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

Abstract

The exponential growth of scientific production makes secondary literature abridgements increasingly demanding. We introduce a new open-source framework for systematic reviews that significantly reduces time and workload for collecting and screening scientific literature. The framework provides three main tools: 1) an automatic citation search engine and manager that collects records from multiple online sources with a unified query syntax, 2) a Bayesian, active machine learning, citation screening tool based on iterative human-machine interaction to increase predictive accuracy and, 3) a semi-automatic, data-driven query generator to create new search queries from existing citation data sets.

To evaluate the automatic screener’s performance, we estimated the median posterior sensitivity and efficiency [90% Credible Intervals] using Bayesian simulation to predict the distribution of undetected potentially relevant records. Tested on an example topic, the framework collected 17,755 unique records through the citation manager; 766 records required human evaluation while the rest were excluded by the automatic classifier; the theoretical efficiency was 95.6% [95.3%, 95.7%] with a sensitivity of 100% [93.5%, 100%].

A new search query was generated from the labelled dataset, and 82,579 additional records were collected; only 567 records required human review after automatic screening, and six additional positive matches were found. The overall expected sensitivity decreased to 97.3% [73.8%, 100%] while the efficiency increased to 98.6% [98.2%, 98.7%].

The framework can significantly reduce the workload required to conduct large literature reviews by simplifying citation collection and screening while demonstrating exceptional
sensitivity. Such a tool can improve the standardization and repeatability of systematic reviews.

**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 accelerated by the COVID-19 pandemic, thanks to faster article processing time by publishers and the greater use of preprint databases (Aviv-Reuven and 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 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 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; Tsafnat et al. 2014; Higgins et al. 2019).

Search queries construction is a complex task (Lefebvre et al. 2011; Hammerstrøm et al. 2010), 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 and 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 (Bannach-Brown et al. 2019; Borah et al. 2017; Allen and Olkin 1999). 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 (Marshall and Wallace 2019; E. Beller et al. 2018; 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 and McNaught 2006; K. B. Cohen and Hunter 2008); using machine learning, such text-derived data can be used to map and reproduce human judgment, automating the screening of citations (Ikonomakis, Kotsiantis, and Tampakas 2005).

Automation of systematic reviews has made significant 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 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 and 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 (Settles 2009; Miwa et al. 2014), 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.

2 Methods

2.1 General description

We built an R (R Core Team 2020) based framework to simplify two aspects of systematic literature review: record acquisition and classification. The code used to generate the results is available at https://github.com/AD-Papers-Material/BART_SystReviewClassifier, while an updated and ready to use version of the framework is distributed as an R package at https://github.com/bakaburg1/BaySREn. The framework includes several modules that communicate through intermediate outputs stored in standard formats, making 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 acquisition and initial labelling

We built a set of tools to allow users to 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 handles authorisation management for non-open databases like WOS and IEEE. It is also possible to import previously downloaded records in the framework; this is particularly useful for acquiring records from 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 also necessary 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 database supported is available in Supplemental Material S3.

The collected 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 subset of records to create the “initial training set” needed to start the automatic classification. We recommend manually labelling the first 250 records (see “hyperparameter optimisation” later). Simple query ordering increases the positivity rate in the initial training set (Wallace, Small, et al. 2010), leading to higher sensitivity during automatic classification (Chawla, Japkowicz, and Kotcz 2004).

2.3 Text feature extraction

The framework models the relevance of a record based on the following fields in the citation data: title, abstract, authors, keywords, MESH terms (Lipscomb 2000). A range of Natural Language Processing (NLP) techniques (Baeza-Yates, Ribeiro-Neto, et al. 1999; Marshall and Wallace 2019; Ananiadou and McNaught 2004) are employed to convert the textual information in these fields into features for machine learning through a bag-of-words approach (Marshall and Wallace 2019). Processing of free text fields (title, abstract) includes: tokenisation (i.e., extracting the terms), removal of common stopwords (i.e., sentence components having no semantic value), part-of-speech filtering (only nouns, adjectives, verbs and untagged terms are retained), and lemmatisation of terms (i.e., reduction to their base grammatical form). Text processing for authors, keywords and MESH terms identifies logical units (e.g., authors’ 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 they do co-occur. To detect nc-ngrams, we generated a word network representation (Francois Rousseau 2015) with edges occurring between terms with a cosine similarity in terms of document 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 are added to the dataset 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 can therefore be considered redundant (i.e., they do not need to be considered separately). These terms are merged to increase computational 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 being relevant, given the information coded into the enriched DTM and the initial training set. We set up the BART model to use 2,000 MCMC iterations (after 250 burn-in iterations) and 50 trees; we used a $k$ value of 2 to regularise 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 the probability of it 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” approach (Settles 2009; Miwa et al. 2014), where a human reviews a specific subset of predictions made by the machine, which is then retrained on the manually reviewed dataset. This process is carried out iteratively to reduce prediction uncertainty. Label assignment is done through identification of an “uncertainty zone”, the construction of which 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:

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

respectively as the lower and upper boundaries of the 98% quantile interval of \( \pi_i \) (98% Predictive Interval, 98% PrI).

