RESEARCH NOTE

Causes of reporting bias: a theoretical framework [version 1; peer review: 1 approved with reservations]

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Abstract

Reporting of research findings is often selective. This threatens the validity of the published body of knowledge if the decision to report depends on the nature of the results. Studies on causes and mechanisms underlying selective reporting may help to avoid or reduce reporting bias. Such research should be guided by a theoretical framework of possible causal pathways that lead to reporting bias. We build upon a classification of determinants of selective reporting that we recently developed in a systematic review of the topic. The resulting theoretical framework features four clusters of causes. There are two clusters of necessary causes: (A) motivations (e.g. a preference for positive findings) and (B) means (e.g. a flexible study design). These two combined represent a sufficient cause for reporting bias to occur. The framework also features two clusters of component causes: (C) conflicts and balancing of interests referring to the individual or the team, and (D) pressures from science and society. The component causes may modify the effect of the necessary causes or may lead to reporting bias mediated through the necessary causes. Our theoretical framework is meant to inspire further research and to create awareness among researchers and end-users of research about reporting bias and its causes.

Keywords

Causality, publication bias, questionable research practice, reporting bias, research design, selective reporting
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Background

The problem of selective reporting and research on reporting bias

Selective reporting of research findings presents a large-scale problem in science, substantially affecting the validity of the published body of knowledge (Bouter et al., 2016; Dwan et al., 2014; van den Bogert et al., 2017). Reporting bias (publication bias or outcome reporting bias) occurs when the decision to report depends on the direction or magnitude of the findings. In clinical research, registration of trials prior to data collection is used to prevent selective reporting with some success (Chan et al., 2017; Gopal et al., 2018). However, it is insufficiently effective because despite registration or publication of the study protocol, trial results often remain partially or completely unpublished (Jones et al., 2013) and selective reporting of “positive findings” also occurs among trials registered at, for example, clinicaltrials.gov (Dechartres et al., 2016).

Although many epidemiological studies have described the occurrence of selective reporting, very few studies have targeted its causes. In particular, there is little high-quality evidence on effective interventions. To develop effective interventions against reporting bias, we need a good understanding of possible contributions of actors involved (such as academic environment, editors, researchers) and of possible mechanisms. We also need clear hypotheses of how causes may be interrelated and how actors are involved.

Basis for a theoretical causal framework: hypothesized determinants of selective reporting and their interrelationships

We recently developed a taxonomy of putative determinants of selective reporting abstracted from the literature. We used qualitative content analyses of empirical and non-empirical studies until we reached saturation, which indicates that the categories likely cover all important putative determinants of selective reporting. This resulted in 12 categories (Table 1).

In the literature review we also found some instances of hypothesized effect modification of the determinants of selective reporting, so that the effects of determinants are assumed not to be simply additive. For example, “Outcomes could be deemed post hoc to have little clinical relevance if they fail to show significant findings and may thus be omitted when accommodating space limitations” (Chan & Altman, 2005). In this case, a preference, namely statistically significant findings, combined with editorial practices lead to reporting bias. Similarly, Ioannidis (2005) hypothesized that a focus on preferred, positive findings could result in reporting of non-reproducible findings (only) if there is also an opportunity to do so through flexibility in study designs and freedom in reporting on it. That is, he concludes that “The greater the flexibility in designs, definitions, outcomes, and analytical modes in a scientific field, the less likely the research findings are to be true” because “Flexibility increases the potential for transforming what would be ‘negative’ results into ‘positive’ results.”

A framework of possible causal pathways to reporting bias

Motivations and means

Along these lines, we hypothesize that the combination of two of the most common categories in our review (van der Steen et al., 2018) — i.e., focusing on preferred findings and employing a poor or flexible study design, suffices to cause bias through selective reporting. Inspired by Rothman’s (1976) framework of necessary, sufficient and component causes, through multiple discussions, we inductively derived Figure 1 which shows clusters covering these and the ten other categories of determinants and their possible interrelationships. The two categories are part of clusters A (motivations) and B (means). We view both clusters A and B as necessary causes, that is, they are both part of any sufficient cause of reporting bias. There is also effect modification between A and B because reporting bias is not possible with A or B alone. Note that the preference need not be authors’ preference; it may also be that of a reviewer or editor. In addition to clusters A and B, we propose clusters C and D containing categories of component causes which are discussed in the next section.

