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Do doctors and other healthcare professionals know overdiagnosis in screening and how are they dealing with it? A protocol for a mixed methods systematic review

Veerle Piessens ,[1] Stefan Heytens ,[1] Ann Van Den Bruel,[2] Ann Van Hecke,[1] An De Sutter[1]

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[1]Department of Public Health and Primary Care, Faculty of Medicine and Health Sciences, Ghent University, Gent, Belgium
[2]Department of Public Health and Primary Care, KU Leuven, Leuven, Flanders, Belgium

Correspondence to
Dr Veerle Piessens;
veerle.piessens@ugent.be

ABSTRACT

Introduction Overdiagnosis is the diagnosis of a disease that would never have caused any symptom or problem. It is a harmful side effect of screening and may lead to unnecessary treatment, costs and emotional drawbacks. Doctors and other healthcare professionals (HCPs) have the opportunity to mitigate these consequences, not only by informing their patients or the public but also by adjusting screening methods or even by refraining from screening. However, it is unclear to what extent HCPs are fully aware of overdiagnosis and whether it affects their screening decisions. With this systematic review, we aim to synthesise all available research about what HCPs know and think about overdiagnosis, how it affects their position on screening policy and whether they think patients and the public should be informed about it.

Methods and analysis We will systematically search several databases (MEDLINE, Embase, Web of Science, Scopus, CINAHL and PsycArticles) for studies that directly examine HCPs' knowledge and subjective perceptions of overdiagnosis due to health screening, both qualitatively and quantitatively. We will optimise our search by scanning reference and citation lists, contacting experts in the field and hand searching abstracts from the annual conference on 'Preventing Overdiagnosis'. After selection and quality appraisal, we will analyse qualitative and quantitative findings separately in a segregated design for mixed-method reviews. The data will be examined and presented descriptively. If the retrieved studies allow it, we will review them from a constructivist perspective through a critical interpretive synthesis.

Ethics and dissemination For this type of research, no ethical approval is required. Findings from this systematic review will be published in a peer-reviewed journal and presented at the annual congress of 'Preventing Overdiagnosis'. In addition, the results will serve as guidance for further research on this topic.

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STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ This is the first systematic review on the perspective of healthcare professionals (HCPs) on the phenomenon of overdiagnosis in screening.
⇒ It combines both qualitative and quantitative data.
⇒ We use Critical Interpretive Synthesis as a review methodology which encourages the development of overarching explanatory concepts.
⇒ We do not include observations of HCPs' actual behaviour in relation to overdiagnosis, but limit our research to statements made by HCPs about what they know and think.
⇒ If the available primary research is scarce, analysis and results will be limited.

INTRODUCTION

Overdiagnosis is the diagnosis of a disease or medical condition that would never have caused any symptom or problem.[1] It is a significant harm of screening. The screening process could turn a previously healthy person into a patient diagnosed with a disease of which he/she would never have had any symptoms if the screening had not taken place.[2] Although the diagnosis is 'correct' according to current diagnostic standards, the early treatment of this screen-detected condition will not benefit the patient. However, overdiagnosis will harm through unnecessary treatment, fear, 'opportunity costs' and financial burden. Overdiagnosis is an issue in several medical domains, such as mental health, cardiovascular diseases and infectious diseases, but it has only attracted widespread attention since its major impact in cancer screening became apparent.[2]

However, the phenomenon is counterintuitive, not tangible and hard to grasp. It can only become visible on a population level, only rarely at the individual one. Once diagnosed with a screen-detected disease, most patients will receive early treatment. Therefore, they will never know what the natural course of their disease would have been or

whether their diagnosis may have been an overdiagnosed condition. The result is that patients and their treating physicians will never perceive the diagnosis as unnecessary or even harmful. On the contrary, they will feel relieved and believe that they avoided complications or death thanks to the early catch.

