differed markedly in content from the recommendations of German guidelines.

Krassuski *et al* conducted a systematic review of effectiveness of DAs for BRCA mutation carriers (with or without breast/ovarian cancer) that have been tested in randomised control trials or pretest and post-test studies. This study reported that DAs significantly improved decision related outcomes in female BRCA mutation carriers, though the authors noted bias concerns regarding most of the included studies.[43]

Kautz-Freimuth *et al* incorporated a review of existing DAs for BRCA mutation carriers as part of their development process for new DAs targeted towards German BRCA mutation carriers. Seven DAs were included in this review and an overview of the structural elements and basic medical contents of these DAs was provided. The authors concluded that due to various limitations related to content of the DAs; none were transferable to the German setting.[40]

Our scoping review differs from these articles in a number of ways. The population of interest for our study was BRCA mutation carriers without a personal history of breast or ovarian cancer often termed ‘previvors’. As such, DAs developed solely for cancer affected women were excluded from this review. In addition, as a scoping review we took a broader approach in terms of included evidence sources by combining a synthesis of features of existing DAs that can be used by cancer unaffected BRCA mutation carriers, the efficacy testing of these DAs and systematic reviews of same. As such, we believe that this work is a useful resource for clinicians and researchers which maps current evidence relating to features and efficacy of existing DAs for cancer unaffected BRCA mutation carriers in a single paper.

The findings described here therefore build on, complement and include those reported by Krassuski and colleagues.[40 43 58]

Our findings demonstrate that only four DAs have been developed exclusively for known BRCA mutation carriers without a personal history of cancer 'previvors'.[28 40 53 54] Considering the unique issues that these women face in relation to their high cancer risk and decision-making about their risk management, DAs designed exclusively for this group may be more appropriate.

Furthermore, of the DAs designed exclusively for cancer unaffected BRCA mutation carriers, only two included the full range of guideline[18] recommended breast and ovarian cancer risk management strategies[53 54] and only one of these is readily available publicly in its full version.[53]

The included DAs span a period of >20 years in terms of their date of development or last update. It is likely that time since development/update may have impacted content and features of DAs. For example, DAs developed recently were more likely to be web-based with four of the six DAs developed in the last 5 years having a web-based format. Furthermore, the evidence base in the BRCA field is continuously evolving. It is noteworthy that content included in some DAs is not in line with current evidence. For example, current evidence does not support screening for ovarian cancer as a valid risk management option for BRCA mutation carriers, therefore, DAs that include this as a risk management option[53 55–57] may no longer be appropriate for use in their current version.

In addition, breast cancer risk reduction was listed as a benefit of BSO in eight DAs.[29 33–36 39 53 54] Due to the conflicting evidence in relation to this[60–62] it may be inappropriate to include breast cancer risk reduction as a benefit of BSO for BRCA1 mutation carriers in DAs presently.

Thus, currently there is no DA publicly available that has been designed exclusively for cancer unaffected BRCA mutation carriers, that includes all breast and ovarian risk management strategies recommended for this population together with a values clarification activity and that aligns with current best evidence in the field.

In terms of effectiveness of the existing DAs for BRCA mutation carriers; the included studies all reported a positive effect of the DA in question on at least one decision related or information related outcome. However, only six DAs were tested in an RCT, and bias concerns have been raised in relation to most of these RCTs.[43] In addition, various instruments were used to assess outcomes in the DA effectiveness studies, some of which were validated and others not. Furthermore, it is possible that publication bias may have contributed to an over-representation of positive findings on DA effectiveness in the literature. Publication bias was not formally evaluated in this scoping review. Thus, while the reported effectiveness of these DAs in improving various decision and information related outcomes is promising; further high-quality studies using validated instruments are required to clarify the influence of DAs on these outcomes.

### Limitations of this review

This scoping review has several limitations. As the intention of this study was to map the landscape of the evidence on development and testing of DAs applicable to cancer unaffected BRCA mutation carriers we took an inclusive approach to eligibility of evidence sources for inclusion. In the absence of a universally accepted definition for 'decision aid' we included DAs that were described as such by their developers and/or included in the Ottawa Hospital Research Institute's patient DAs inventory and/or that in the author's judgement could be considered a DA based on the DA definition provided by the IPDAS Collaboration.[59] In addition, DAs included in this review were identified by searching databases, reference lists and the internet. It is possible that other relevant DAs may exist elsewhere in the grey literature. Several of the included DAs were not readily accessible as full versions in the public domain; as such, details of their features and content were derived from the articles describing their development rather that the full DA version. This may have resulted in some DA features being omitted in this report. Finally, as a scoping review a formal quality appraisal of included evidence sources was not conducted thus the evidence on DA quality and the quality of studies testing DA effectiveness reported here was drawn from reports by other authors[43 58] rather than an independent appraisal.

## Conclusions

### Implications for research or practice

The features of existing DAs and evidence relating to their efficacy testing reported here and by others will serve as a useful basis for identifying which DAs are suitable for various populations of BRCA mutation carriers and will assist in the development of new DAs for this population.

**X** Sarah A McGarrigle @drsmcgarrigle

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**ORCID iDs**
Sarah A McGarrigle http://orcid.org/0000-0003-4404-6564
Anne-Marie Brady http://orcid.org/0000-0002-7112-6810

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