Poor or flexible study design may offer the means for selective reporting in addition to limitations in reporting and editorial practices (cluster B in Figure 1). In parallel, we placed “prejudice” in cluster A together with “preference for particular findings” because both may, whether consciously or not, represent a motivation for behaviour that leads to reporting bias. The possible motivations, wishes and beliefs in cluster A are different concepts that may result in “wishful thinking” (Bastardi et al., 2011) and in motivated reasoning around the interpretation of scientific findings (e.g. to serve political interests; Colombo et al., 2016; Kraft et al., 2015). Persons may or may not be fully aware of their motivations and the resulting behaviour may or may not be intentional (Greenland, 2009). At the root of reporting bias may thus lay a basic human attitude, the very natural tendency to make public our successes (Dickersin & Min, 1993).

The pertinence of the second necessary cause (cluster B)—multiple opportunities to select what to analyse or report—is illustrated by the many degrees of freedom that researchers have but should not be tempted to use (in performing psychological research: Wicherts et al., 2016). The necessary causes thus represent having a motive (preference or prejudice; cluster A) and the means (opportunities in study design or reporting; cluster B). Together they form a sufficient cause for reporting bias.

Obviously, researchers and editors are key stakeholders because commonly they decide what is actually being reported and what is not. It can be argued that researchers are the most important because a single editor’s decision is not decisive for non-publication or selective publication. Researchers are actors in three of the four categories in clusters A and B that represent the necessary causes, while editors are key players in only one
### Table 1. Twelve categories of determinants of selective reporting.


<table>
<thead>
<tr>
<th>Determinant category</th>
<th>Description</th>
<th>Examples</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>A. Motivations</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Preference for particular findings</td>
<td>A particular preference motivates a focus on finding results that match preferences, mostly statistically significant or otherwise positive findings, wishful thinking and acting</td>
<td>Significance chasing, finding significant results, larger effect size, suppressing publication of unfavourable results, not being intrigued by null findings</td>
</tr>
<tr>
<td>Prejudice (belief)</td>
<td>A conscious or unconscious belief that may be unfounded, and of which one may or may not be aware</td>
<td>Prior belief about efficacy of treatment, author reputation or gender bias in the phase of review</td>
</tr>
<tr>
<td><strong>B. Means</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Opportunities through poor or flexible study design*</td>
<td>Attributes of study design relating to power and level of evidence provide much leeway in how studies are performed and in interpretation of their results</td>
<td>Not a controlled or blinded study, study protocol unavailable, small sample size</td>
</tr>
<tr>
<td>Limitations in reporting and editorial practices</td>
<td>Constraints and barriers to the practice of reporting relevant detail</td>
<td>Journal space restrictions, author writing skills</td>
</tr>
<tr>
<td><strong>C. Conflicts and balancing of interests</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Relationship and collaboration issues</td>
<td>Intellectual conflict of interest between reporting and maintaining good relationships</td>
<td>Disagreements among co-authors and between authors and sponsors, sponsors prefer to work with investigators who share the sponsor’s position</td>
</tr>
<tr>
<td>Dependence upon sponsors</td>
<td>Financial conflict of interest resulting in lack of academic freedom</td>
<td>Requirements and influence of funding source with financial interests in study results</td>
</tr>
<tr>
<td>Doubts about reporting being worth the effort</td>
<td>Weighing investment of time and means versus likelihood of gain through publication</td>
<td>Anticipating disappointment of yet another rejection or low chances of acceptance of a manuscript, belief that findings are not worth the trouble</td>
</tr>
<tr>
<td>Lack of resources, including time</td>
<td>Insufficient manpower or finances</td>
<td>Lack of time resulting from excessive workload, or lack of personnel due to life events</td>
</tr>
<tr>
<td><strong>D. Pressures from science and society</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Academic publication system hurdles</td>
<td>Various hurdles to full reporting related to submission and processing of manuscripts (other than reporting) including those that represent an intellectual conflict of interest</td>
<td>Solicited manuscripts, authors indicating non-preferred reviewers, editor’s rejection rate</td>
</tr>
<tr>
<td>High-risk area and its development</td>
<td>Area of research or discipline or specialty including its historical development and competitiveness, the currently dominant paradigms and designs, and career opportunities</td>
<td>Ideological biases in a research field, area with much epidemiological research versus clinical or laboratory research (“hard sciences”), humanities, experimental analytic methods, “hot” fields, publication pressure in the specific field</td>
</tr>
<tr>
<td>Unfavourable geographical or regulatory environment</td>
<td>Geographical or regulatory environment that affects how research is being performed</td>
<td>Continents under study included North America, Europe and Asia; few international collaborations; no governmental regulation of commercially sponsored research</td>
</tr>
<tr>
<td>Potential harm</td>
<td>Publishing data can harm individuals</td>
<td>Risk of bioterrorism, or confidentiality restriction</td>
</tr>
</tbody>
</table>