Then we identify the “uncertainty zone” as:

\[ U_\pi = [\max \bar{\pi}_{u} - \min \bar{\pi}_{l}^+ \]

with \( \bar{\pi}_{u} \) being the vector of \( \pi_{i,u} \) with a negative label and \( \bar{\pi}_{l}^+ \) the vector of \( \pi_{i,l} \) with a positive label. That is, \( U_\pi \) 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 records previously labelled positive and should 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), and 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 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

As a measure of feature importance, we computed the “inclusion rate”, that is, the proportion of times a term is used in a posterior tree over the sum of total inclusions of all variables (Kapelner and Bleich 2013). We extracted the terms, the portion of the citation data in which they were used, 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 the 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). In the Results, we only listed the first 15 terms with IS > 1.5 (in order of inclusion rate), while the first fifty terms, regardless of inclusion stability, are listed in Supplemental Material S2.

2.6 New search query generation

We developed an algorithm that generates a new search query to find further relevant publications missed in the initial search, possibly at a reasonable cost to specificity (i.e., a higher number of negative results). The algorithm is composed of the following steps:

- A partition tree (Therneau and Atkinson 2019) is built between the DTM and 800 samples from the PPD; if a term is present multiple times in the DTM (e.g., both in the title and abstract), it is counted just once, 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 and select one or more rules from each group or edit overly specific rules (e.g., citing a non-relevant concept casually associated with a paper, like a numeric value or indicator). It is possible to exclude a group of rules altogether, especially those with the poorest 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 set.
- Redundant (i.e., rules whose positive records are already included in more specific ones) and non-relevant rules (i.e., conditions that when removed do not impact sensitivity and specificity) are removed.
- Finally, the rules are re-elaborated in a query that can be used to perform a new citation search.

Because the algorithm is data-driven, it creates queries that effectively select positive records from the input dataset but may be not specific enough when applied to actual research databases. Therefore we added an extra subquery in _AND_ that specifies the general topics of our search and narrows 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 consistency of the classification model (see Discussion for the theoretical justification). The surrogate model uses as predictor the lower boundary of the 98% PrI of the PPD of the records with weakly regularising, robust priors for the intercept (Student T with ν = 3, μ = 0, σ = 2.5) and the linear coefficient (Student T with ν = 3, μ = 0, σ = 1.5).

The quality of the model was evaluated through the 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 characterised 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 dataset. This distribution allows estimating the expected total posterior “Sensitivity” and “Efficiency” of the classification model in the whole (unreviewed) dataset. Efficiency is summarised by the “Work saved over random” (WSoR) statistic: one minus the ratio between the number of records manually reviewed and those that would be required to find the same number of positives if classification were performed choosing records randomly; this last quantity is estimated through a negative hypergeometric distribution (Chae 1993) over the predicted number of positive records.

For the number of predicted positive records, sensitivity and efficiency, we reported the “truncated 90% PrI”
2.8 Hyperparameter evaluation

Our classification algorithm has a limited 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 and Casella 2004), MCMC plus data bootstrapping (Breiman 1996) of the training set.

To evaluate the hyperparameter effect on performance, we set up a “grid search” (Claesen and De Moor 2015; L. Yang and Shami 2020) on a prelabelled “validation set” derived from the first 1,200 records of the first session dataset. Each hyperparameter combination was tested until four CR iterations were completed with no positive records or until the whole dataset was labelled. For each combination, a performance score was computed as the product of “Efficiency” (1 minus the ratio of records that required reviewing over the total number of records) and “Sensitivity” (number of positive records found over the total number of positive records). We then used a partition tree (Therneau and Atkinson 2019) to identify homogeneous “performance clusters” of scores given hyperparameter values. 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 the Methods and Supplemental Material S1.

The first search session returned a total of 27,600 records, specifically 12,719 (71.6% of the total) records from the EMBASE database, followed by 9,546 (53.8%) from Pubmed, 3,175 (17.9%) from SCOPUS, 2,100 (11.8%) from WOS, and 60 (0.34%) from IEEE (Table 1). There were various degrees of overlapping between sources, with 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 of 17,755 unique records. The first 250 records (based on “simple query ordering”) were categorised manually. Of these 43 (17.2%) were labelled as positive, and 207 (82.8%) as negative.

