

Further exploration of dissemination bias in qualitative research required to facilitate assessment within qualitative evidence syntheses

Toews, Ingrid; Booth, Andrew; Berg, Rigmor C.; Lewin, Simon; Glenton, Claire; Munthe-Kaas, Heather M.; Noyes, Jane; Schroter, Sara; Meerpohl, Joerg M.

Journal of Clinical Epidemiology

DOI:

[10.1016/j.jclinepi.2017.04.010](https://doi.org/10.1016/j.jclinepi.2017.04.010)

Published: 01/08/2017

Peer reviewed version

[Cyswllt i'r cyhoeddiad / Link to publication](#)

Dyfyniad o'r fersiwn a gyhoeddwyd / Citation for published version (APA):

Toews, I., Booth, A., Berg, R. C., Lewin, S., Glenton, C., Munthe-Kaas, H. M., Noyes, J., Schroter, S., & Meerpohl, J. M. (2017). Further exploration of dissemination bias in qualitative research required to facilitate assessment within qualitative evidence syntheses. *Journal of Clinical Epidemiology*, 88(August), 133-139. <https://doi.org/10.1016/j.jclinepi.2017.04.010>

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Dissemination Bias in Qualitative Research: conceptual considerations

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1. Abstract

When the findings of relevant studies are not disseminated, and are therefore not accessible, data within evidence syntheses may be considered inadequate. In addition, bias will result if disseminated studies and findings differ systematically from non-disseminated studies and findings. Such bias could occur due to several mechanisms and is called *dissemination bias*. We have defined dissemination bias in qualitative research as a systematic distortion of the phenomenon of interest due to selective dissemination of studies or findings of studies. The presence of such bias could impact on our confidence in findings from qualitative evidence syntheses.

Objective

To conceptualise and discuss dissemination bias in qualitative research.

Results

It is likely that the mechanisms leading to dissemination bias in quantitative research, including time lag, language, grey literature publication and truncation, also contribute to dissemination bias in qualitative research. These conceptual considerations have informed the development of a research agenda.

Conclusion

Further exploration of dissemination bias in qualitative research is needed, including the extent of non-dissemination and dissemination bias, and how to assess dissemination bias within qualitative evidence syntheses. We also need to consider the mechanisms through which dissemination bias in qualitative research could occur in order to explore approaches for reducing it.

Keywords: Dissemination bias, publication bias, qualitative research, qualitative evidence synthesis, systematic review, non-dissemination

Running title: Dissemination bias in qualitative research

Word count: 2817

2. Qualitative research in health and social care: what is it used for?

Qualitative research aims to understand people's experiences and perspectives, and can influence how health care and social interventions are conceptualized, developed and implemented.

Qualitative research is well suited to understanding factors that affect the acceptability and feasibility of interventions, as well as implementation fidelity [1]. Qualitative research can also explore how and why interventions, and different intervention components, might lead to specific outcomes, and contribute to theory development and the creation of explanatory hypotheses. Findings from qualitative research can inform decisions on the use of evidence-based health and social care interventions and contribute to policy decisions in these fields. Decision-makers in health and social care are therefore increasingly using qualitative evidence alongside other forms of evidence to inform decisions [2-6].

2.1. Qualitative evidence synthesis

Qualitative evidence is increasingly brought together in qualitative evidence syntheses [7].

Qualitative evidence syntheses provide an overview of people's views, perspectives and experiences of a particular phenomenon. A qualitative evidence synthesis analyses and further interprets evidence from individual qualitative research studies addressing similar research questions or phenomena of interest. There are over 20 methods of qualitative evidence syntheses to select from and new guidance has been published on selecting the most appropriate method for a specific context [8]. Qualitative evidence syntheses are designed to create new understanding of phenomena of interest, generate theoretical and conceptual models, identify research gaps, and provide evidence for the development, implementation and evaluation of interventions. These syntheses can be used when developing fields of research, for instance by contributing to empirical generalizations [9]. They can also be used to complement systematic reviews of quantitative evidence as part of clinical and health system decision-making processes. For instance, qualitative evidence syntheses are increasingly used in the development of clinical and health system guidelines [6, 10]. Here, they can help define the scope of the guideline, including detailing the populations, interventions, comparisons and outcomes on which each guideline question should focus [11]. They can help assess the acceptability of the intervention to key stakeholders as well as the intervention's feasibility [11]. They can also ascertain how different stakeholders and population groups value different outcomes and help ensure that the voices of important and sometimes underrepresented groups of people are heard. Lastly, they can identify implementation considerations for interventions that a guideline recommends (see Textbox 1) [11].

