How do patients and the public understand overtesting and overdiagnosis? A protocol for a thematic meta-synthesis of qualitative research

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ABSTRACT
Introduction Examining patient and public understanding of overtesting and overdiagnosis (OverTD) is vital for reducing the burden of OverTD. Studies from disparate contexts, disciplines and focusing on disparate healthcare issues have examined patient and public understanding of OverTD. A synthesis is needed to bring this literature together, examine common themes, strengthen conclusions and identify gaps. This will help steer further research, policy and practice to improve patient and public understanding of OverTD. The objective of this study is to synthesise qualitative research data about patient and public understanding of OverTD.

Methods and analysis A thematic meta-synthesis will be used to synthesise primary qualitative research and qualitative components of primary mixed-methods research about patient and public understanding of OverTD. Studies published in English will be included. These will be identified using systematic searches from inception to March 2020 in the Scopus, CINAHL, PsycINFO and MEDLINE databases. Studies that satisfy eligibility criteria will be assessed for methodological quality using the Critical Appraisal Skills Programme (CASP) checklist. Thematic meta-synthesis will comprise three stages: (1) line-by-line coding; (2) generation of descriptive themes and (3) generation of analytic themes. Confidence in the synthesis findings will be assessed using the Grading of Recommendations Assessment, Development and Evaluation Confidence in Evidence (GRADE CERQual) approach. A summary of GRADE CERQual results will be presented alongside the key themes. Study eligibility screening, data extraction, analysis and the CASP and GRADE CERQual assessments will be undertaken independently by two review authors.

Ethics and dissemination Ethics approval is not required for this secondary analysis of published data. The results will be disseminated in peer-reviewed journals and may be presented in conference papers and elsewhere.

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INTRODUCTION
The high prevalence of overtesting, overdiagnosis and overtreatment across a range of health conditions is a global challenge.1 Overtesting is when diagnostic tests that are not indicated are used.2 It can lead to overdiagnosis, which is when a diagnosis is made according to professional standards, but when it is unlikely to benefit the patient.4 Overtesting and overdiagnosis (OverTD) can lead to overtreatment,2,5 which is treatment that does more harm than good.6

It is important to reduce overtesting, overdiagnosis and overtreatment.7,8 Overtesting can lead to harms including unnecessary invasive procedures, false positives and misdiagnoses.3 Overdiagnosis can lead to unwanted behavioural and psychological responses in patients, such as reduced participation in usual activities,9 stress and anxiety.10,11 A diagnosis primes patients and physicians to commence treatment, even for benign conditions.8 Overtreatment can lead to patient suffering, treatment-related complications, loss of quality of life, lost productivity and other burdens.6,12 Medical overuse is massively costly to healthcare systems and to patients and their families,2,13,14 and must be reduced to maintain healthcare system sustainability.15 Improving patient and public understanding of OverTD is key to reducing their incidence as well as the incidence of...
Both patients and the public need to be aware of OverTD, as people regularly transition between being in and out of medical care, and their medical decision making is informed by beliefs that are formed and reformed across contexts. Presently, patients and the public often drive medical overuse. Some patients and members of the public tend to over-rely on tests and diagnoses, overestimating their benefits, underestimating their risks, as they cope with uncertainty poorly. Few are aware that overtreatment or overdiagnosis occurs, and those who are often find the phenomena difficult to understand. Research suggests that patient outcomes would be improved if they understood OverTD better. Patients with better knowledge about OverTD make more appropriate screening and treatment decisions. Patient knowledge also influences the tests and treatments prescribed by the medical practitioners, who in some cases overuse medical interventions. Patients and the public want to be informed about OverTD, and need to understand both risks and benefits of medical interventions in order to participate in shared decision making.

Research is increasingly examining patient and public understanding of OverTD. Patient and public understandings of OverTD have been surveyed qualitatively, and examined in relation to a range of conditions and in multiple contexts. Researchers have studied the challenges of communicating about OverTD to the general public, as well as to particular patient groups, such as patients with low health literacy. Strategies are being developed to overcome these communication challenges. They include the development of decision aids, which inform patients about the risks as well as benefits of particular medical interventions, such as breast cancer screening, and assist them in making evidence-backed healthcare decisions. Other research has focused on refining patient educational tools. This includes studying how different concepts of OverTD resonate with patients and the public, the effects of information about overdiagnosis on patient screening decisions, and studying patients’ understandings of their own diagnoses. The use of mass media to reduce OverTD has also been studied, such as how media narratives can influence cancer screening decisions or promote better management of back pain.