*With study design, we mean broader design issues than just type of research design, including also definitions, outcomes, analytic plans etc.

Conflicts and balancing of interests and the wider environment

After a series of rejections researchers may doubt whether reporting is worth the effort under the pressure of lack of resources such as time. Balancing effort and output is placed in cluster C (component cause conflicts and balancing of interests; Figure 1). Cluster C also includes relationship and collaboration issues and dependence upon sponsors. Cluster C thus represents conflicts of interests, individuals and teams juggling with harmony in relationships and time investments.

Other component causes represent pressures from science and society (cluster D), so from the wider environment. The individual researcher has less control over type C, and in
Figure 1. A theoretical framework for reporting bias. Bullet points indicate the 12 categories of determinants of selective reporting subsumed under four higher-level clusters A, B, C, and D. Note that the figure implies effect modification between A and B (necessary causes) because there will be no reporting bias with A or B alone. Effect modification (“X”) may also occur by C or D and thus make the joint effect of A and B stronger. Mediation (“M”) may occur if the necessary causes (A and B) mediate the effect of D. Mediation may also occur if C mediates the effects of D on A and B, which in its turn leads to reporting bias.

Discussion
We propose a theoretical framework of reporting bias by relating and ordering 12 determinant categories that we derived from the literature (van der Steen et al., 2018). We further combined these categories in four clusters (A-D).

The model has more layers and is more refined than we anticipated when we wrote a protocol to develop a taxonomy of determinants of selective reporting and their interrelationships. We then expected a central role for preferences for particular “positive” findings only (van der Steen et al., 2018 Supplement 1, Figure 1). However, having the means is necessary as well. Although the determinants in our model are mostly based on research in the biomedical area, the model fits well with the “Desire-Belief-Opportunity” (DBO) model that analytical
sociologists use to explain various phenomena (Hedström, 2005) and which we came across after developing our theoretical framework. Desire and Belief concur with the two motivations in cluster A, while opportunities (alternative actions available to the actor) represent the means in cluster B.

Theory may guide the development of interventions as research often does not systematically consider contextual and individual factors that influence delivery of an intervention. Thus, theory may help avoid an ad hoc or data-driven approach to attempts to reduce reporting bias. Although one might assume that interventions addressing reporting bias effectively will be complex, the removal of a single necessary cause is obviously effective. For example, a potentially very effective measure that funders and (medical) ethics committees could adopt is systematic monitoring of all written research outputs and comparing the outcomes reported therein to the corresponding research protocols and statistical analysis plans and eventual amendments. This would require that these organizations make submission to them or to a publicly available repositories mandatory. The approach would become practically feasible if software comparing protocols to publications becomes available (ter Riet & Bouter, 2016). In the jargon of this paper, this approach would eliminate the necessary cause ‘means.’

Given suitable negative reinforcements (punishments) following incomplete reporting, such measures may also reduce motivation to report selectively. Similarly, elements from the component causes contained in cluster C and D that are highly prevalent and strongly modify the combined effect of cluster A and B may be prioritized targets. Mediators can also be good candidates for intervention. For example, component causes contained in cluster C may mediate the impact of elements of D on elements of clusters A or B.

