An open-source integrated framework for the automation of citation collection and screening in systematic reviews.

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Abstract

The exponential growth in scientific production makes its summarisation through secondary literature exceedingly demanding in terms of time and human resources. We introduce a new open-source framework that significantly increases efficiency during the citation collection and screening phases of systematic reviews.

The framework introduces three main tools: 1) an automatic citation search engine and manager that allows collecting records from multiple citation databases (PUBMED, WOS, SCOPUS, EMBASE, IEEE) with a unified query syntax; 2) a citation screening tool based on Bayesian active machine learning and natural language processing, which requires users feedback only on uncertain classifications to increase predictive accuracy iteratively; 3) a semi-automatic data-driven query generator to create new search queries from existing reviewed citation data sets.

The framework was applied on an example topic performing citation collection and screening. We estimated median posterior Sensitivity and Efficiency [90% Credible Intervals] using Bayesian simulation to predict the distribution of possibly missed relevant matches in the unreviewed records.

17755 unique records were collected through the framework citation manager; 101 over 766 records were found to be relevant after manual evaluation, while the rest were excluded by the automatic classification; the expected Efficiency was 95.7% [95.3%, 95.7%] for a Sensitivity of 100% [93.5%, 100%]. A new search query was generated from the labelled dataset, and 82579 more records were collected. Only 567 records required human review after the automatic screening, and six additional positive matches were found. Including the

additional records, the overall expected Sensitivity dropped to 97.3% [73.8%, 100%] while the Efficiency raised to 98.6% [98.2%, 98.7%].
The framework can significantly reduce human resources requirements for systematic reviews by simplifying citation collection and screening while demonstrating exceptional sensitivity even on large data sets.

Keywords Systematic review automation · Citation management · Online data collection · Active machine learning · Natural language processing · Bayesian modeling

1 Introduction

Scientific production has experienced continuous exponential growth in the last decades (Larsen and Von Ins 2010; Bornmann and Mutz 2015). This is especially true for biomedical research, a trend further increased by the COVID-19 pandemic, thanks to faster article' processing time by publishers and the diffusion of preprint databases' usage (Aviv-Reuven and Rosenfeld 2021; Horbach 2020; Hoy 2020). Consequently, it gets harder for researchers and practitioners to stay up-to-date on the latest findings in their field. Secondary research is of paramount relevance in this scenario, providing valuable summaries of the latest research results, but is getting ever more demanding in terms of time and human resources (Allen and Olkin 1999; Borah et al. 2017; A. M. Cohen et al. 2010; Bastian, Glasziou, and Chalmers 2010).
The article collection and screening phases of a systematic review are particularly demanding (Babar and Zhang 2009). First, relevant published research needs to be collected from scientific databases through appropriately built search queries (retrieval phase); secondly, the acquired scientific citations need to be screened, selecting only those relevant to the topic (appraisal phase) (Bannach-Brown et al. 2019; Tsafnat et al. 2014; Higgins et al. 2019).
The construction of search queries is a complex task (Lefebvre et al. 2011; Hammerstrøm et al. 2010), requiring both domain and some knowledge of the databases' query languages; the goal is to produce a set of results containing all relevant articles (high sensitivity) while keeping the total number low (high specificity), focusing on the first aspect at the cost 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 in the number of records that can be downloaded at once and the necessity to harmonise different formats and resolve record duplication (Marshall and Wallace 2019).
The citation screening phase is usually the more resource-demanding task of a systematic review: even with appropriately built search queries, the returned results easily range in the tens of thousands of which just a small fraction is actually relevant (Lefebvre et al. 2011). It was estimated that labelling 10,000 publications may take as much as 40 weeks of work and that the average clinical systematic review takes 63 weeks to be completed (Bannach-Brown et al. 2019; Borah et al. 2017; Allen and Olkin 1999). A consequence is that often systematic reviews are already outdated once they are published (E. M. Beller et al. 2013).
The field of Data Science applied to evidence synthesis and acquisition has greatly maturated in the last years (Marshall and Wallace 2019; E. Beller et al. 2018; Tsafnat et al. 2014). Through the application of natural language processing (NLP), it is possible to transform free text into quantitative features, with various levels of abstraction and generalisation (Ananiadou and McNaught 2006; K. B. Cohen and Hunter 2008); with machine learning, such text-derived data can be used to map and reproduce human judgment, automating the citation screening (Ikonomakis, Kotsiantis, and Tampakas 2005).
The automation of systematic reviews has been ripe with improvements in the last years (Ananiadou et al. 2009; O'Mara-Eves et al. 2015; Tsafnat et al. 2013; Jonnalagadda, Goyal, and Huffman 2015), and it is possible to foresee that it is going to become the standard approach in the field (E. Beller et al. 2018), with many solutions already being turned into commercial and free-to-use tools (see Marshall and Wallace 2019, table 1).
In this manuscript we present an open source, production-ready framework which 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 automatic collection and management of scientific literature from multiple online sources. For the citation screening (appraisal) phase, we built an active machine learning-based Miwa et al. (2014) protocol, which exploits a Bayesian framework to efficiently identify potentially relevant documents that require human review while automatically screening-out the large majority of clearly non-relevant ones; the algorithm then uses human reviews to iteratively increase classification accuracy. Finally, we included a tool to generate new search queries given an already labelled citation data set, in order to identify relevant research possibly missed by human-made queries.
We tested the framework in the retrieval and appraisal phases of an example topic of interest for our group: the evaluation of the mathematical modelling of patient referral networks among hospitals and their impact