Despite progress in research, important gaps in knowledge remain. First, existing studies are scattered across disciplines, contexts and focus on disparate medical conditions. It is difficult to appraise the overall state of research or glean its collective insights. Second, while it is known that patients and the public find OverTD unintuitive, little is known about why. A meta-synthesis of qualitative data from research examining patient and public understanding of OverTD will help address these gaps. It will systematise insights from disparate disciplines, contexts and topic areas by identifying descriptive themes in the body of literature. The synthesis will also identify analytic themes about the reasons for poor public and patient understanding of OverTD. These findings will inform future research by highlighting priority areas for further enquiry. An increased understanding about why patients and the public struggle to understand OverTD may inform the development of educational interventions and other practice to improve their understanding.

OBJECTIVE

The objective of this study is to synthesise data from qualitative research on patient and public understanding of OverTD.

METHODS

Thematic meta-synthesis will be used to examine primary qualitative research and qualitative components of mixed-methods research about patient and public understanding of OverTD.

The protocol is presented in accordance with the Preferred Reporting Items for Systematic review and Meta-Analysis Protocols (PRISMA-P) checklist (available in online supplementary appendix 1).

Study selection criteria

Study selection criteria and their rationale are described in table 1.

Search methods

The search process will comprise first an informal scoping stage to develop search strategies, and then a formal main stage to identify and collate eligible studies. The main stage will identify English language studies indexed in four databases from inception until March 2020.

The scoping stage will be exploratory. Its aims are to become familiar with the literature, refine search parameters, identify MeSH terms and keywords to test the preliminary search strategy.

The main stage will comprise the formal literature search. It will be informed by the scoping stage, by search strategy guidelines from the Cochrane Collaboration, and guidelines for optimising database searches for medical qualitative research and guidelines for searching the individual databases used, such as for MEDLINE and PsycINFO. Search filters will be identified for each of the inclusion criteria. A subject librarian will contribute to the development of the search strategy.

The following databases will be used: Scopus, CINAHL, MEDLINE and PsycINFO. These were chosen because they are most likely to index studies about patient and public understanding of OverTD: social research (Scopus); medicine/public health/health communication research (MEDLINE, CINAHL); psychological research (PsycINFO) and generalist fields (Scopus). Database selection was also informed by research showing that Scopus, MEDLINE and CINAHL searches retrieve some of the largest numbers of qualitative health studies, and the largest number of qualitative health studies not listed...
Despite indexing relatively few unique studies, it was predated by earlier terms, and it is important to capture these earlier studies. Synonymous concepts to overtesting and overdiagnosis will be included, such as ‘over-detection’ and ‘overuse of diagnostic testing’. The term ‘overdiagnosis’ was popularised relatively recently, but it was predated by earlier terms, and it is important to capture these earlier studies. Studies which did not explicitly aim to understand how to inform patients or the public about overdiagnosis are outside the scope of this synthesis and will be excluded. Studies that only address overtreatment and not overtesting or overdiagnosis will also be excluded.

Studies examining understanding

The synthesis will examine understanding among both patients and the public. People regularly transition between being one or the other, and make diagnostic and screening decisions drawing on understanding they developed over time and in either role. So, it is appropriate to examine understanding of OD among both groups. It will be distinguished whether studies are about patients, the public or both. People have a differing engagement with health decision making when they are patients or the public. To account for this, synthesis results for each group will be compared, and important intergroup differences will be considered in study outcomes.

Among patients and/or the public

The synthesis will examine understanding among both patients and the public. People regularly transition between being one or the other, and make diagnostic and screening decisions drawing on understanding they developed over time and in either role. So, it is appropriate to examine understanding of OD among both groups. It will be distinguished whether studies are about patients, the public or both. People have a differing engagement with health decision making when they are patients or the public. To account for this, synthesis results for each group will be compared, and important intergroup differences will be considered in study outcomes.

Of overtesting and/or overdiagnosis (OverTD)

Studies about both overtesting (OT) and overdiagnosis (OD) will be included, as both are deeply interlinked and underpinned by common broader patient ideas about healthcare. However, understandings of OT and OD may differ. To account for this, studies will be classified based on whether they examine OT, OD or both. The synthesis results will be compared by these classifications, and important differences will be accounted for.

Synonymous concepts to overtesting and overdiagnosis will be included, such as ‘over-detection’ and ‘overuse of diagnostic testing’. The term ‘overdiagnosis’ was popularised relatively recently, but it was predated by earlier terms, and it is important to capture these earlier studies. Studies which did not explicitly aim to understand how to inform patients or the public about OD are outside the scope of this synthesis and will be excluded. Studies that only address overtreatment and not overtesting or overdiagnosis will also be excluded.