Accordingly, systematic review organisations such as Cochrane, NICE Public Health Guidelines, the EPPI Centre, Joanna Briggs, and UK funders such as the National Institute for Health Research (NIHR), increasingly value syntheses of qualitative health and social care research [3]. A challenge to using evidence from qualitative research, however, has been assessing and communicating how much confidence decision makers should have in the review findings.

The benefit of clinical safety checklists for patient safety has been demonstrated in a large, prospective study [12], but the uptake of checklists in clinical practice is slow [13]. In order to find out why clinical checklists are not regularly and successfully used in clinical settings, Bergs et al. [14] synthesized 18 qualitative studies in a qualitative evidence synthesis aiming to identify the barriers and facilitators to implementing clinical checklists. The evidence suggests that staff perceptions of checklists play a major role, with some staff being reluctant to use a checklist because they doubt its evidence base. Staff's perceptions of patient safety also

influenced the use of checklists: for example, nurses would not read out checklist items that might cause distress to patients. Lastly, workflow adjustments, such as changing the workflow of the involved staff, were identified as a barrier to implementing clinical checklists. The authors also highlighted aspects which could improve the use and success of clinical safety checklists.

Textbox 1: Example of how findings from a qualitative evidence synthesis can inform understanding of the factors affecting implementation of a health care intervention

2.2. Assessing confidence in findings from qualitative evidence syntheses

The Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach was originally designed to assess how much confidence to place in findings from reviews of quantitative studies of the effectiveness of interventions. The GRADE Working Group has since expanded its remit, and now includes approaches for assessing confidence in a range of different types of evidence. The GRADE-CERQual (Confidence in the Evidence from Reviews of Qualitative research: www.cerqual.org) approach was specifically developed for findings from syntheses of qualitative evidence [15]. According to the GRADE-CERQual approach, review authors and/or end users may have less confidence in a review finding if there are concerns regarding: methodological limitations of the studies contributing to the review finding, relevance of the included studies to the review question, coherence of the review finding, or adequacy of data supporting the review finding. In the development of this approach, however, there has also been much discussion on the degree to which dissemination bias might influence our confidence in a review finding. An assessment of dissemination bias is not currently part of the GRADE-CERQual approach in recognition of the very limited empirical evidence on its extent in qualitative research and its impact on findings of qualitative evidence syntheses. In addition, we have little knowledge on ways of detecting such bias. Further research is needed to establish the extent of non-dissemination and related dissemination bias in qualitative research, to determine how dissemination bias can be identified and to assess its impact on findings from qualitative evidence syntheses.

In this paper we conceptualise and discuss the issue of dissemination bias in qualitative research. While evidence on dissemination bias in qualitative research is scarce, the phenomenon has been investigated intensively in the quantitative research environment. Our discourse about the causes and consequences of dissemination bias in qualitative research was therefore informed by reflecting on the available evidence from the quantitative research arena. We will highlight how mechanisms that cause dissemination bias in quantitative research might also play a role in qualitative research.

3. What is dissemination bias?

Non-dissemination and irretrievability of studies is first and foremost unethical and a waste of resources [16]. In the case of systematic reviews of quantitative studies with meta-analyses, such non-dissemination might lead to inadequacy of data, which, in turn, might lead to imprecision of pooled effect estimates. Where non-dissemination is systematic rather than random – in other words, if disseminated studies and findings differ systematically from non-disseminated studies and findings – this will distort review findings and cause dissemination bias.