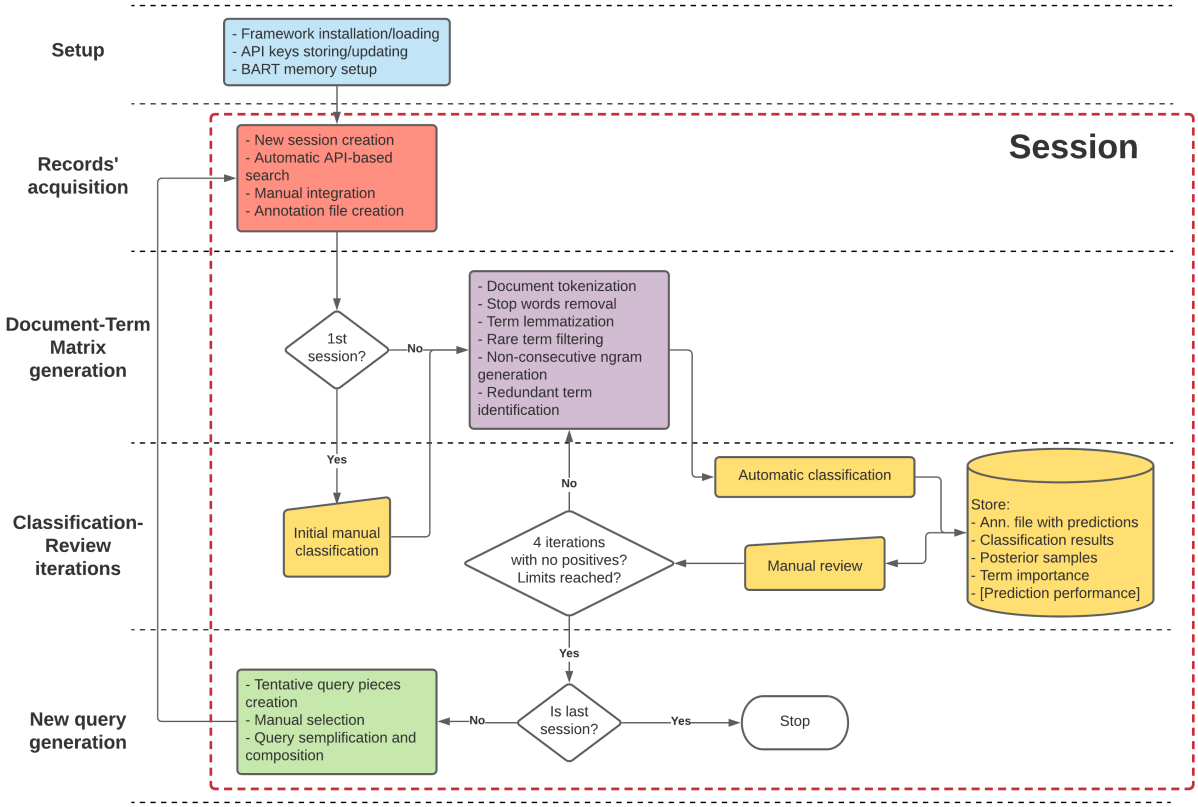

Figure 1.Framework's visual depiction.

on the diffusion of healthcare-associated pathogenic microorganisms; the protocol is published in (Sadaghiani et al. 2020).
We give an overview of the framework in the Methods section; in the Result section, we show the framework outputs and performance once applied to the example topic; in the Discussion, we lay out the methodological justification behind the different components and features of the framework.

2 Methods

2.1 General description

We built an R (R Core Team 2020) based framework to simplify two aspects of systematic reviews: record acquisition and classification. The code used to generate the results in the manuscript is available at X while an update version of the framework is distributed as an R package at Y. The framework includes several modules that communicate through intermediate outputs stored in standard formats, which make it possible for users to extend the framework or easily integrate it with other tools in their pipeline. See Supplemental Material S1 for an in-depth description of the framework and how to use it.
The tasks carried out by the framework are grouped into "sessions", which comprise obtaining scientific citation data (records) using a search query and then labelling them as relevant ("positive" in the rest of the text) or not ("negative") for the topic of interest with the help of a machine learning engine (Fig. 1). The initial search query should be built using domain knowledge, trying to achieve a high relevant/non-relevant record ratio.
The framework can then generate a new data-driven query from this labelled set to perform a new session to find records possibly missed by the first query.

2.2 Record's acquisition and initial labelling

We built a set of tools to let users automatically search and download citation data from three major scientific databases ("sources"): Pubmed (`https://pubmed.ncbi.nlm.nih.gov/`), Web Of Science (WOS, `https://apps.webofknowledge.com/`) and the Institute of Electrical and Electronics Engineers (IEEE, `https://ieeexplore.ieee.org/Xplore/home.jsp`). The framework takes care of authorization management for non-open databases like WOS and IEEE. It is also possible to download and import records in the framework manually; this is particularly useful to acquire records from the SCOPUS (`https://www.scopus.com/`) and EMBASE databases (`https://www.embase.com/`), for which a comprehensive API interface was not easy to build. An extra manual search was necessary also for Pubmed, since the API and the web interface have different rule expansion algorithms and return slightly different results ('NCBI Insights : Updated Pubmed e-Utilities Coming in April 2022!', n.d.). A short guide on how to set up the framework for each supported database is available in Supplemental Material S3.

The acquired records are merged into a single database, resolving duplicates and different formatting between sources. The records are ordered according to the frequency of the positive query terms (e.g., not preceded by a *NOT* modifier) in the title and abstract ("simple query ordering").

The researcher is then asked to label a number of records to create the "initial training set" needed to start the automatic classification. We suggest manually labelling the first 250 records (see "hyperparameter optimization" later). The simple query ordering increases the positivity rate in the initial training set (Wallace, Small, et al. 2010), which provide higher sensitivity during automatic classification (Chawla, Japkowicz, and Kotcz 2004).

2.3 Text feature extraction

The collected citation data have a number of fields characterising a scientific publication. The framework models the relevance of a record based on the following fields: title, abstract, authors, keywords, MESH terms (Lipscomb 2000). A series of Natural Language Processing (NLP) techniques (Baeza-Yates, Ribeiro-Neto, et al. 1999; Marshall and Wallace 2019; Ananiadou and McNaught 2006) are employed to transform the textual information in these fields into features for machine learning through a bag-of-words approach (Marshall and Wallace 2019). The processing of free text fields (title, abstract) includes: tokenization (i.e., extracting the terms), common stopwords (i.e., sentence components bringing no meaning) removal, part-of-speech filtering (only nouns, adjectives, verbs and untagged terms are retained), and lemmatization of the terms (i.e., reduction to their base grammar form). Text processing for authors, keywords and MESH terms identifies logical units (e.g., author's full names, composite keywords) and extracts them.

Terms appearing in less than 5% of the labelled documents are removed from negative records. All terms in the positive set are kept to increase sensitivity at the cost of specificity.

Some terms tend to co-appear in records (non-consecutive ngrams, nc-ngrams), often carrying a particular meaning when copresent. To detect nc-ngrams, we generated a word network representation (Francois Rousseau 2015) posing edges between terms with a cosine similarity in terms of record co-occurrence > 0.5. We extracted the maximal cliques in the network (Eppstein, Löffler, and Strash 2010) representing highly correlated groups of terms; These groups of terms are added to the data set as individual features. Only nc-ngrams comprising a maximum of ten terms are kept.

