

# Introduction

2022-02-27

## Introduction

Scientific production has experienced continuous exponential growth in the last decades (Bornmann & Mutz, 2015; Larsen & Von Ins, 2010). This is especially true for biomedical research, a trend further accelerated by the COVID-19 pandemic, thanks to faster article processing time by publishers and the greater use of preprint databases (Aviv-Reuven & Rosenfeld, 2021; Horbach, 2020; Hoy, 2020). Consequently, it has become harder for researchers and practitioners to stay up to date on the latest findings in their field. Secondary research is of paramount importance in this scenario in that it provides valuable summaries of the latest research results; however, it is becoming ever more challenging in terms of time and human resources required (Allen & Olkin, 1999; Bastian et al., 2010; Borah et al., 2017; A. M. Cohen et al., 2010).

The article collection and screening phases of a systematic review are particularly demanding (Babar & Zhang, 2009). First, relevant published research must be collected from scientific databases using appropriately built search queries (retrieval phase); secondly, the scientific citations collected must be screened, selecting only those that are relevant to the topic (appraisal phase) (Bannach-Brown et al., 2019; Higgins et al., 2019; Tsafnat et al., 2014).

Search queries construction is a complex task (Hammerstrøm et al., 2010; Lefebvre et al., 2011), requiring both expertise in the scientific field of interest and some knowledge of the database query languages. The goal is to obtain a set of results that contains all relevant articles (high sensitivity) while keeping the total number of records low (high specificity), possibly focusing on the first at the expense of the second (Hammerstrøm et al., 2010).

If an integrated search tool is not used, manual work is required to download, store and organise the publication data; this approach is complicated by limits to the number of records that can be downloaded at any one time and the need to harmonise different formats and resolve record duplication (Marshall & Wallace, 2019).

The citation screening phase is usually the more resource-demanding task in a systematic review: even with appropriately built search queries, the results may easily range in the tens of thousands, of which just a small fraction are actually relevant (Lefebvre et al., 2011). It has been estimated that labelling 10,000 publications can take up to 40 weeks of work and that the average clinical systematic review takes 63 weeks to complete (Allen & Olkin, 1999; Bannach-Brown et al., 2019; Borah et al., 2017). A consequence of this is that systematic reviews are often already out-of-date by the time they are published (E. M. Beller et al., 2013). The field of Data Science applied to evidence synthesis and acquisition has greatly matured in the last years (E. Beller et al., 2018; Marshall & Wallace, 2019; Tsafnat et al., 2014). By applying natural language processing (NLP), it is possible to transform free text into quantitative features, with various levels of abstraction and generalisation (Ananiadou & McNaught, 2006; K. B. Cohen & Hunter, 2008); using machine learning, such text-derived data can be used to map and reproduce human judgment, automating the screening of citations (Ikonomakis et al., 2005).

Automation of systematic reviews has made significant improvements in the last years (Ananiadou et al., 2009; Jonnalagadda et al., 2015; O'Mara-Eves et al., 2015; Tsafnat et al., 2013), and it is possible foreseeable that it will become the standard approach in the field (E. Beller et al., 2018), with many solutions already being implemented into commercial or free-to-use tools (see Marshall & Wallace, 2019, table 1).

This manuscript introduces an open-source, production-ready framework that further contributes to the state-of-the-art in systematic review automation (SRA) and helpers (SRH) tools. We improve the “retrieval phase” by providing a unified framework for the automated collection and management of scientific literature from multiple online sources. For the citation screening (appraisal) phase, we built an active machine

learning-based protocol (Miwa et al., 2014; Settles, 2009), which utilises a Bayesian framework to efficiently identify potentially relevant documents that require human review while automatically screening-out the vast majority of clearly non-relevant ones; the algorithm then requires human review to increase classification accuracy iteratively. Finally, we included a tool to generate new search queries based on an already categorised citation data set, to identify relevant research that manually-made queries may have possibly missed.

We tested the framework in the retrieval and appraisal phases of an example topic of interest to our group: the evaluation of the mathematical modelling of patient referral networks among hospitals and their impact on the diffusion of healthcare-associated pathogenic microorganisms; the protocol is published in (Sadaghiani et al., 2020).

In the Methods, we give an overview of the framework, in the Result, we show the outputs and performance of the framework applied to the example topic, and in the Discussion, we explain the methodological rationale for the different components and features of the framework.