Either qualitative or mixed-method study design

Mixed-methods studies will be included where their qualitative components can still be examined in the thematic meta-synthesis. Quantitative components of mixed-method studies will be excluded, as will studies where it is not possible to differentiate between quantitative and qualitative components of analysis.

Published in the English language

Only English language studies will be included, as the authors are English speakers, and relying on translations of non-English studies could introduce inaccuracies into the analysis.

Published in any year

There will be no date restrictions: older insights may still be relevant.

Conducted in any setting

There will be no setting restrictions: studies from all settings may potentially contain transferable insights about patient and public understandings of OD.

Focusing on the general concepts of OD and/or in relation to any condition/s or interventions

While patient and public understanding of OD may differ depending on medical conditions, there may be underlying themes across conditions, so it is relevant to include studies relating to any conditions. The condition/s which a study focuses on will be noted. Study themes will be compared by conditions in analysis if the sample characteristics make this viable.

Selection of studies

Study selection will comprise the following steps:
1. All study records identified using the search strategy will be extracted with a PDF of the study manuscript into EndNote reference management software.
2. Duplicate studies will be removed from the data.
3. Study titles and abstracts will be screened for eligibility by two authors (TR and RH) working independently. Eligible studies and studies where eligibility cannot be clearly determined from the abstract and title will be included for full-text review.
4. Full texts will be independently read and examined for eligibility by TR and RH using a standardised form. Ineligible studies will be screened out, and the reason for exclusion recorded. Eligible studies will be included in the analysis. Where the two authors cannot agree on eligibility after discussion, a third author (DAO) will judge whether the study should be included.
5. The final sample of full text studies will be extracted to NVivo research software. The screening process will be reported in a PRISMA diagram.

Data extraction
The complete study manuscript will be extracted into NVivo. Analysis will be undertaken on the Results sections of manuscripts, including themes, subthemes and primary data as reported in the manuscripts, such as participant quotes. Primary data included in tables and sections of manuscripts may also be analysed. For each study, a standardised data collection form will be completed to capture:

- Study details: authors, year of publication, journal in which study was published.
- Research question/s.
- Participants: sample size, demographic characteristics, whether they are patients and/or the public, methods of participant recruitment and selection.
- Setting: type/s of healthcare and/or conditions the study focused on, whether the study examined over-testing and/or overdiagnosis, country where study was completed, whether study was in urban or rural settings.
- Method of data collection (such as interview or survey).
- Method of data analysis (such as narrative analysis or discourse analysis).

These details will be added as classifying information to the extracted full text studies in NVivo.

Assessment of quality of included studies
The Critical Appraisal Skills Programme (CASP) qualitative checklist56 will be used to systematically examine the reliability, validity and usefulness of individual studies in the synthesis. The 10-item checklist comprises nine fixed-response questions that can be answered: yes/can’t tell/no (‘yes’ indicates a positive score), and one text-response question. Two authors (TR and RH) will independently complete the CASP checklist for each study, and any disagreements in scoring will be resolved by a third author (DAO). A summary of CASP checklist results will be reported as a table and interpreted in text.

Synthesis and analysis
A thematic meta-synthesis of the Results sections of manuscripts will be undertaken. Analysis will comprise three main stages:55 first, line-by-line coding; next, descriptive thematic development, and finally; analytical theme development.

The thematic meta-synthesis method was chosen for several reasons. It fits the gaps this research responds to: the descriptive phase will address the need to systematise insights from disparate disciplines, contexts and topic areas, while the analytical phase is an interpretive tool with which synthesised studies can be re-examined to study why patients and the public find OverTD so difficult to understand. Furthermore, thematic analysis is suitable for handling data from disparate contexts,58 which fits this synthesis where included studies are likely to be heterogeneous. Finally, thematic meta-synthesis is particularly suited to informing policy and practice,59 which is an important consideration for this research. The synthesis assumes an objective idealist epistemic position. The synthesised studies are considered to convey something about reality, but this reality is conveyed through a subjective lens.60 This is also assumed for the findings of this synthesis.

The first stage of analysis will be line-by-line coding. Authors will familiarise themselves with the data. TR will inductively generate initial codes for ideas in the data, coding over several iterations until no new codes are needed to capture ideas. Single data fragments can be assigned multiple codes. Once TR is satisfied with the code frame, he will code the whole dataset, checking coding for data coverage and refining it as necessary. A second author (RH) will check a randomly selected sample of 10% of coded data for coding accuracy. A disagreement score will be calculated, and disagreements discussed and resolved, drawing on the wider team if required. An agreement score of 85% or higher will be targeted.61 If the agreement score is low, reasons for this will be investigated, and line-by-line coding may need to be repeated.