A second network is built using a co-occurrence threshold of 0.9. In this case, the cliques represent terms that always appear together and therefore can be considered redundant (i.e., they do not need to be considered separately). These terms are merged to increase computation efficiency and reduce overfitting.

The output is a Document-Term Matrix (DTM), with N_d rows representing the records (D_i), N_t terms column for the t_{field} terms (divided by record field) and $0, 1$ values whether $t_{field} \in D_i$. We also enriched the DTM with features referencing the number of terms in each field to help the model scale term importance based on the field length.

2.4 Label prediction

We used a Bayesian Additive Regression Trees (BART) machine learning "classification model" (Chipman et al. 2010) (in the implementation of Kapelner and Bleich 2013) to predict the probability of a record of being relevant, given the information coded into the enriched DTM and the initial training set. We set up the BART model to use 2000 MCMC iterations (after 250 burn-in iterations) and 50 trees; we used a k value of 2 to regularize extreme prediction and let the model use missing fields in the DTM as features (Kapelner and Bleich 2015). Positive records are oversampled ten times to increase sensitivity (Batista, Prati, and Monard 2004).

The output is a posterior predictive distribution (PPD) for each record describing its probability of being relevant (i.e., a positive match). An ensemble of ten models was fitted to improve prediction stability by averaging the PPD between models (Zhou 2021; Dietterich 2000).

To assign the labels, we employed an "active learning" Miwa et al. (2014) approach, where a human reviews a specific subset of predictions made from the machine, which is then retrained on the manually reviewed data set. This process proceeds iteratively, decreasing prediction uncertainty.
Label assignment is done through the identification of an "uncertainty zone" whose construction is possible thanks to the Bayesian nature of BART, which provides full PPDs instead of point-wise predictions for each record.
To describe the process formally, we define

$$\pi_i = \frac{1}{M} \sum_{j=1}^{M} Pr(L_i = 1 | DTM, m_j)$$

as the PPD of a record D_i being relevant (i.e, having a positive label, $L_i = 1$), averaging the PPDs of the ensemble of $M = 10$ models m, and

$$\pi_{i,l} = \{\pi_i : Pr(\pi_i) = 1\%\}$$
$$\pi_{i,u} = \{\pi_i : Pr(\pi_i) = 99\%\}$$

as respectively the lower and upper boundaries of the 98% quantile interval of π_i (98% Predictive Interval, 98% PrI).
Then we identify the "uncertainty zone" as

$$U_\pi = [\max \vec{\pi}_u^-, \min \vec{\pi}_l^+]$$

with $\vec{\pi}_u^-$ being the vector of $\pi_{i,u}$ with a negative label and $\vec{\pi}_l^+$ the vector of $\pi_{i,l}$ with a positive label. That is, U_π defines a range of values between the smallest $\pi_{i,l}$ in the set of already labelled positive records L_p and the largest $pi_{i,u}$ related to the negative ones L_n, noting that the two limits can appear in any order.
Consequently, a record D_i will be labelled as positive if

$$\pi_{i,l} > \max_{\pi \in U_\pi} \pi$$

that is, the record lower 98% PrI boundary should be higher than every value in the uncertainty zone. In other words, for a record to be labelled positive, its PPD should be within the range of the mixture of PPD of the previously labelled positive records and not cross the distributions of the negative records.
Conversely, a record is labelled as negative if

$$\pi_{i,u} < \min_{\pi \in U_\pi} \pi$$

All other records are labelled as "uncertain".

Manual review is then necessary for: 1) uncertain records, 2) positive records (to avoid false positives), 3) records whose predicted label differs from the existing manual one. The last case helps identify human errors or inconsistent labelling criteria.

The automatic classification and the manual review steps alternate in a loop (CR iterations) until no new positive matches are found in four consecutive iterations.

2.5 Relevant term extraction

BART model it is possible to extract the proportion of times a term was used in a posterior tree over the sum of total inclusions of all variables (Kapelner and Bleich 2013), as a measure of feature importance for the model. We extracted the terms, the part of the citation data they were used in, the average "inclusion rate" among the ensemble models (over 10,000 inclusions) and its ratio over the standard deviation of this

inclusion (inclusion stability, IS). For each term we ran a Poisson regression to get its linear association with a positive label and reported it as Relative Risk (RR) with the number of standard errors as significance index (Statistic); the comparison between the inclusion rate in the BART models and the linear association allows to spot relevant non-linear effects (i.e, the feature is relevant only in association with others). We reported in the Results only the first (in order of inclusion rate) 15 terms with IS > 1.5 while the first fifty terms regardless of inclusion stability are reported in Supplemental Material S2.

2.6 New search query generation

We created an algorithm that generates a new search query to acquire further relevant publications missed during the first search, possibly at a reasonable cost in specificity (i.e., a higher number of negative results). The algorithm encompasses a number of steps:

- We fit a partition tree (Therneau and Atkinson 2019) between the DTM and 800 samples from the PPD; if a term is present multiple times in the DTM (e.g., both title and abstract), they are counted just one, and field term count features are removed. This step generates a list of rules composed by AND/NOT "conditions" made of terms/authors/keywords/MESH tokens, which together identify a group of records.
- For each rule, negative conditions (i.e., NOT statements) are added iteratively, starting from the most specific one, until no conditions are found that would not also remove positive records.
- The extended set of rules is sorted by positive-negative record difference in descending order. The cumulative number of unique positive records is computed and used to group the rules. Rules inside each group are ordered by specificity.
- The researcher is then asked to review the rule groups, selecting one or more rules (useful if they convey different meaning) from each, or edit them (in case too specific positive or negative conditions were included). It is possible to exclude a group of rules altogether, especially those with the worse sensitivity/specificity ratio.
- The selected rules are joined together by OR statements, defining a subset of records with a sensibly higher proportion of positive records than the original one.
- Redundant rules (i.e., rules whose positive records are already included in more specific ones) and conditions (i.e., conditions that once removed do not decrease the total number of positive or do not increase the negative records) are removed.
- Finally, the rules are re-elaborated in a query usable on the major scientific databases.

Since the algorithm is data-driven, it creates queries that effectively select positive records from the input data set but may be not specific enough once applied to actual research databases. Therefore we appended an extra subquery in AND, which specifies the general topics of our search and delimitates the search domain. The new query was used to initiate a second search session.

