



BMJ Open Decision aids for female BRCA mutation carriers: a scoping review protocol

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ABSTRACT

Introduction Women who inherit a pathogenic mutation in Breast Cancer Susceptibility Genes 1 or 2 (BRCA1 or BRCA2) are at substantially higher risk of developing breast and ovarian cancer than the average woman. Several cancer risk management strategies exist to address this increased risk. Decisions about which risk management strategies to choose are complex, personal and multifactorial for these women. This scoping review will map evidence relevant to cancer risk management decision making in BRCA mutation carriers without a personal history of cancer. The objective is to identify and describe the features of patient decision aids that have been developed for BRCA mutation carriers. This information may be beneficial for designing new decision aids or adapting existing decision aids to support decision making in this population.

Methods and analysis This scoping review will be conducted according to the Joanna Briggs Institute's scoping review methodological framework. The Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews checklist will be used for guidance. Studies on decision aids for women with a BRCA mutation who are unaffected by breast or ovarian cancer will be considered for inclusion. Five electronic databases will be searched (MEDLINE, EMBASE, Cochrane Library, CINAHL, Web of Science) with no restrictions applied for language or publication date. Studies for inclusion will be selected independently by two review authors. Data will be extracted using a predefined data extraction form. Findings will be presented in tabular form. A narrative description of the evidence will complement the tabulated results.

Ethics and dissemination Ethical approval for conducting this scoping review is not required as this study will involve secondary analysis of existing literature. Findings will be published in a peer-reviewed journal and presented at relevant conferences.

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

Strengths and limitations of this study

- This study will provide a comprehensive mapping of literature relevant to cancer risk management decision-making aids in BRCA mutation carriers without a personal history of cancer.
- This scoping review will be conducted according to the Joanna Briggs Institute's scoping review methodological framework and will be guided by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews checklist.
- The search strategy will include five databases with no restrictions applied for language or publication date.
- Relevant evidence may exist in the grey literature, but our review is limited to the published literature.

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) 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 in various formats have been developed internationally to support decision-making for BRCA mutation carriers.^{19–23} Such tools require sophisticated design to effectively support decision-making, communicate risk and clarify patients' values and preferences.²⁴ Decision aids for BRCA mutation carriers have not yet been widely incorporated into routine clinical practice.

Rationale

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

This enquiry is guided by one key question, namely, what are the features of patient decision aids that have been developed to support risk management decision-making for female BRCA mutation carriers.

This scoping review will map evidence relevant to cancer risk management decision making supports for female BRCA mutation carriers without a personal history of cancer. This information may be beneficial for designing new decision aids or adapting existing decision aids to support decision-making in terms of cancer risk management for female BRCA mutation carriers.

A preliminary search of the literature (Medline and Embase conducted in July 2020) revealed that a number of decision aids have been developed for this population; therefore, ample evidence sources exist on which to base this review.

The preliminary search did not reveal any existing scoping reviews of patient decision aids developed to support risk management decision making in BRCA mutation carriers. One quantitative systematic review of effectiveness of decision aids for BRCA mutation carriers that have been tested in randomised control trials or pre-test and post-test studies was identified.²⁵ The proposed

review will differ from this review in that it will summarise the features of all published decision aids for cancer unaffected female BRCA mutation carriers irrespective of whether their effectiveness has been tested in randomised control trials or pre-test and post-test studies.

This scoping review will identify all patient decision aids developed to support decision-making in female BRCA mutation carriers who have not had a cancer diagnosis and will examine the range of features contained in these decision aids. Effectiveness of these decision aids on improving the decision-making process will also be appraised where applicable.

A scoping review can be used to identify, map and discuss certain characteristics in papers or studies.²⁶ The aim of this review is to summarise the key characteristics (content and features) of patient decision aids for female BRCA mutation carriers. 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.²⁷

Review question

The question that this scoping review aims to answer is:

What are the features of patient decision aids that have been developed to support risk management decision-making in female BRCA mutation carriers.

Objectives

The objectives of this scoping review are:

(1) to identify and describe the features of patient decision aids that have been developed for female BRCA mutation carriers to support decision-making in terms of choosing which cancer risk management options to opt for.

METHODS AND ANALYSIS

This scoping review will be conducted according to the Joanna Briggs Institute's scoping review methodological framework.²⁶ In addition, the Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) checklist will be used for guidance.²⁸

Inclusion criteria

Types of participants

This review will consider studies on decision aids for cancer risk management in women with a pathogenic BRCA1 or BRCA2 mutation who are unaffected by breast or ovarian cancer.

Concept

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

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 patient decision aids for BRCA mutation carriers pertaining to any contextual setting are eligible for inclusion.

Types of evidence sources

This review will consider studies that describe the development and/or testing of a patient decision aid for female BRCA mutation carriers to support decision making in terms of choosing which cancer risk management options to opt for. Systematic reviews of the above-mentioned evidence sources will also be considered. This review will not include case reports, non-systematic reviews, protocols, letters, posters or conference abstracts. Studies that describe patient decision aids aimed solely at BRCA mutation carriers with a personal history of breast or ovarian cancer will be excluded also.