Dissemination bias therefore describes the systematic error that occurs from non-dissemination of studies and findings. The key underlying concern is the (non-) dissemination of studies due to the nature of their content and message [17]. In the context of this paper, we discuss dissemination bias resulting from the non-dissemination of studies and findings due to their content. We do not use the

term to describe the effects of the non-dissemination of studies and findings due to other factors, such as the study's design or the population under investigation etc. For example, the extent to which journals decide to publish qualitative research in general and the editorial policies that apply are not our primary area of interest [18] nor do we categorise this as dissemination bias in qualitative research. However, we are aware that some journals are less likely to publish qualitative research than quantitative research and that journals might not have specific publication guidelines and policies for qualitative research. This in turn may contribute to non- or incomplete dissemination of qualitative studies [19]. Given that these mechanisms would affect any qualitative study irrespective of the nature of its findings we do not consider these mechanisms as contributing to dissemination bias within qualitative research itself as there are a high number of journals that readily publish qualitative research.

3.1. Dissemination bias in *quantitative* research: causes

While the systematic non-dissemination of research has been commonly referred to as *publication bias*, the term *dissemination bias* is becoming more commonly used as this allows us to acknowledge the underlying mechanisms more comprehensively [20]. Three issues are particularly relevant when discussing the term dissemination bias. Firstly, while scientific evidence is usually made available in journal publications, other dissemination channels such as study registries or online data repositories are becoming increasingly important [17]. Secondly, the term *dissemination bias* describes both the non-dissemination of an entire study (non-publication) as well as the selective non-dissemination of individual results (selective reporting). In addition to selective reporting of results, for example from individual participants of the study, selective outcome reporting describes the non-reporting of findings related to entire outcomes. The most dominant mechanisms underlying the selective dissemination of quantitative studies and results, and resulting in dissemination bias, are described in Table 1 [17].

Thirdly, *dissemination bias* also covers the practice of duplicate publication. *Duplicate publication* is an aspect of dissemination that describes the practice of producing multiple publications reporting the same findings from a single study [21]. Outcomes and results might, sometimes unreasonably, be split up into several reports. Duplicate publications are not always clearly discernible and might be confused for reports of different studies. As a consequence, the same study results might be included multiple times in meta-analyses and thus bias the overall effect estimate.

3.2. Non-Dissemination and Dissemination bias in *quantitative* research: prevalence and impact

Clear empirical evidence demonstrates that a large proportion of quantitative studies in clinical research remain unpublished after completion [22-24]. For example, in a systematic review of methodological research projects including randomised controlled trials and other interventional and observational studies from general medicine, different medical specialties and epidemiology, Schmucker et al. [25] found that only half of all studies (46.2%) approved by research ethics committees were published. The bias resulting from such non-dissemination has been found to have consequences for the evidence-base for clinical, and political, decision-making [26]. Dissemination bias was repeatedly found to lead to an overestimation of the reported effects of health interventions because statistically significant and positive results had an increased probability of getting published [16, 25, 27]. For example, the drug *reboxetine* was approved and consequently used as a safe and effective treatment for depression. A recent systematic review [28], however,

revealed that the beneficial effect of *reboxetine* was based on selected patient data. Data for 74% of the patients were not published in the primary studies. The review authors repeated the analyses with published and unpublished primary data and found that *reboxetine* was not more effective than placebo and caused more adverse events. This demonstrates that dissemination bias is a threat to decisions in health and health care and consequently to the health and safety of individuals.

3.3. Non-Dissemination and Dissemination bias in *qualitative* research

We have previously defined dissemination bias in qualitative research as a systematic distortion of the phenomenon of interest due to selective dissemination of studies or findings of studies [15].

Although little empirical research is available on either the extent of non-dissemination of qualitative research or on the extent of dissemination bias in this domain, it is very likely to be present. In clinical effectiveness research, the most common concern about dissemination effects is that the benefits or harms of a clinical intervention will be over- or underestimated [25, 26]. This distinction between “positive” and “negative” findings is unhelpful in qualitative research which focuses on the varying views and experiences of participants regarding a health issue or intervention, and not on the direction of the overall effect. Dissemination bias in qualitative research therefore cannot be articulated within a discourse of outcome, but rather needs to be viewed in relation to the complete and accurate representation of the phenomenon of interest. Consequently, it is challenging to explore whether particular types of content or types of findings or conclusions from qualitative studies are more or less likely to be published.