2.7 Performance evaluation

We trained a simple Bayesian logistic regression (surrogate model) on the reviewed records to evaluate the classification model consistency (see Discussion for the theoretical justification). The surrogate model uses as predictor the lower bound of the 98% PrI of the records' PPD with weakly regularizing, robust priors for the intercept (Student T with $\nu = 3, \mu = 0, \sigma = 2.5$) and the linear coefficient (Student T with $\nu = 3, \mu = 0, \sigma = 1.5$).
The quality of the model was evaluated through Bayesian R^2 (Gelman et al. 2019), of which we reported the posterior median and 90% Credible Interval [90% CrI]. The R^2 also provides an evaluation of the consistency of the original classification model. Given that this model is conditional only on the BART predictions and not on the DTM, it is characterized by more uncertainty, providing plausible worst-case scenarios.
The surrogate model is then used to generate the predictive cumulative distribution of the number of total positive records in the whole data set. This distribution allows estimating the expected total posterior "Sensitivity" and "Efficiency" of the classification model in the full (unreviewed) data set. Efficiency is summarised by the "Work saved over random" (WSoR) statistic: one minus the ratio between the number of record manually reviewed and those that would be required to find the same number of positives if classification was performed choosing records randomly; this last quantity is estimated through a negative hypergeometric distribution (Chae 1993) over the predicted amount of positive records.

For the number of predicted positive records, the sensitivity and the efficiency, we reported the "truncated 90% PrI" [trunc. 90% PrI], which is the uncertainty interval bounded at the number of observed total positive records (i.e., there cannot be less predicted positive records than observed).

2.8 Hyperparameter evaluation

Our classification algorithm has a number of hyperparameters:

- Size of the initial training set: 50, 100, 250, 500 records;
- Number of models in the ensemble: 1, 5, 10, 20, 40, 60 repetitions;
- Oversampling rate of positive records: (1x (i.e., no oversampling), 10x, 20x;
- PrI quantiles for building the uncertainty zone: 80%, 90%, 98%;
- Source of randomness between models in the ensemble: MCMC sampling only (Robert, Casella, and Casella 2004), MCMC plus data bootstrapping (Breiman 1996) of the training set.

To evaluate the hyperparameter effect of performance, we set up a "grid search" (Claesen and De Moor 2015; L. Yang and Shami 2020) on a prelabelled "validation set" made of the first 1200 records from the first session data set. The framework tested each hyperparameter combination until four CR iterations with no positive records were returned or the whole data set got labelled.
For each combination, a performance score was computed as the product of "Efficiency" (1 minus the ratio of records that required review over the total) and "Sensitivity" (number of positive records found over the total of positives). We then identified homogeneous "performance clusters" of scores given hyperparameter values using a partition tree (Therneau and Atkinson 2019). For the rest of the study, we used the best hyperparameter set in terms of Sensitivity followed by Efficiency from the cluster with the highest average score.

3 Results

3.1 First session

The initial search query for the example topic was:

((model OR models OR modeling OR network OR networks) AND (dissemination OR transmission OR spread OR diffusion) AND (nosocomial OR hospital OR "long-term-care" OR "long term care" OR "longterm care" OR "long-term care" OR "healthcare associated") AND (infection OR resistance OR resistant))

selecting only results between 2010 and 2020 (included). Results were collected from Pubmed, WOS, IEEE, EMBASE and SCOPUS, using the framework tools as described in Methods and Supplemental Material S1.

The first search session returned a total of 27600 records, specifically 12719 (71.6% of the total) records from the EMBASE database, followed by 9546 (53.8%) from Pubmed, 3175 (17.9%) from SCOPUS, 2100 (11.8%) from WOS, and 60 (0.34%) from IEEE (Table 1). There were various degrees of overlapping between sources, with the 38.4% of records being present in more than one database and EMBASE and IEEE being the databases with the higher uniqueness ratios. The final data set was composed by 17755 unique records. The first 250 records (based on "simple query ordering") were manually labeled. Of these 43 (17.2%) were labeled as positive, and 207 (82.8%) as negative.

The labeled records were used to train the Bayesian classification model used to label the remaining records. After seven classification and review (CR) iterations (three resulting in new positive matches and four extra replications to account for stochastic variability), a total of 101 positives matches were found, requiring manual review of 766 records (13.2% positivity rate).
It is possible to observe how the number of records that required manual review dropped rapidly between iterations (Table 2), indicating that the engine was converging while the uncertainties were resolved.
This phenomenon is better depicted in Fig. 1 of the Supplemental Material S2, showing the mixture distribution of the PPDs of the records, specifically for the reviewed positive and negative records, and for records that need manual review after the classification step: it can be noticed how the distribution of the uncertain records shrinked (they concentrate in a shorter probability range) and shifted toward the negative zone as more positive matches are found and reviewed.

Table 1. Distribution of retrieved records by source and session. For each source it is reported the number of records, percentage over the session total (after removing duplicates), and number or records specific for a source as absolute value and as percentage over the source total. All session shows records after joining and deduplication of the Session 1 and Session 2 data set.

Session	Source	Records	% over total	Source specific records	% over source total
Session1	Total	17755			
	Embase	12719	71.6%	6683	52.5%
	Pubmed	9546	53.8%	3457	36.2%
	Scopus	3175	17.9%	298	9.39%
	WOS	2100	11.8%	473	22.5%
	IEEE	60	0.34%	29	48.3%
Session2	Total	82579			
	Embase	48396	58.6%	40826	84.4%
	Pubmed	28811	34.9%	18021	62.5%
	Scopus	17070	20.7%	4908	28.8%
	WOS	12956	15.7%	2817	21.7%
	IEEE	61	0.074%	22	36.1%
All Sessions	Total	98371			
	Embase	59604	60.6%	46942	78.8%
	Pubmed	37278	37.9%	21371	57.3%
	Scopus	19353	19.7%	5181	26.8%
	WOS	14367	14.6%	3175	22.1%
	IEEE	108	0.11%	48	44.4%

Table 2. Results of the automatic classifaction and manual review rounds. For each iteration, the cumulative number of positives and negative records and their sum (Total labelled) and percentage over total are shown. Also, the number of changes after review and their description is reported."Unlab." indicates unlabelled records marked for review. For each Iteration, also the number of features used by the engine is reported. The first row reports the results of the initial manual labelling of records, which acted as input for the automatic classification in Iteration 1. In Session 2, the engine uses the labels at the end of Session 1 to classify the newly added records.