Additionally, research to estimate and quantify the problem is challenging, requires long-term follow-up of screening data and high-quality morbidity registers, is prone to bias and confounding factors and outcomes rely highly on the basic assumptions of the researchers.[3–5]

Nevertheless, overdiagnosis is nowadays recognised as a non-neglectable harm of screening. Since the late 20th century, there has been a rise of dissident voices in the dominant discourse of propagating screening. They claim that people should not be 'motivated' to participate in screening programmes but instead informed about benefits and harms and encouraged to make an informed decision that relates well with their preferences and values in health.[6 7]

Since then, multiple studies have tried to assess to what extent this information has reached the general public, especially those who already participated or are about to participate in a screening programme.[8–11] A recent systematic review synthesised all qualitative research on lay people's understanding of overtesting and overdiagnosis.[12] Only a minority of patients seems familiar with the phenomenon, and most people cannot remember receiving information about the risk of overdiagnosis before getting screened. Participants find the concept difficult to understand, are often baffled that it exists and generally prefer to be informed about it. Providing information had mixed effects on the respondents' intention to participate in screening.

It is perhaps not surprising that people are unaware of this problem. After all, in the early years of organised screening programmes, none of the leaflets and websites of the official cancer screening programmes mentioned overdiagnosis as possible harm.[13 14] Although the quality and comprehensiveness of information about screening has improved recently, still not all programmes inform the public about the risk of overdiagnosis.[15] Moreover, a systematic review of patient decision aids (PDA) for cancer screening found that one in five decision aids does not address overdiagnosis at all. However, there was significant variability between screening programmes, with nearly all PDAs for lung and prostate cancer screening addressing overdiagnosis, but only 61% of those for breast cancer screening.[16]

Public awareness and accurate understanding of this intricate problem need widely available information and doctors willing to address this topic in shared decision-making about screening. However, the findings in patients and the general public suggest that doctors do not discuss overdiagnosis with their patients,[11 17] and the research about websites and PDAs[15 16] proves that one in five omits this information.

It is essential to find out what might lie behind this lack of transparent and balanced information. Could it be that doctors and healthcare professionals (HCPs) involved in providing or organising screening are perhaps not fully aware of this problem themselves? Does knowledge of this phenomenon affect their opinion on screening policy? Are they in favour of disclosing this information to the public? Or, do other considerations and concerns (like the fear that it might lower screening uptake) make them decide not to address the issue?

In contrast to the multiple studies in the general public, research on this topic in HCPs is far less prominent. However, a preliminary search found that HCPs attribute different levels of value to the phenomenon, depending on their role, intrapersonal and contextual factors,[18 19] suggesting that apart from knowledge, other factors, such as moral position, emotions, perceived expectations and dominant discourse, might steer doctors and other HCPs in their decisions to disclose information on overdiagnosis. Insights in how HCPs perceive and deal with this problem might provide a substantial knowledge base to explain why the information has not disseminated well in the general public until now.

Research question

The research questions of this mixed methods systematic review are:

- Are doctors and other healthcare providers involved in offering or organising screening aware of overdiagnosis resulting from health screening?
- How important do they think this problem is, and to what extent does it influence their position on screening?
- How do they feel about informing their patients or the public about overdiagnosis?

We translated our research question into a modified population/patient–intervention–comparator–outcome (PICO) format. It is a tool that helps researchers to be as clear and precise as possible about the exact question they want to answer. Because our research question does not involve an intervention or a comparator, we used population–concept of interest–outcomes (PCO), one of the alternative formats, to formulate a research question.[20] PCO stands for:

- Population: doctors and other HCPs offering or organising screening.
- Concept of interest: overdiagnosis as a result of health screening (cancer screening as well as other screenings).
- Outcomes: awareness, knowledge, perception and appreciation of the phenomenon of overdiagnosis and effect on position on screening policy and informing patients or the public about overdiagnosis.

METHODS AND ANALYSIS

This protocol is written according to the Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols (PRISMA-P) guidelines[21] (online supplemental appendix 1).