The second stage of analysis will be the development of descriptive themes to organise existing ideas in the data. TR and RH will independently organise individual codes into broader themes. The two authors will then cooperate to develop one set of common descriptive themes, discussing them with the wider author group. Themes will be checked for data coverage and internal homogeneity.62 External heterogeneity will not be assessed, as this is problematic where individual data can be multi-coded. Themes will be revised until their fit with data is optimised.

The third stage of analysis will be the development of analytical themes capturing the barriers and enablers to patient and public understanding of OverTD. This stage will be interpretative and will seek to generate new ideas.63 TR and RH will independently re-examine the data organised into descriptive themes to infer what the barriers and enablers to understanding OverTD are.64 This phase will rely on the authors’ subjectivities, and the authors will take a reflexive approach to minimise problems in interpretation and improve transparency in analysis.65 TR and RH will meet to compare their analytical themes. As part of researcher reflexivity, they will discuss the factors that led to their interpretations, including their assumptions, logical inferences and how their interpretations may have been shaped by the predetermined research aims. Researcher reflexivity will also be addressed in peer-reviewed publications resulting from this research, including consideration about the ways in which the authors’ own positions could have influenced the study design, analysis and the interpretation of findings. TR and RH will determine the analytical themes,
which will be discussed and finalised with the wider author group.

Descriptive and analytical thematic results will be compared across a range of classifying variables, such as whether data are from studies about patients/the public/both, and whether data are from studies investigating understanding of overtesting/overdiagnosis/both. Notable comparative differences will be reported in the Results. Descriptive and analytical themes will be tabulated and paired with exemplary data fragments. A separate table will display how the data from each study is represented in the coding.

Assessment of confidence in findings
The Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach\(^6\) will be used to assess confidence in the analytic synthesis results. GRADE-CERQual is used to consider four factors about studies contributing to review findings: (1) methodological limitations; (2) relevance; (3) adequacy of supporting data and (4) coherence. The overall confidence in each review finding (ie, for each theme generated) will be graded as: high, moderate, low or very low. GRADE-CERQual assessments will be undertaken independently by two authors (TR and RH). Any disagreements will be discussed until consensus is achieved. Review findings, the confidence judgement for each finding and an explanation of the judgement will be presented in a Summary of Qualitative Findings table.

Assessment of methodological limitations
Methodological limitations in the synthesis will be judged based on the aggregated CASP checklist results for all included studies (described earlier).

Assessment of relevance
Relevance is ‘the extent to which the body of data from the primary studies supporting a review finding is applicable to the context specified in the review question’ (Noyes et al, p53).\(^6\) Across synthesised studies contributing to each review finding, we will consider the years of publication, settings in which studies were conducted, target audiences and specificity of the findings. These will determine how relevant the body of synthesised studies is for developing knowledge about contemporary patient and public understanding of OverTD in general.

Assessment of adequacy
Adequacy is the quantity and richness of data contributing to a review finding.\(^6\) Quantity is defined as the number of studies or data fragments supporting a theme. Richness is defined as the extent to which themes are supported by detailed, qualitative descriptions. Both parameters will be considered to judge the adequacy of data for supporting each theme in the synthesis results.

Assessment of coherence
Coherence is ‘how clear and cogent the fit is between the data from the primary studies and a review finding that synthesises that data’ (Colvin et al, p35).\(^6\) To examine coherence, the synthesis themes will be compared against the results of individual synthesised studies, examining the extent to which the synthesis findings align with individual study findings.

Patient and public involvement
A health consumer advocate from the Consumer’s Health Forum of Australia was consulted in the development of this protocol. They will advise on the interpretation of the synthesis results.

RESULTS
The Results will comprise two subsections:
1. The sample profile, describing classifying information about the synthesised studies.
2. The thematic meta-synthesis results. Both descriptive and analytical themes will be reported. The descriptive themes will form a minor part of the Results, summarised in a table and briefly interpreted in text. The analytical themes will form a main part of the Results, with all major analytical themes tabulated, described in text and paired with exemplary data fragments and GRADE-CERQual assessment findings. The meta-synthesis will be reported in accordance with the enhancing transparency in reporting the synthesis of qualitative research statement.\(^7\)

ETHICS AND DISSEMINATION
Ethics approval is not required for this secondary analysis of published data. The findings may be disseminated in peer-reviewed publications, conference papers and elsewhere.

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Contributors TR, DAO and RB conceived the study and wrote the first draft of the protocol. RH, RT, KM and SC contributed to refining the protocol design and preparing subsequent protocol drafts. All authors approved the submitted protocol and are accountable for its content.

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