Session	Iteration	Positives	Negatives	Total labelled (%)	Unlab. -> y	Unlab. -> n	Unlab. -> *	n -> y	Changes	N. features
Session1 (n = 17755)	Initial labelling	43	207	250 (1.41%)	43	207	0	0	250	2289
	1	93	529	622 (3.5%)	50	322	0	0	372	2289
	2	100	614	714 (4.02%)	6	86	0	1	93	3750
	3	101	625	726 (4.09%)	1	11	0	0	12	3834
	4	101	648	749 (4.22%)	0	23	0	0	23	3856
	5	101	651	752 (4.24%)	0	3	0	0	3	3856
	6	101	660	761 (4.29%)	0	9	0	0	9	3856
	7	101	665	766 (4.31%)	0	5	0	0	5	3856
Session2 (n = 98371)	1	106	934	1040 (1.06%)	5	270	998	0	1273	4729
	2	107	1123	1230 (1.25%)	1	189	0	0	190	4729
	3	107	1176	1283 (1.3%)	0	53	0	0	53	4733
	4	107	1200	1307 (1.33%)	0	24	0	0	24	4729
	5	107	1209	1316 (1.34%)	0	9	0	0	9	4729
	6	107	1226	1333 (1.36%)	0	17	0	0	17	4729

We extracted the 15 term more relevant for the classification model, described as: Term (citation part): Inclusion Rate (Inclusion Stability) [linear Relative Risk, Statistic].

Patient Transport (Keyword): 61.2 (3.77) [99.1, 21.3], Transfer (Abstract): 57 (3.93) [22.5, 15.4], Network (Title): 56.5 (2.91) [18, 14.2], Network & Patient (Abstract): 54.2 (4.66) [26.3, 15.2], Donker T (Author): 53.5 (4.56) [159, 16.5], Worker (Abstract): 50 (3.33) [0.421, -1.21], Hospitals (Keyword): 49.8 (4.31) [27.8, 16.5], Movement (Abstract): 47.8 (2.7) [27.2, 15], Spread (Title): 46.6 (2.25) [16.2, 12.1], Facility (Abstract): 45 (2.22) [19.6, 14.8], Orange County (Keyword): 44.3 (3.19) [199, 17.2], Conduct (Abstract): 42.6 (3.7) [0.221, -2.57], Patient (Abstract): 42 (3.61) [27.6, 7.23], Perform (Abstract): 41.9 (2.38) [0.342, -2.55], Hospital (Title): 39 (1.95) [12.5, 12.5].

The "&" indicates nc-ngrams, i.e., terms strongly co-occurrent in the documents.
The engine was able to pick up the central concept of the research, i.e., "patient transport" or "transfer" through a "network" of "facility"ies that facilitates the "spread" of infections, and even one of the authors of this study (Donker T.) as well as the region of interest ("Orange County") of another research group active on the topic of healthcare asociated pathogen spreading over hospital networks. Some terms were considered highly relevant for the BART models (e.g., "Worker" in 6th position out of more than 3800 terms considered) although in a simpler linear model their effect would be hardly significant (statistic: -1.21 s.e.); these are terms which are relevant only in conjunction with other terms but not by themselves, highlighting the extra predictive power brought by a non-linear model like BART.
A more extensive set of terms is presented in Table 1 of Supplemental Material S2.

3.2 Second session

The results of the first classification session were used to create a second, data-driven query with the purpose of performing a more large-spectrum search to find records which may have escape the first search session. The resulting query was the following:

(((Donker T) NOT (bacterium isolate)) OR ((network patient) AND (resistant staphylococcus aureus) NOT (monte carlo) NOT isolation) OR (facility AND (network patient) AND regional NOT hospitals NOT increase NOT (patient transport) NOT (control infection use)) OR ((patient transport) NOT (Donker T) NOT worker) OR (hospitals AND (network patient) NOT (patient transport) NOT regional NOT clinical) OR (facility AND (network patient) NOT hospitals NOT (patient transport) NOT regional NOT prevention NOT medical) OR ((healthcare facility) NOT (Donker T) NOT worker NOT positive) OR (hospitals NOT (network patient) NOT medical NOT environmental NOT outcome NOT global) OR ((network patient) NOT facility NOT hospitals NOT (patient transport) NOT therapy NOT global)) AND ((antimicrobial resistance) OR (healthcare infection))

The final piece *AND ((antimicrobial resistance) OR (healthcare infection)* was added manually to better define the search domain, since the algorithm was trained on documents that were all more or less related to these topics.
The generated query also provides a more nuanced understanding of the engine's internal classification logic, and this is helpful to spot possible biases in the model.

The search was done with the same year filter and procedures of the first session.

The new search produced 107294 records (Table 1), of which 48396 (58.6%) from the EMBASE, followed by 28811 (34.9%) from Pubmed, 17070 (20.7%) from SCOPUS, 12956 (15.7%) from WOS, and 61 (0.074%) from IEEE; compared with the first session, the relative weight of EMBASE and Pubmed was decreased, while the amount of content specificity was greatly increased, as it was for SCOPUS. After removal of duplicates, 82579 unique records were obtained. Once joined with the session 1 records and duplicates removed, we obtained 98371 unique records, with just 1963 shared records between searches, that is the 2%. The percentage of records shared by two or more source dropped to 22%.

Six CR rounds were necessary to complete the second session classification, with just 6 new positive found after reviewing 568 extra records. The first CR iteration required the user to review a substantial number of records (1,273), but just labelling 275 of them (the canonical 250 plus 25 that were already labelled during the framework hyperparameter tuning) was sufficient to drop this number to just 190 in the subsequent round. An evaluation of the convergence (Figure 1, Supplemental Material S2) showed that, in addition to the dynamics already observed in session 1 (shrinkage and negative shift), a second mode appeared in the mixture distribution of the records to be reviewed, centred in a highly positive zone. The interpretation is that as the number of negative training records increases, the engine gets more and more skeptical and asks to review even some records labelled as positive in the initial training set generated during Session 1. This behaviour can be useful to spot classification errors and inconsistencies. Considering both sessions, 1333 records were reviewed and 107 (8.03%) were found.

Again, the evaluation of the inclusion rate of the terms showed that the engine was quite capable of internalizing the concepts behind the research topic A subsample of the terms is reported in Table 2 of Supplemental Material S2.

3.3 Hyperparameter selection

As described in the methods, the selection of hyperparameters was achieved via evaluation of sensibility and efficiency through a grid search on a validation set of 1200 completely manually labelled records. The best set of parameters suggested an initial input of 250 labelled records with 10x positive matches oversampling, an averaged ensemble of 10 models, no bootstrapping and an uncertainty zone defined by the 98% predictive interval. On the validation set, this combination of parameters reached a sensitivity of 98.8% (81 / 82 positive matches found) and efficiency of 61.5% (462 / 1200 records evaluated). The results of the hyperparameter tuning are reported in Table 3 of Supplemental Material S2. Figure 2 in Supplemental Material S2 demonstrates that the positive record oversampling rate, the number of ensemble models and the size of the initial training set were the parameters that most impact performance.