In qualitative evidence syntheses, omission of data may result in the loss of a particular perspective altogether or may lead to a less nuanced interpretation of the phenomenon. As a consequence, we may place more confidence in a finding than we should, or a synthesis may be limited by the omission of findings. Decision-making might therefore be hampered by an incomplete evidence base or flawed assessments of confidence in the evidence. However, because we are only now starting to explore dissemination bias in qualitative research, we can only speculate about its consequences for the body of qualitative evidence and for decision making.

3.4. Dissemination bias in qualitative research: possible causes and consequences

Based on what is known about dissemination bias in quantitative research, it may be reasonable to assume that the same mechanisms lead to dissemination bias in qualitative research. Table 1 presents a description of how time lag, language, grey literature and truncation bias may occur in qualitative research and impact on qualitative evidence syntheses. Additional factors, observed in quantitative research, which may also lead to dissemination bias in qualitative research include findings that oppose current beliefs, findings that may be viewed as unpopular by opinion leaders, findings that are discordant with the stance of those funding the research, and findings that have cost or other implications that are not seen as feasible [17].

Table 1: Biases and underlying mechanisms identified to play a role in quantitative research that might influence selective reporting of studies and findings in *qualitative* research

Bias	Description	Causes	Impact on systematic reviews of quantitative results	Impact on evidence syntheses of qualitative findings
Time-lag	Striking findings are	Authors might pursue	Relevant and new	Evidence syntheses

bias	published sooner after completion of a study than less noteworthy results [29]	the publication of certain findings more vigorously so that more striking findings or findings supporting a popular view are published sooner; editors might prioritize the publication of findings that they consider more newsworthy	results of no, little or even harmful effects might not be available at a given point in time [30]	might be lacking up to date, relevant studies that report a wider variety of findings
Language bias	Striking findings from a study might be more likely to be published in the English language in an international journal. This, in turn, might increase the retrievability and accessibility of these findings, compared to those that were seen as less striking	Expressing small nuances of speech and language that might add to the correct understanding of the phenomenon of interest is more challenging for researchers who are not writing in their first language [31]; less striking findings are probably more likely to be published in journals publishing in the native language and national context of the researchers, for which the reports are more difficult to access	Studies in languages other than English are harder to identify and retrieve [21]	Studies in languages other than English are harder to retrieve and identify, and therefore some findings may be less represented in evidence syntheses. It is prohibitively expensive to translate and back translate the study to ensure that conceptual meaning is not lost in translation.
Grey literature bias	Increased publication of less noteworthy study findings [29] in outlets other than peer reviewed journals	Limitations on article length can be overly restrictive for the full reporting of qualitative research; many researchers publish their findings in reports, on websites and social media, and in newsletters [31]; Qualitative research is frequently conducted outside of	Studies showing less striking results are not indexed in major scientific databases and harder to be retrieved and included in systematic reviews	Grey literature is not indexed in major scientific databases and harder to retrieve for evidence syntheses.

		an academic context and published routinely in organisational grey literature reports. Small effect sizes are more likely to be published in grey literature [26]		
Truncation bias	Studies that are published in outlets such as reports, books, theses and dissertations might be more likely to report fuller findings than those where an arbitrary word limit is prescribed [32]	The artificial word limit of scientific journals is often too restrictive for the full reporting of qualitative research [31]; researchers often choose to employ books and reports as a medium for communication as these allow longer articles and a wider variety of formats than journal articles [33]	Studies published in outlets with strict manuscript word limits, such as in scientific journals, might contain incomplete reporting of findings	Literature searches confined to journal articles may lead to 'truncation' bias as the full details and findings of a qualitative study may not be published in a journal article

3.5. Non-dissemination and dissemination bias in qualitative research: empirical evidence

To date, very few studies on non-dissemination and dissemination bias in qualitative research have been conducted and, more generally, meta-research on qualitative research is rare. This scarcity of research on dissemination bias may be a consequence of the relative novelty of qualitative evidence synthesis when compared to its quantitative counterparts, and highlights the need for more research to investigate the issue comprehensively. The research priorities outlined below focus on non-dissemination of qualitative research as a first step in exploring the issue of dissemination bias. This research will also contribute to developing a broader research agenda on dissemination bias in qualitative research.