In addition to informing the development of interventions that are subsequently evaluated, our framework may also help to identify high risk scientific fields. For example, areas where designs offer considerable flexibility or where the researchers’ degrees of freedom are combined with strong beliefs or a mission to disseminate particular outcomes. Of course, based on research, our theoretical framework may need to be adapted. Motive and means may be stable clusters but the C and D type causes may change as science changes. Future work may also help to refine the framework’s relevance for specific disciplinary fields (e.g., non-clinical biomedical research). Nevertheless, because the causal pathways seem plausible, we derived from the literature on selective reporting and is congruent with theory developed in the social sciences (Hedström, 2005), we feel that the current work can already help to design further research on the effectiveness of interventions.

Data availability

Underlying data

PLOS ONE Supplement 2 to article van der Steen et al., 2018. Determinants of selective reporting abstracted from the selected literature. “S2 File. Dataset with determinants.” In Excel available from: https://doi.org/10.1371/journal.pone.0188247.s003 (van der Steen et al., 2018)

PLOS ONE Supplement 3 to article van der Steen et al., 2018. Categories of determinants of selective reporting with literature references. “S3 file. References to the 64 articles included in the determinant analysis, per category.” In Word available from: https://doi.org/10.1371/journal.pone.0188247.s004 (van der Steen et al., 2018)

Data are available under the terms of the Creative Commons Attribution 4.0 International license (CC-BY 4.0).

Grant information

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The funders had no role in study design, data collection and analysis, decision to publish, or preparation of the manuscript.

References


Dwan K, Altman DG, Clarke M, et al.: Evidence for the selective reporting of...


Open Peer Review

Ksenija Bazdaric
Department of Medical Informatics, Faculty of Medicine, University of Rijeka, Rijeka, Croatia

I was happy to review a manuscript about a theoretical framework in the field of reporting bias. I think the authors have proposed an interesting perspective but my major remark is that they try to explain human behaviour with an epidemiological model for which I don’t find a body of evidence in the literature that could convince me.

Comments:

1. Background: In clinical research, registration of trials prior to data collection is used to prevent selective reporting with some success – please delete “some success” because it is further explained.

2. “A framework of possible causal pathways to reporting bias - Motivations and means.
   Along these lines, we hypothesize that the combination of two of the most common categories in our review (van der Steen et al., 2018) — i.e., focusing on preferred findings and employing a poor or flexible study design, suffices to cause bias through selective reporting..." – how do you then comment on the replication crisis in psychology and the experiments that were replicated in the same laboratories? They were motivated to replicate and for sure were not sloppy. Do we have a poor designed field here or are there other factors? I would like a more detailed explanation.

3. A theoretical framework for reporting bias. Rothman’s theoretical model – is there any evidence in practice for this model in relation to human behaviour?
   Figure 1 and the model: It is an interesting figure, but the same could be explained by some other general theories, for example ‘Theory of planned behaviour’ (of course evidence cannot be confirmed as we have a replication crisis in psychology). I don’t believe human behaviour can be explained with an epidemiological model although it is very nice. Also, the model itself does not have a word about ethical climate and other possible external factors. Why did you exclude them? Do you consider them stable in all environments?
   There is some evidence that peers can predict replication? How do you comment? Could you include some external factors in your model? Like environment, ethical climate, etc...

4. The sentence: “At the root of reporting bias may thus lay a basic human attitude, the very natural tendency to make public our successes” – this is not clear at all. At a root of everything probably lies personality and attitude, but I don’t understand the meaning of the sentence here.

5. Obviously, researchers and editors are key stakeholders because commonly they decide what is actually being reported and what is not. – I would say that sometimes we cannot report everything because we have only 3000-4000 words (here is the role of editors).
6. After a series of rejections researchers may doubt whether reporting is worth the effort under the pressure of lack of resources such as time. I advise you to read a case study from Helene Speyer - The value of a “failed” trial.4.

References

Is the work clearly and accurately presented and does it cite the current literature?
Partly

Is the study design appropriate and is the work technically sound?
Partly

Are sufficient details of methods and analysis provided to allow replication by others?
Yes

If applicable, is the statistical analysis and its interpretation appropriate?
Not applicable

Are all the source data underlying the results available to ensure full reproducibility?
No source data required

Are the conclusions drawn adequately supported by the results?
Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: research ethics, psychology, medical informatics

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.
- Your article is published within days, with no editorial bias
- You can publish traditional articles, null/negative results, case reports, data notes and more
- The peer review process is transparent and collaborative
- Your article is indexed in PubMed after passing peer review
- Dedicated customer support at every stage

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