3.4 Performance evaluation

To evaluate the theoretical performance of the engine, a surrogate Bayesian logistic regression model was trained on the manually reviewed labels using only the lower bound of the record PPDs as predictor (see Methods for details). The surrogate model show the high predictive power of the scores produced by the classification model (Bayesian R2: 98.1% [97.4%, 98.3%] for session 1 and 98.2% [97.6%, 98.3%] for session 2).

Figure 2 presents the actual and predicted (from the surrogate model) cumulative number of positive matches, ordered by the initial simple ordering query: the median of surrogate models' cumulative predictive distributions matches quite well the actual number of positive records found. It is striking how many more records would have needed to be evaluated manually to find the same number of positive matches without using a smart classification tool; some relevant record was found even close to the end of the heuristically ordered list of records.

Table 3 shows various performance indexes for both sessions, both descriptive (Total records, Reviewed records, Observed positive matches) and estimated through the surrogate model (Expected efficiency, Predicted positive matches, Expected sensitivity, R^2).
In session 1 we observe an expected total number of positives of 101 [101, 108] for an estimated sensitivity of 100% [93.5%, 100%] and efficiency of 95.7% [95.3%, 95.7%]. In session 2 we observed a drop in the expected sensitivity, especially in the lower margin (97.3% [73.8%, 100%]), due to the fact that as the number of records grows, even a small probability can translate, in the worst case scenario, into a relevant number of missed positive matches (145 in this case). To ascertain that no evident positives were missed, we evaluated 100 more records between the unreviewed ones with the highest median predicted probability produced by the engine and found no additional positive matches.

Table 3. Estimated performance summary. The table reports for each session, the number of reviewed records and the percentage over the total. Also, the posterior expected number of positive records, "Sensitivity" and "Efficiency" (as WSoR) are reported, with their 90% PrI truncated to the observed realization in the dataset [trunc. PrI] (see. methods). Finally the median Bayesian R^2 [90% CrI] of the logistic models is reported. PrI: Predictive Intervals; CrI: Credibility Intervals.

Indicator	Session 1	Session 2
Total records	17755	98371
Reviewed records (% over total records)	761 (4.29%)	1316 (1.34%)
Expected efficiency (over random) [trunc. 90% PrI]	95.7% [95.3%, 95.7%]	98.6% [98.2%, 98.7%]
Observed positive matches (% over total records)	101 (0.57%)	107 (0.11%)
Predicted positive matches [trunc. 90% PrI]	101 [101, 108]	110 [107, 145]
Expected sensitivity [trunc. 90% PrI]	100% [93.5%, 100%]	97.3% [73.8%, 100%]
Simple Model R^2 [90% CrI]	98.1% [97.4%, 98.3%]	98.2% [97.6%, 98.3%]

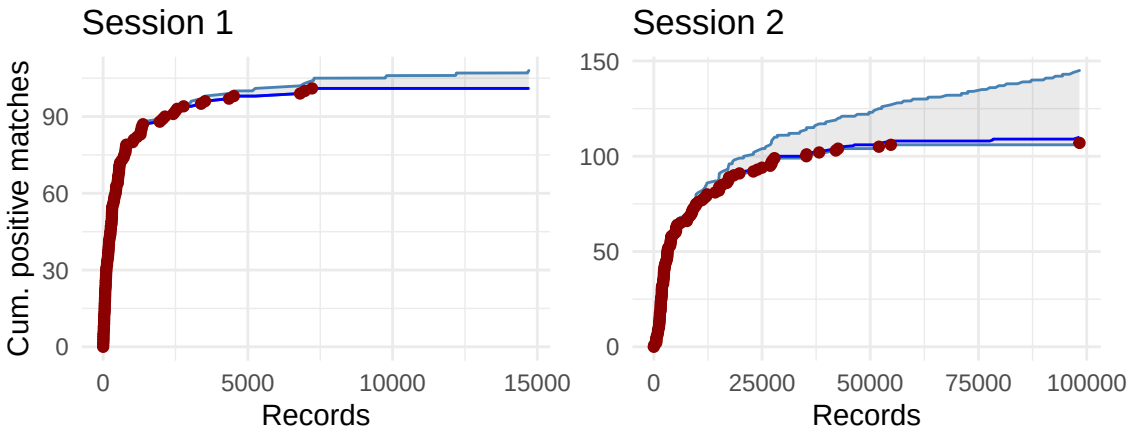

Figure 2. Observed cumulative number of positive matches (red dots) sorted by simple query ordering. The [trunc. 90% PrI] of the cumulative positive matches estimated by the logistic Bayesian model is shown as shaded area delimited by the 95% quantile of the PrI and by the observed number of positive matches (light blue lines). The median of the PrI is represented by a darker blue line.

4 Discussion

We propose a new integrated framework to help researchers collect and screen scientific publications characterised by high performance and versatility, joining the growing field of systematic review automation (SRA) and helpers (SRH) tools (A. M. Cohen et al. 2006, 2010; Ananiadou et al. 2009; O'Mara-Eves et al. 2015). This framework joins standard approaches and uses ad-hoc solutions to deal with common SRA issues. By freely sharing the tool as an open-source R package and by following a modular design, we tried to adopt some of the so-called Vienna Principles advocated by the International Collaboration for the Automation of Systematic Reviews (ICASR) (E. Beller et al. 2018).
The framework consists of four main components: 1) an integrated query-based citation search and management engine, 2) a Bayesian active machine learning-based citation classifier, and 3) a data-driven search query generation algorithm.

The framework's search engine module is capable of automatically collecting citation data from three well-known scientific databases (i.e., Pubmed, Web of Science, and the database of the Institute of Electrical and Electronics Engineers) as well as process manually downloaded results from both the mentioned and other (SCOPUS, EMBASE) databases. In comparison, most SRH tools, commercial or free to use, rely either on internal databases (e.g., Mendeley `https://www.mendeley.com/`) sometimes focusing just on a particular topic (Visser 2010) or on a single external data source (Thomas and Brunton 2007; Poulter et al. 2008; Soto, Przybyła, and Ananiadou 2019).
Mixing different databases is fundamental to have a more comprehensive view of the literature (Bajpai et al. 2011; Wilkins, Gillies, and Davies 2005; Woods and Trewheellar 1998): in our results, 18.7% of the positive matches found were unique for one of the different data sources, and no positive record was present in all of them (data not shown).
The online search algorithms are efficient enough to manage tens of thousands of search results, using various expedients to overcome the limitations of the queried databases in terms of traffic and download quota. The results are then automatically organized, deduplicated and arranged by "simple query ordering" in a uniform corpus. The preliminary ordering allow to increase the positivity rate in the initial training set (Wallace, Small, et al. 2010).