One of the few studies on this topic followed a cohort of 224 qualitative studies presented at a single medical sociology conference to assess what proportion of these studies remained unpublished in the following two years [34]. The study searched for subsequent publication of the studies in relevant databases and by contacting the study authors. They found that less than half (44%) of the studies had been published up to seven years after publication. Reporting quality in the abstracts was positively related to the subsequent publication of the study. The authors concluded that the extent of non-publication of qualitative studies is similar to that for quantitative studies.

A second study, an explorative cross-sectional survey of authors of qualitative studies, peer reviewers and editors of scientific journals, demonstrated that non-dissemination in qualitative

research is substantial, and that several stakeholder groups play an important role in the ‘non-dissemination’ pathway [31]. Non-dissemination, and the dissemination bias that may result, was not seen by participants as merely a theoretical problem but was seen as having important impacts on health and social care research, practice and policy. Over half of researchers reported that one or more of their qualitative studies had not been published in a peer-reviewed journal (62%) or in another publicly accessible format (52%). Around one third reported that important individual findings were missing in one or more of their published reports.

4. Research priorities

The increasing use of qualitative research findings in clinical guidelines and health and social care decision-making emphasises the need to explore further the extent and implications of non-dissemination, and related dissemination bias, in qualitative research. As a starting point, we need to develop an evidence-informed taxonomy of the different routes through which dissemination bias may arise in the context of qualitative studies. A comprehensive mapping review that can inform this taxonomy is currently prepared. It describes the quantity and characteristics of papers reporting non-dissemination and dissemination bias in qualitative research and sets out to describe and categorize the mechanisms that contribute to dissemination bias. Further studies are also needed across a range of qualitative research domains, including different disciplines (sociology, anthropology etc.) and areas of research (health systems research, social welfare research etc.), of the conversion rate of funded projects, abstracts or submissions into publicly accessible dissemination formats. In addition, we need to explore the causes of dissemination bias in qualitative research – why some qualitative studies or findings are not published or disseminated – and find feasible ways for decreasing and preventing dissemination bias.

Distortions in the results of reviews of quantitative effectiveness evidence can, under certain circumstances, be detected and adjusted for by statistical methods, so increasing the validity of the overall estimate of effect [35]. Currently, no established methods or guidance exist on how to assess whether, and to what extent, dissemination bias might be present in the findings of qualitative evidence syntheses. We also lack guidance on what precautions can be taken when interpreting the findings of these syntheses. Further research on these questions is needed, as we discuss elsewhere [36]. People interested in this topic are encouraged to join the GRADE-CERQual DissQuS (Dissemination bias in Qualitative Synthesis) sub-group and contribute to taking forward this area of research.

What is new: Key findings: Evidence on dissemination bias in qualitative research is scarce. Plausible biases that might affect the full dissemination of qualitative studies include time-lag bias, language bias, grey literature bias and truncation bias.

What this adds to what is known: Given the paucity of literature on dissemination bias in qualitative research, several sub-biases are proposed to help conceptualize dissemination bias in qualitative research. Based on conceptual considerations a research agenda has been developed.

What is the implication: More evidence on the extent of dissemination bias in qualitative research and its effects is needed; and we need to further explore the underlying mechanisms of dissemination bias in qualitative research.

Acknowledgements

Our thanks for their feedback to those who participated in the GRADE-CERQual Project Group meetings in January 2014 and/or June 2015.

Funding

This work was supported in part by funding from the Alliance for Health Policy and Systems Research (www.who.int/alliancehpsr/en/), WHO: Norad (Norwegian Agency for Development Cooperation: www.norad.no) and the Research Council of Norway (www.forskningsradet.no). SL is supported by funding from the South African Medical Research Council (www.mrc.ac.za). The funders had no role in preparation of the manuscript or the decision to publish.

Conflicts of interest: none.

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