

COMMENTARY

A two-step manuscript submission process can reduce publication bias

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Accepted 27 March 2013; Published online 8 July 2013

**Abstract**

Much of what is researched is never published. This would not be of great concern if the selection of what we read would occur irrespective of study outcomes. Unfortunately, the reverse is the case: “positive” studies have a much larger chance of acceptance after editorial and peer review than “negative” ones. Several solutions to this problem of publication bias have been discussed or implemented, but none seem to be very effective.

In this article, the approach of implementing an editorial and peer-review procedure that is blinded to study outcomes is discussed. This would require a two-step submission procedure of manuscripts: first a version including just the introduction and methods and in some cases followed by a second submission including results and discussion. The pros and cons of such an approach are discussed. © 2013 Elsevier Inc. All rights reserved.

*Keywords:* Publication bias; Peer review; Editorial review; Bias; Statistics; Blinding

Publication bias, the preferential dissemination of “positive” or statistically significant studies via medical journals, is a threat to the credibility and ethical integrity of medical science. Positive studies are much more likely to be published (reported odds ratios vary from 2.2 to 4.7) and appear earlier as well as in higher impact journals than negative studies [1,2]. Closely related to publication bias is outcome reporting bias, the selective reporting of significant results, often violating original analysis protocols. Changing the primary outcome is another way of distorting the presentation of study results and occurs in 40–62% of published studies [2]. The impact of unbalanced dissemination of positive studies/results is further amplified by preferential subsequent citation of such studies over negative ones. Publication bias is strongest in laboratory-based, experimental, and small clinical studies [1], but large studies are certainly not immune to this phenomenon [3]. Taken together, publication bias distorts medical science and may hurt our patients.

Although the awareness of publication bias is not at all new, it seems we have not been willing or able to learn to reduce it. On the contrary, there are signs that publication bias is increasing over the years. One of the first reports

on publication bias was by Sterling in 1959, after he noticed that 97% of psychology studies showed statistically significant results. Looking back in 1995, the same author concluded that the practices contributing to publication bias had hardly improved [4]. A more recent analysis addressed the period between 1990 and 2007 and revealed no less than a 22% increase in the fraction of positive articles [5].

The causes of publication bias are complex and occur in different phases of the publication process. First, authors can feel discouraged to submit negative or statistically non-significant results. Recent evidence suggests that an increasingly competitive scientific environment may even amplify such behavior [6]. Apart from authors, commercial sponsors of studies may feel the same disappointment toward negative results and may attempt to prevent or postpone dissemination of such studies [3].

Obviously, it is not just in the submission phase in which things go wrong. The role of editors and external peer reviewers in generating publication bias is also important. It appears that negative studies have a much higher likelihood of rejection than positive studies, although study quality may not differ. This may occur inside editorial offices and on the desks of external reviewers. A recent study randomized journal reviewers to receive a sham article that had a negative or positive primary outcome but was identical in other respects [7]. Not only did the reviewers assign a higher priority to the positive outcome version but also they rated the identical methods section significantly higher

Conflict of interest: None.

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for the positive article. Finally, reviewers were less attentive to deliberate errors that were part of both versions of the manuscript.

What are the solutions for publication bias? Many hoped that trial registration would be an effective remedy. Unfortunately, trial registration by no means guarantees protocol adherence and (timely) publication [8]. Statistical methods have been developed to attenuate the impact of publications bias in literature reviews and meta-analyses, but these methods have intrinsic limitations and are incompletely implemented [3,9].

Three key questions come to mind, the answers to which seem essential to make progress in the struggle against publication bias. First, how can we address the discouragement authors have to submit “negative” or nonsignificant results? Second, how could editors become more neutral to the outcomes of a study in deciding whether to forward manuscripts to external reviewers? Finally, what could make reviewers equally neutral to the study results in their initial evaluation of an article? The answer to each of these questions seems quite simple: blind both editors and reviewers to the study outcomes. This would be possible by implementing a two-stage manuscript submission process: authors first submit their introduction and methods section, including the statistical paragraph. They may add a baseline characteristics table to properly describe the study population, but no further parts of the results section, except perhaps empty tables and figures. Editors and reviewers then make a preliminary decision based on what really should count in medical science: “Is the question relevant, and has it been properly addressed?” If this preliminary decision turns out favorable, at once or after revision, the authors are notified and encouraged to submit their full results and discussion. In such a second phase, there would be a relatively high threshold for rejecting the full article. After all, results themselves are unlikely to be a proper justification for rejection, unless, for example, the results suggest that the protocol was violated or the data collection was clearly insufficient. In some other cases, the discussion could be of such poor quality that the article must be rejected only because of this. In any event, it would require relatively exceptional circumstances to overturn an initial favorable decision. This makes perfect sense because this initial decision was, again, based on the most crucial questions editors and reviewers should ask themselves: “Does the study address a relevant question, and was that question appropriately addressed?”

What are the disadvantages and limitations of such an approach? A few come to mind. Surely, it might be unpleasant for editors and reviewers to read a “result-less” article. On the other hand, initially submitted manuscripts would become much shorter, reducing the burden of assessing the large number of submitted articles. Another limitation could be that some study results were already presented at medical conferences or published in abstract

form. True as this may be, it would still be difficult for editors and reviewers to reject an introduction and methods section without reference to prior knowledge of the outcome from any other source.

The solution presented here is not completely new. Several authors have presented similar suggestions, some even many years ago [10–13], but the impact of their calls seems to have been small. Now may be a good time to act: we have strong empirical evidence for the magnitude of the problem, we have electronic submission procedures facilitating implementation, and we have the ability to monitor the success rate of the intervention using literature databases.

Should one or several journals decide to implement such a submission procedure, they may consider doing this in the form of a study: randomize submitted manuscripts immediately after receipt to editorial and external review of the full article in one group and of introduction and methods sections only in the other. Who knows what a difference in preliminary recommendations would occur. Editors of one or a few of the larger journals could at least consider this option and discuss it within their editorial staff. They may well set an example in the ongoing struggle against publication bias.

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