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Table 1 Study selection criteria

Criteria	Justification
Original research	To avoid duplication, we restrict our review to primary data of original research.
No language restrictions	The researchers master English, French, Dutch and German and assume that these languages will cover most of the available research. However, if our search would retrieve relevant publications in other languages, we will try to translate them.
Research that questions doctors and other healthcare professionals (HCPs) directly on this topic	We look for studies that question HCPs directly on their perception of this topic, for example, through questionnaires or interviews. We will not include studies that only use indirect methods, for example, field observations or analysis of written materials.
Both qualitative, quantitative and mixed methods research	To assess the proportion of HCPs familiar with the phenomenon, we need quantitative research, like surveys, whereas qualitative research is more appropriate for gaining insight into participants' perceptions, considerations and values.
Examining overdiagnosis in screening	The topic overdiagnosis has to be an explicit research theme, although it can be a research topic among other screening aspects. We will not include studies addressing other definitions of overdiagnosis not related to screening[35].
In doctors and HCPs, implicated in offering or organising screening	This review focuses explicitly on doctors and other HCPs. Therefore, publications only dealing with patients' or the general public's perspectives will not be included.
No restrictions in the time frame	We do not set any limitations on the publication date.

Study selection criteria

All study selection criteria and their justification are summarised in table 1.

Search strategy

We will perform systematic searches to retrieve studies from medical, sociological and psychological perspectives in the following databases: MEDLINE (via the PubMed interface), Embase (via the embase.com interface), Web of Science, Scopus, CINAHL and PsycArticles (via ProQuest). We plan to search for quantitative and qualitative studies at once with a dedicated search filter for qualitative research and surveys. A specialised librarian helped develop the search strategy for the different databases (online supplemental appendix 2). We will also scan reference and citation lists of the included articles and contact experts in the field for additional publications. Finally, we will hand search all abstracts of the annual conference on 'Preventing Overdiagnosis'.

Selection and further processing

Three researchers will select eligible studies in a stepwise approach and report the selection process in a PRISMA diagram.

- We will import all references into EndNote reference software (EndNote V.X9.3.3) and Rayyan software for systematic reviews.
- VP and a second reviewer will independently make the first selection for full-text review based on title and abstract. In case of disagreement, we will delay the decision until after the full-text review.
- VP and a second reviewer will independently screen all full texts for eligibility and reasons for exclusion will be recorded. In case of disagreement, AVH will assist in the discussion until consensus is reached.
- All eligible studies will be organised into three subsets for quantitative, qualitative and mixed-method publications during the selection process.
- If a possible eligible study provides insufficient or unclear information for a final decision, we will contact the author for clarification and wait another 2 weeks before deciding on inclusion.
- After quality assessment, we will extract qualitative data into NVivo research software V.1.4.1, and quantitative data will be managed in MS Excel for spreadsheets.

Quality assessment

On the one hand, there is much debate on whether it is appropriate to subject qualitative research to formal quality assessment and, if so, what criteria should be assessed and how researchers should make decisive evaluations. The available quality checklists do not provide formal scoring systems and require substantial interpretive judgement from the researcher.[22] On the other hand, a quality assessment will provide valuable information on the credibility of the results and the study design's appropriateness. Therefore, we decided to perform a formal quality appraisal, mainly for informative reasons, not excluding studies.

For the qualitative studies, we will use the 'Framework for Assessing the Quality of Qualitative Research Evidence'[23] and for quantitative data, a 'Guide for appraising Survey Reports' (online supplemental appendix 3).[24]

VP and FS will independently appraise the included studies. AVH will supervise and assist in discussions in case of disagreements.

Data extraction

For each study, we will complete a predefined and piloted data extraction form, including:

- Title, author and bibliographical details.

- Research question.
- Methodology:
  Study design, setting, sample size, inclusion and exclusion.
  Epistemological position of the authors, if available, and theory for data analysis.
- Results:
  Number and characteristics of participants.
  Main conclusions and author interpretations.
  Strengths and limitations.
  Reviewer comments, if available.

All manuscripts of qualitative studies and the qualitative components of mixed-method studies will be imported in NVivo software to analyse the results section, primary data, developed themes, author interpretation and discussion. We will extract results from quantitative studies and the quantitative results of mixed-method studies to MS Excel for further analysis.