For the framework's record classification module we employed an active machine learning approach Miwa et al. (2014) based on the best practices from other SRA studies, bringing further improvements at various levels.
The feature extractor module uses modern NLP techniques (Ananiadou and McNaught 2006; K. B. Cohen and Hunter 2008) to transform text into input data for machine learning. We did not include classical n-grams (Schonlau and Guenther 2017), but we used network analysis to find non-consecutive frequently associated terms, a generalisation of n-grams relaxing the term adjacency assumption. This approach allows

also to find term connections across fields, for example a term having a different relevancy if associated with a specific author. The same technique but with different parameters was applied to merge redundant terms to make model estimation more efficient and reduce noise.
The use of concurrency network-driven modelling of text is not new (Francois Rousseau 2015; Violos et al. 2016; François Rousseau, Kiagias, and Vazirgiannis 2015; Ohsawa, Benson, and Yachida 1998) and is a valuable tool to extract semantic information not evident in one-word or consecutive n-gram models.

The automatic classification algorithm is based on Bayesian Additive Regression Trees (BART) (Chipman et al. 2010; Kapelner and Bleich 2013). As with other boosted trees algorithms (Hastie, Tibshirani, and Friedman 2009), BART can explore complex non-linearities, perform variable selection, manage missing data while sporting high performance in predictive power.
However, its Bayesian foundation provides further benefits: less sensitivity on hyperparameter choices, natural regularisation through priors, and, especially, predictive distributions as output in place of point-wise predictions (Soria-Olivas et al. 2011; Joo, Chung, and Seo 2020; Jospin et al. 2020). By selecting relatively tight prior distributions, we discouraged excessively deep trees, long sequences of trees, or extreme predicted probabilities, to decrease the risk of overfitting.
The algorithm runs multiple replications of the model and averages their predictive distributions creating an "ensemble"; this technique has been shown to improve out-of-sample predictive performance (Zhou 2021; Dietterich 2000), as we were able to confirm during the hyperparameter evaluation (Supplemental Material S2). Ensembling reduces the uncertainty in the predictive distribution tails related to the randomness in the MCMC fit (Robert, Casella, and Casella 2004), generating a shift of probability mass towards the distribution centre and stabilising it (i.e., decreasing variance without impacting bias). On the other hand, just imposing robust uninformative priors against extreme predictions would have decreased variance but also shifted the distribution towards a non-decision zone, increasing bias (Hansen et al. 2000).
Since the number of model replications significantly impacts computation times, we decided to use ten replicas, the lower value after which showed performance stabilisation during the hyperparameter evaluation.
We also investigated whether bootstrapping between replications (Breiman 1996) would improve performance, but, contrary to theory (Díez-Pastor et al. 2015), it was demonstrated to be slightly detrimental (Supplemental Material S2) compared to simple ensembling.

A low rate of relevant matches (class imbalance) is typical in literature reviews (Sampson, Tetzlaff, and Urquhart 2011; Wallace, Trikalinos, et al. 2010; O'Mara-Eves et al. 2015), and such strong imbalance between positive and negative records can affect sensitivity (Khoshgoftaar, Van Hulse, and Napolitano 2010; Chawla, Japkowicz, and Kotcz 2004).
To overcome the problem, we oversampled (Batista, Prati, and Monard 2004) the positive records ten times before model fitting. Our hyperparameter analysis showed that together with model ensembling, the oversampling rate was the parameter with the highest impact on performance.
A known risk with positive oversampling is the misclassification of negative records (Ramezankhani et al. 2016). However, since all predicted positives get manually reviewed in our approach, we are always ensured to achieve 100% specificity/positive predictive value: the only price for the increased sensitivity due to oversampling is a larger number of records to review.
An alternative to oversampling would be applying different weights and/or cost to the classes (Abd Elrahman and Abraham 2013; Díez-Pastor et al. 2015), but the BART implementation we used did not have this feature; also, using simple oversampling permits broader compatibility with different modelling engines (Galar et al. 2011; Roshan and Asadi 2020).
Finally, ordering the records by query term frequency (simple query ordering) generates a far higher rate of relevant records in the initial training set (17.2%) compared to the overall data (0.11%), and this boosts the sensitivity of the model.

One of the central innovations we introduced is the concept of "uncertainty zone", whose implementation is possible thanks to the Bayesian foundation of the classification model. This construct guides the selection of the records to review, dynamically updating and shrinking after every CR iteration, as more uncertain predictions are evaluated (Supplemental Material S2 Fig. 1).
This approach overcomes the usual requirement of dataset-specific hard thresholds in active machine learning, and also allows to review multiple items at once between iterations (Laws and Schütze 2008; Miwa et al. 2014; Zhu et al. 2010). The parameters our algorithm needs are instead quite general and non task-specific, like the PPD intervals based on which the uncertainty zone is built ,and the maximum number of iterations

with no positive matches after which a session is concluded; the hyperparameter evaluation shows that the algorithm is robust against variations in these parameters and we expect the default values to preform well on most datasets.
Since researchers are asked to review both records with a positive predicted label and those inside the uncertainty zone, this method can be considered as a unifying synthesis of the "certainty" and "uncertainty" paradigms of active learning (Miwa et al. 2014).

We evaluated performance as the capability of the screening procedure (automatic classification plus manual review) to find the largest number of relevant records while reviewing as few of them as possible (i.e., sensitivity × efficiency).
We avoided the classic out-of-sample approaches like train-test sampling, out-of-bag bootstrapping or cross-validation (Kohavi et al. 1995; James et al. 2013). Such methods primarily assume that the rate of positivity is equal on average in every random subset of the data (Tashman 2000); this uniformity is broken by how the initial training set and the subsequent reviewed records are selected by the query-based ordering and the active learning algorithm, determining a lower positivity rate in the unlabelled records (Fig. 2). Also, a literature corpus is unique per search query/database combination, and therefore any out-of-sample performance estimate is not replicable since no new data can be acquired related to the current corpus.
Instead, to estimate overall sensitivity, we employed simple Bayesian regression (surrogate model) on the manually reviewed data to abstract the classification model predictions and achieve a maximum entropy (Harremoës and Topsøe 2001) estimate of the number of missed positive matches among the unreviewed records in the whole dataset. This simple surrogate model fitted the data very well (R^2 consistently above 97%) using just the lower 98% PrI bound of the PPDs as predictor, indicating predictive consistency in the classification model. The surrogate model posterior predictive distribution could be exploited to explore worse case scenarios in terms of sensitivity.

