While a systematic review is meant to provide a high-quality appraisal of the evidence regarding a healthcare intervention, the conclusions are predicated on the reliability of the included studies [1]. The existence of unreliable or problematic studies in the literature presents difficulties for researchers performing systematic reviews and jeopardizes the credibility of their results. Retracted studies have the most obvious flaws. Retraction is defined as the removal of a published paper from a journal to warn readers of the significant problems identified in the article as a means of maintaining the integrity of scientific literature [2]. Including retracted studies in a systematic review may impact the outcomes and level of evidence, and ultimately provide inaccurate medical guidelines [3,4]. Thus, handling retracted studies in systematic reviews is a critical issue.

Recently, we discovered that one of the studies cited in a systematic review and meta-analysis (SRMA) [5], which was accepted through the peer review process in the Korean Journal of Anesthesiology (KJA), was confirmed to have been retracted during the proofreading stage. Given the ambiguous timing, several rounds of editorial discussions were necessary to thoroughly consider the options regarding how to proceed. The KJA editorial board made a concerted effort to perform peer review and editorial assessment in a robust, transparent, and credible manner regarding this disputed SRMA [6].

What happened during the review process?

Following acceptance of the SRMA [6], a manuscript editor evaluating the proof found that one [5] of the included studies had been retracted for containing too many flaws to be corrected on September 9, 2021. The SRMA protocol was registered with the International Prospective Register of Systematic Reviews (PROSPERO) on April 29, 2021 (registration number: CRD42021252062), and a literature search was conducted in May 2021 on papers published between January 2000 and January 2021. The authors first submitted their original SRMA to the KJA on July 26, 2021. The revised paper was submitted on September 28, 2021, and the final acceptance date after the revision was reviewed was October 12, 2021. During the KJA editorial board review process for the revision of the original SRMA, neither the reviewers nor the editors requested that the related studies be searched again, and the authors did not perform a second search.

A systematic review focuses on selecting and analyzing the findings of previously published papers; thus, each included study per se can be considered important material of the systematic review. The inclusion of a retracted study in the analysis could have significantly impacted the overall content and findings of the systematic review. Consequently, determining how to deal with this SRMA under these circumstances became a
top priority. To resolve this issue, the following concerns had to be addressed through multiple rounds of discussion by the KJA editorial board.

How should a retracted study be dealt with at this point?

Our editorial board had two conflicting points of view. One view was to publish the SRMA with the retracted study included. The retraction date was September 9, 2021, and the first submission date of the original SRMA was July 26, 2021. Therefore, the SRMA had already been completed at the time of retraction and the SRMA was not in progress at that time. Given that requesting the authors to conduct further research after the study had been completed could be interpreted as not following protocol, it would be acceptable to publish the SRMA with the retracted study included, according to this viewpoint. In PROSPERO, the literature search was planned to begin in May 2021, and study searches in systematic reviews should be conducted systematically, transparently, and reproducibly, according to a pre-defined process. The retraction had not yet occurred at this stage. Proofreading is the last step in the process to ensure that the text is accurate and ready for publication. Thus, according to this viewpoint, altering the contents of an SRMA should not occur at this stage.

The other viewpoint of the editorial board was to publish the SRMA following additional revisions in light of the recent retraction. Because a study is not completed until it is published, and this SRMA was still in the proofreading stage, it was argued that revisions at this stage are feasible when necessary. Readers have the right to see the most accurate and up-to-date research; therefore, it is appropriate to revise the content if it can be amended, even if it goes against the concept of the purpose of the proofreading stage. Readers also have the right to know whether a study has been retracted, as well as the reasons of its retraction. If the reason for retraction is that a published paper has too many flaws to be corrected, as in this case, the results of the SRMA may contain significant bias. Publishing the results of an SRMA that may contain errors could be regarded as unethical as it would infringe on the readers’ right to know the truth. Because the SRMA had not yet been published, according to this viewpoint, it was reasonable to consider the proofreading process as a stage where alterations could occur and to conduct a systematic review again, if necessary.

What should be considered in the review process if a retracted study is to be excluded?

In this case, the decision was made regarding the two previous viewpoints and the SRMA was to be re-submitted following a new analysis that excluded the retracted study [5]. The review process for the re-submitted SRMA could be seen from three perspectives: 1) treat the re-submitted SRMA as a completely new submission, for which a new peer review process for acceptance, revision, or rejection would be required. 2) treat the re-submitted SRMA as an extension of the previously submitted SRMA, and add an extension to the review process, and 3) only review the new additions with the retracted study excluded and uphold the editorial decision of the previously submitted SRMA.

Once a retracted study is excluded, the results of the analyses may change, affecting the effect size and statistical significance. Additionally, the main flow of the SRMA may be altered if the outcomes change as a result of excluding the retracted study.

In this case, the SRMA was eventually accepted following a defined peer review process and proofreading conducted by the KJA editorial board. The research protocol of the SRMA was registered with PROSPERO and the SRMA was performed according to the registered protocol. Importantly, the process of omitting the retracted paper did not result in a deviation from the registered protocol. At least at the time of the original SRMA submission, all the processes appear to have been completed appropriately. Therefore, including a retracted study should have had no influence on the acceptance or rejection of an SRMA, as this could be to the cause of another publication bias.

How did the KJA reach a consensus?

The editorial decision made regarding the previously submitted SRMA was upheld after the retracted paper was discovered. However, we had to account for the fact that excluding the retracted study could have altered the results of the SRMA. From a broad perspective, proofreading is deemed to be a part of the review process, which includes everything from submitting to evaluating papers. In this case, we requested that the authors add a new section to the latest version of their manuscript by performing a systematic review without the retracted paper. The authors were also asked to declare that a retracted study was included in their previous SRMA to provide the reason for the retraction. A second literature search, data extraction process, and analysis were conducted and the results were included as supplemental files at the time of retraction.

This topic may be relevant to readers in terms of the decision to include or exclude retracted studies in systematic reviews, and it may also be intriguing on its own given the various potential viewpoints on the issue. The following message was conveyed to all the study participants: systematic reviews may involve retract-
ed citations, which journal editors, reviewers, and researchers should be aware of [4]. Ensuring that the included references have not been retracted should be part of the peer-review process. The authors must regularly check the status of the included studies and ensure that they do not cite retracted papers [7]. In this case, it was impossible to avoid including a retracted study when submitting the original version, whereas it was possible to exclude a retracted study before publication because the retraction occurred during the proofreading stage. Given this ambiguous timing, establishing an appropriate review process was our primary challenge. Because no similar cases have been reported to date, clearly disclosing our process here allows for this to be used as a reference when a similar scenario arises in the future. It can also be used to demonstrate how the editorial board at the KJA strives to make the review process robust, transparent, and credible, as well as how it handles problems that occur during the review process using a rigorous and reasonable approach.

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Author Contributions

Geun Joo Choi (Conceptualization; Data curation; Formal analysis; Investigation; Methodology; Project administration; Writing – original draft; Writing – review & editing)
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