Data analysis

Qualitative studies

This appears to be a relatively untrodden field of research, and our preliminary search learns that the available studies seem scarce. Therefore, we propose a double-layered analysis. First, we believe that it might be of essential informative value to summarise the available research and perform a content analysis in a 'neutral' descriptive way.

However, if enough data were available, a more interpretive analysis from a constructivist perspective would provide a valuable and insightful second layer with the generation of new theories and concepts. We plan to use 'critical interpretive synthesis' (CIS) as our review method. CIS uses an iterative and inductive approach to generate the research question and the emerging themes, concepts and theories. It allows to combine quantitative and qualitative data and, more importantly, to enrich the data with research from adjacent research fields.[25 26]

VP will thoroughly familiarise herself with the available publications and gain insight into each study's position and context separately and into what it adds in relation to the other studies. She will draft a coding tree guided by the principles of a 'line-of-argument (LOA) synthesis'. This method builds on several researchers' work; it is a central technique in meta-ethnography and adapted by the authors who presented CIS as a novel methodology to review qualitative research.[25] LOA analyses the findings in separate layers or lines, where 'first-order constructs' are how participants in the primary studies see and understand certain phenomena, whereas 'second-order constructs' consist of the interpretations, explanations and theories introduced by these studies' researchers.

Additionally, two research team colleagues will independently code a subset of the articles, and after discussing discrepancies and similarities, we will adapt the coding tree where necessary. We will present a first overview of the emerging themes and constructs to the research team and several researchers with different academic backgrounds (psychology, sociology, educational sciences, medicine) and invite them to suggest modifications and relevant theories, to point out inconsistencies or gaps and to pose critical questions. We will then reassess all publications in light of these review sessions' outcomes and repeat this iterative and reflexive process until no new comments or insights emerge.

We might complete the LOA synthesis with 'synthetic constructs' and a 'synthesising argument' if enough data is available. Dixon-Woods *et al*, who introduced CIS as a review method, conceive synthetic constructs as "the result of a transformation of the underlying evidence into a new conceptual form… they are grounded in the evidence, but result from an interpretation of the whole of that evidence, and allow the possibility of several disparate aspects of a phenomenon being unified in a more useful and explanatory way". The authors define that "a synthesising argument integrates evidence from across the studies in the review into a coherent theoretical framework comprising a network of constructs and the relationships between them".[25]

Quantitative studies

Our first exploratory literature search did not reveal any quantitative studies so far. If some were to be found with our thorough search in various databases, we would analyse them separately from the qualitative studies in a segregative design for mixed methods reviews.[27] With this method, we treat qualitative and quantitative findings as conceptually distinct approaches for gaining insight into the phenomenon of interest. Therefore, we will select, appraise, analyse and synthesise qualitative and quantitative data separately and bring both syntheses as complementary outcomes together in a mixed-method synthesis. Depending on the number and the content of the quantitative studies, we will use the appropriate methodology to synthesise them. If the data would allow it, we plan to use meta-analytic techniques to combine data from different sources.[28] If not, we will present the quantitative findings merely descriptively. However, if no quantitative studies are revealed, this will become a single-method systematic review with only qualitative studies.

Strength of evidence

We will apply the Grading of Recommendations Assessment, Development and Evaluation-Confidence in the Evidence from Reviews of Qualitative research (GRADE-CERQual) framework criteria for assessing the strength of evidence of this review's findings. CERQual is a novel approach developed by a subgroup of the GRADE working group to 'assess the extent to which a review finding is a reasonable representation of the phenomenon of interest'.[29] It provides guidance for evaluating each review fin ding on four distinct components: (1) methodological limitations, (2) coherence, (3) adequacy of the data and (4) relevance.[30–33] Additionally, attention is drawn to the 'risk of dissemination bias' as the fifth component of

interest, although without specific guidance on assessing it, as this theme is still a work in progress.[34]

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ORCID iDs
Veerle Piessens http://orcid.org/0000-0002-2305-1762
Stefan Heytens http://orcid.org/0000-0003-1097-4987

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