- Allen, I. E., & Olkin, I. (1999). Estimating time to conduct a meta-analysis from number of citations retrieved. *Jama*, 282(7), 634–635.
- Ananiadou, S., & McNaught, J. (2006). *Text mining for biology and biomedicine*. Citeseer.
- Ananiadou, S., Rea, B., Okazaki, N., Procter, R., & Thomas, J. (2009). Supporting systematic reviews using text mining. *Social Science Computer Review*, 27(4), 509–523.
- Aviv-Reuven, S., & Rosenfeld, A. (2021). Publication patterns’ changes due to the COVID-19 pandemic: A longitudinal and short-term scientometric analysis. *Scientometrics*, 1–24.
- Babar, M. A., & Zhang, H. (2009). Systematic literature reviews in software engineering: Preliminary results from interviews with researchers. *2009 3rd International Symposium on Empirical Software Engineering and Measurement*, 346–355.
- Bannach-Brown, A., Przybyła, P., Thomas, J., Rice, A. S., Ananiadou, S., Liao, J., & Macleod, M. R. (2019). Machine learning algorithms for systematic review: Reducing workload in a preclinical review of animal studies and reducing human screening error. *Systematic Reviews*, 8(1), 1–12.
- Bastian, H., Glasziou, P., & Chalmers, I. (2010). Seventy-five trials and eleven systematic reviews a day: How will we ever keep up? *PLoS Medicine*, 7(9), e1000326.
- Beller, E. M., Chen, J. K.-H., Wang, U. L.-H., & Glasziou, P. P. (2013). Are systematic reviews up-to-date at the time of publication? *Systematic Reviews*, 2(1), 1–6.
- Beller, E., Clark, J., Tsafnat, G., Adams, C., Diehl, H., Lund, H., Ouzzani, M., Thayer, K., Thomas, J., Turner, T., others. (2018). Making progress with the automation of systematic reviews: Principles of the international collaboration for the automation of systematic reviews (ICASR). *Systematic Reviews*, 7(1), 1–7.
- Borah, R., Brown, A. W., Capers, P. L., & Kaiser, K. A. (2017). Analysis of the time and workers needed to conduct systematic reviews of medical interventions using data from the PROSPERO registry. *BMJ Open*, 7(2), e012545.
- Bornmann, L., & Mutz, R. (2015). Growth rates of modern science: A bibliometric analysis based on the number of publications and cited references. *Journal of the Association for Information Science and Technology*, 66(11), 2215–2222.
- Cohen, A. M., Adams, C. E., Davis, J. M., Yu, C., Yu, P. S., Meng, W., Duggan, L., McDonagh, M., & Smalheiser, N. R. (2010). Evidence-based medicine, the essential role of systematic reviews, and the need for automated text mining tools. *Proceedings of the 1st ACM International Health Informatics Symposium*, 376–380.
- Cohen, K. B., & Hunter, L. (2008). Getting started in text mining. *PLoS Computational Biology*, 4(1), e20.
- Hammerstrøm, K., Wade, A., Jørgensen, A.-M. K., & Hammerstrøm, K. (2010). Searching for studies. *Education*, 54(11.3).
- Higgins, J. P., Thomas, J., Chandler, J., Cumpston, M., Li, T., Page, M. J., & Welch, V. A. (2019). *Cochrane handbook for systematic reviews of interventions*. John Wiley & Sons.
- Horbach, S. P. (2020). Pandemic publishing: Medical journals strongly speed up their publication process for COVID-19. *Quantitative Science Studies*, 1(3), 1056–1067.
- Hoy, M. B. (2020). Rise of the rxivs: How preprint servers are changing the publishing process. *Medical Reference Services Quarterly*, 39(1), 84–89.

- Ikonomakis, M., Kotsiantis, S., & Tampakas, V. (2005). Text classification using machine learning techniques. *WSEAS Transactions on Computers*, 4(8), 966–974.
- Jonnalagadda, S. R., Goyal, P., & Huffman, M. D. (2015). Automating data extraction in systematic reviews: A systematic review. *Systematic Reviews*, 4(1), 1–16.
- Larsen, P., & Von Ins, M. (2010). The rate of growth in scientific publication and the decline in coverage provided by science citation index. *Scientometrics*, 84(3), 575–603.
- Lefebvre, C., Manheimer, E., Glanville, J., Higgins, J., & Green, S. (2011). Searching for studies (chapter 6). *Cochrane Handbook for Systematic Reviews of Interventions Version*, 510.
- Marshall, I. J., & Wallace, B. C. (2019). Toward systematic review automation: A practical guide to using machine learning tools in research synthesis. *Systematic Reviews*, 8, 1–10.
- Miwa, M., Thomas, J., O’Mara-Eves, A., & Ananiadou, S. (2014). Reducing systematic review workload through certainty-based screening. *Journal of Biomedical Informatics*, 51, 242–253.
- O’Mara-Eves, A., Thomas, J., McNaught, J., Miwa, M., & Ananiadou, S. (2015). Using text mining for study identification in systematic reviews: A systematic review of current approaches. *Systematic Reviews*, 4(1), 1–22.
- Sadaghiani, C., Donker, T., Andrianou, X., Babarczy, B., De Boer, G., Di Ruscio, F., Cairns, S., Crépey, P., Fortaleza, C. M., Freyler, P., Gagliotti, C., Liljeros, F., Moro, M. L., Oteo, J., Palos, C., Pilarski, G., Reilly, J., Robotham, J., Sá-Leão, R., ... Mutters, N. T. (2020). *National health care infrastructures, health care utilization and patient movements between hospitals - networks working to improve surveillance: A systematic literature review*. PROSPERO. [http://www.crd.york.ac.uk/PROSPERO/display/\\_\\_record.asp?ID=CRD42020157987](http://www.crd.york.ac.uk/PROSPERO/display/__record.asp?ID=CRD42020157987)
- Settles, B. (2009). *Active learning literature survey*.
- Tsafnat, G., Dunn, A., Glasziou, P., & Coiera, E. (2013). *The automation of systematic reviews*. British Medical Journal Publishing Group.
- Tsafnat, G., Glasziou, P., Choong, M. K., Dunn, A., Galgani, F., & Coiera, E. (2014). Systematic review automation technologies. *Systematic Reviews*, 3(1), 1–15.