Our framework achieved very high sensitivity by screening a markedly small fraction of all records, bringing a sensible reduction in workload.
Based on the surrogate model, we predicted a predicted median sensitivity of 100% [93.5%, 100%] in the first session (screening 4.29% of records) and of 97.3% [73.8%, 100%] in the second (screening 1.34% of records): efficiency increased significantly in the second session since only a few new positive matches were found, but given the large number of records, uncertainty regarding sensitivity also expectedly increased.
Both results are above the usual performance in the field (O'Mara-Eves et al. 2015) and in line with the 92% average sensitivity estimated after human-only screening (Edwards et al. 2002). In one interesting case, the model spotted a human-made misclassification error, demonstrating its robustness and value as a second screener, a role already suggested for SRA tools by previous studies (Frunza, Inkpen, and Matwin 2010; Bekhuis and Demner-Fushman 2012, 2010). Finally, albeit the simple query ordering already concentrated most of the relevant matches in the first 20-25 thousand records, without the tool support some relevant records would have required almost the complete data set to be manually checked to be found.

The model took ~5-20 minutes per iteration to perform predictions in session 1 (17,755 documents) and 20-40 minutes in session 2 (98,371 documents) on an eight-core, 2.5 GHz, 16 GB RAM laptop from 2014; including manual record review, one session required 1-3 days of work, for a total of 1-2 weeks for the whole process (including record collection). That is a considerable saving of time compared to the multiple months usually required for the screening phase of systematic reviews (Bannach-Brown et al. 2019; Borah et al. 2017; Allen and Olkin 1999). To our knowledge, the amount of data processed (~100.000 records) were larger than what is typical in most SRA studies (O'Mara-Eves et al. 2015; Olorisade et al. 2016), emphasising the reliability of the tool in real-world scenarios.

The last module of our framework is a data-driven query generation algorithm. Creating an efficient and efficacious search query is a complex task (Lefebvre et al. 2011; Hammerstrøm et al. 2010) since it requires building a combination of positive and negative terms to maximise the number of relevant search results while minimising the total number of records to review. Our solution joins a sensitivity-driven subquery proposal engine based on concurrent decision trees (Blanco-Justicia and Domingo-Ferrer 2019; Moore et al. 2018) built on the BART ensemble PPD with a human review step and an efficiency-driven query builder. The aim is to generate a second query that helps find records missed during the first session search. The generated query allowed indeed to retrieve few more positive matches not found in session 1, but at the cost of a significant increase in the number of documents.

One interesting aspect of this functionality is that it provides a human-readable overview of the classification rules learned by the classification model, showing which combination of terms was particularly relevant and even spotting authors and geographical locations associated with the study topic. The generated query, therefore, acted as a tool for machine learning explainability (Bhatt et al. 2020; Burkart and Huber 2021), a feature useful to understand and spot bias in black-box classification algorithms (Malhi, Knapic, and Främling 2020); explainability is often required or even legally mandatory for high-stake machine learning applications (Bibal et al. 2021, 2020).
It is important to note that this process is entirely data-driven. The algorithm is only aware of the "world" defined by the data set used as input, which is generated by a specific search query focused on a particular topic. Therefore, the new query may not be specific enough once applied to an unbounded search domain, returning an unmanageable amount of unrelated results. The solution we found was to add another component to the query, specifying the general topic (antimicrobial resistance and healthcare-associated infections) of our research.

As reported, our framework builds on modularity. We designed it to easily implement complete independence of the main modules in future iterations, making it possible for users to add custom features like citation search and parsing for other scientific databases, alternative text processing algorithms or machine learning modules. We deem such interoperability extremely relevant because the main strength of our tool is the composition of many solutions and the general idea of Bayesian active machine learning and uncertainty zone. However, each of its components could benefit considerably from the recent improvements in text mining.
For example, our text processing approach is quite simple, based on the boolean bag-of-words paradigm, and indeed could be improved by more nuanced text representations. It could be evaluated if feature transformations like TF-IDF (Baeza-Yates, Ribeiro-Neto, et al. 1999; Ananiadou and McNaught 2006) would be advantageous, even if we hypothesise that tree-based classification algorithms like BART are robust enough not to need such operations. Word embedding could be worth exploring: this technique transforms terms in semantic vectors derived from the surrounding text Minaee et al. (2021) and could be used to eliminate semantically redundant terms or differentiate identical terms with different meanings given the context. Another option would be to employ unsupervised learning models like Latent Dirichlet Analysis, Latent Semantic Analysis (Pavlinek and Podgorelec 2017; Q. Chen, Yao, and Yang 2016; Landauer, Foltz, and Laham 1998) or graph-of-word techniques (Ohsawa, Benson, and Yachida 1998; Francois Rousseau 2015) to extract topics to enrich the feature space.
Our classification algorithm can be implemented with any Bayesian supervised machine learning method that provides full PPDs; therefore alternative classification models could be evaluated, like Gaussian Processes which are known for their flexibility (Jayashree and Srijith 2020; S.-H. Chen et al. 2015). Even more interesting would be to test advanced learning algorithms that surpass the bag-of-words approach, taking into consideration higher-level features in the text like term context and sequences, long-distance term relationships, semantic structures, etc., (Cheng et al. 2019; Minaee et al. 2021; Li et al. 2020; J. Yang, Bai, and Guo 2020; Lai et al. 2015; Farkas 1995), given that a Bayesian implementation of such algorithms is available (for example C. Chen, Lin, and Terejanu (2018)).
Finally, a natural improvement would be to provide a graphical user interface to make the framework easy to use also for less technical users.

The field of literature review automation is maturing rapidly, and we expect an increasing use of such technologies to manage the ever-faster rate of scientific production. We believe it is appreciable that a multiplicity of tools are being made available to let researchers and policymakers find the instrument that better fits their needs.
We contribute to this field with an innovative framework that provide excellent performance and easy integration with existing systematic review pipelines. The value of this work lies not only in the framework itself, which we provide as open-source software, but in the set of methodologies we developed to solve various SRA issues and that can be used to improve already existing solutions.

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