Search strategy

A three-step search strategy will be 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 (online supplemental appendix 1) will then be undertaken across all included databases (MEDLINE, EMBASE, Cochrane Library, CINAHL, Web of Science) (online supplemental appendix 1). Databases will be searched from inception to present. No restrictions will be applied for language or publication date. The National Institute for Health and Care Excellence guidelines¹⁸ will also be searched for relevant evidence. Thirdly, the reference list of reports and articles selected for inclusion in the review will be searched for additional sources.

Source of evidence selection

Search results will be uploaded to EndNote X8 (Clarivate Analytics, Pennsylvania, USA) and duplicate records will be removed. Retrieved studies will initially be 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 will be resolved by discussion. Full text papers and reports will be retrieved for potentially relevant studies. For these studies, Covidence software will once again be used to assess and document studies for inclusion and exclusion according to the inclusion criteria. Studies for inclusion will be selected independently by two review authors. Disagreements will be resolved by discussion. In cases of no consensus, final resolution will be achieved by involving a third review author as arbiter. Study selection will be documented in a PRISMA-ScR flow chart. Reasons for exclusion of articles will be included in the scoping review report.

Data extraction

Data will be extracted from included articles and other evidence sources using a data extraction form developed

by the reviewers (online supplemental appendix 2). The design of this instrument is based on the Joanna Briggs Institute instrument for extracting details of the studies characteristics and results. Data extraction will be performed independently by two reviewers. Disagreements that arise between the reviewers will be resolved through discussion, or by involving a third review author as arbiter. Key items to extract include: author, year, country, type of document, objective, features/components of patient decision aids for BRCA mutation carriers, format of patient decision aids, patient and public involvement in decision aid development, efficacy of patient decision aids (where applicable). Data extracted will include specific details about the population, sample size (where applicable), study methods and key findings relevant to the scoping review objectives. The data extraction tool will be modified and revised as required during the process of data extraction. Modifications will be detailed in the full scoping review report. In cases where pertinent information is not reported efforts will be made to contact study authors to request this information.

Analysis of the evidence

Data presentation

Extracted data will be presented in tabular form in a manner that aligns with the objective of this scoping review. A narrative description of the evidence will complement the tabulated results. In addition, a description of how the results answer the objectives and questions of this review will be provided. The review findings and their implications for research and practice in the field of decision-making for female BRCA mutation carriers will be discussed.

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.

ETHICS AND DISSEMINATION

Ethical approval for the conduct of this scoping review is not required as this study will involve secondary analysis of the existing literature. Findings will be published in a peer-reviewed journal and presented at relevant conferences. Findings will be disseminated further through social media platforms.

DISCUSSION

This scoping review will fill an important gap in the literature, as no scoping reviews of evidence relevant to cancer risk management decision-making supports for BRCA mutation carriers currently exist that map the features of patient decision aids developed for these women.

As a scoping review, results from this study will provide a descriptive overview of available evidence. An additional limitation of this review is that we will limit our search to

the published literature, which could potentially result in relevant evidence in the grey literature being overlooked.

Overall, evidence collated through this scoping review may be beneficial for designing new patient decision aids or adapting existing decision aids to support decision-making in terms of cancer risk management for female BRCA mutation carriers.

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Contributors YPH and A-MB led the conceptualisation, design and development of this study. GP conceptualised the scoping review approach. YPH, A-MB, SAM, GP, CS, NB and AD were involved in developing the review questions. SAM drafted the protocol. DM developed the search strategy with input from SAM. EMC provided feedback on the study design from a clinical perspective. All authors reviewed and approved the final version of the manuscript.

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Appendix 1: Search strategy

A Medline (OVID) search strategy^{yes} has^{ve} been provided as an example; the below search strategy^{yes} will be adapted for EMBASE, Cochrane Library, CINAHL and Web of Science.

Decision Aids

Medline (OVID)

1. Decision Support Techniques/
2. (Decision adj3 (support* OR aid* OR navigation OR patient* OR tool*)).ti,ab.
3. or/1-2
4. Genes, BRCA1/
5. BRCA1 Protein/
6. Genes, BRCA2/
7. BRCA2 Protein/
8. (BRCA* or brca* or hereditary breast ovarian cancer syndrome or hereditary breast ovarian cancer syndrome or HBOC).mp.
9. ((BRCA* or brca*) adj5 (carrier* or tumor* or tumour* or gene* or suppress* or protein* or mutat* or alter* or damage* or inherit* or heredit*)).mp.
10. or/4-9
11. 3 AND 10

Appendix 2: Data Extraction Form**Appendix II: Data Extraction Template for studies on decision aids.**

Evidence source Details and Characteristics	
Author	
Year	
Title	
Country	
Aims/Purpose	
Study population and sample size	
Concept	
Context or Setting	
Study design	
Methodology	
Details/Results extracted from source of evidence (in relation to the concept of the scoping review)	
Risk management options addressed in decision aid	
Format of decision aid (paper, web-based etc.)	
Presentation of risks and benefits in decision aid	
Patient & Public Involvement (PPI) in decision aid development	
Other features of decision aid	
Efficacy of decision aid on decision related outcomes (if applicable)	
Author correspondence (Details of correspondence with study authors for additional information or clarification of queries)	
References (Additional relevant articles cited in reference list of article)	

*Modified from JBI methodology guidance for scoping reviews.