The categorised 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 noticeable how the number of records that required manual review decreased rapidly between iterations (Table 2), indicating that the engine was converging while the uncertainties were resolved. This phenomenon is better illustrated in Fig. 1 of Supplemental Material S2. It shows the mixture distribution of the PPDs of the records, specifically for records that were manually reviewed, before and after the classification step: it can be seen how the distribution of uncertain records shrinks (i.e., it becomes concentrated in a shorter probability range) and shifts 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, we reported the number of records, percentage over the session total (after removing duplicates), and the number of 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  | 17,755  |              |                         |                     |
|          | Embase | 12,719  | 71.6%        | 6,683                   | 52.5%               |
|          | Pubmed | 9,546   | 53.8%        | 3,457                   | 36.2%               |
|          | Scopus | 3,175   | 17.9%        | 298                     | 9.39%               |
|          | WOS    | 2,100   | 11.8%        | 473                     | 22.5%               |
|          | IEEE   | 60      | 0.34%        | 29                      | 48.3%               |
| Session2 | Total  | 82,579  |              |                         |                     |
|          | Embase | 48,396  | 58.6%        | 40,826                  | 84.4%               |
|          | Pubmed | 28,811  | 34.9%        | 18,021                  | 62.5%               |
|          | Scopus | 17,070  | 20.7%        | 4,908                   | 28.8%               |
|          | WOS    | 12,956  | 15.7%        | 2,817                   | 21.7%               |
|          | IEEE   | 61      | 0.074%       | 22                      | 36.1%               |
| All Sessions | Total | 98,371  |              |                         |                     |
|          | Embase | 59,604  | 60.6%        | 46,942                  | 78.8%               |
|          | Pubmed | 37,278  | 37.9%        | 21,371                  | 57.3%               |
|          | Scopus | 19,353  | 20.7%        | 5,181                   | 26.8%               |
|          | WOS    | 14,367  | 14.6%        | 3,175                   | 22.1%               |
|          | IEEE   | 108     | 0.11%        | 48                      | 44.4%               |

Table 2. Results of the automatic classification and manual review rounds. The cumulative numbers of positives and negative records and their sum (Total labelled) and percentage over total are shown for each iteration. Also, the number of changes after review and their description is reported. 'Unlab.' indicates unlabelled records marked for review. For each iteration, the number of features used by the engine is also reported. The first row reports the results of the initial manual labelling of records, which served 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 (n = 17755) | Iteration | Positives | Negatives | Total labelled (%) | Unlab. -> y | Unlab. -> n | Unlab. -> * | n -> y | Changes | N. features |
|---------------------|-----------|-----------|-----------|--------------------|-------------|-------------|-------------|--------|----------|-------------|
| Session1            | Initial labelling | 43        | 207       | 250 (1.41%)        | 43          | 207         | 0           | 0      | 250      | 2,289       |
| 1                   | 94        | 529       | 622 (3.5%) | 50                | 322         | 0           | 0           | 0      | 372      | 2,289       |
| 2                   | 100       | 614       | 714 (4.02%)| 6                 | 86          | 0           | 1           | 93     | 3,750    | 4,729       |
| 3                   | 101       | 625       | 726 (4.09%)| 1                 | 11          | 0           | 0           | 12     | 3,854    | 4,729       |
| 4                   | 101       | 648       | 749 (4.22%)| 0                 | 23          | 0           | 0           | 23     | 3,854    | 4,729       |
| 5                   | 101       | 651       | 752 (4.24%)| 0                 | 3           | 0           | 0           | 3      | 3,854    | 4,729       |
| 6                   | 101       | 660       | 761 (4.29%)| 0                 | 9           | 0           | 0           | 9      | 3,854    | 4,729       |
| Session2 (n = 98371)| 1         | 106       | 934       | 1040 (1.06%)      | 5           | 270         | 0           | 0      | 1,273    | 4,729       |
| 2                   | 107       | 1,123     | 1,230 (1.25%)| 1      | 189        | 0           | 0           | 190    | 4,729    | 4,729       |
| 3                   | 107       | 1,176     | 1,283 (1.3%)| 0                 | 53          | 0           | 0           | 53     | 4,729    | 4,729       |
| 4                   | 107       | 1,200     | 1,307 (1.33%)| 0                 | 24          | 0           | 0           | 24     | 4,729    | 4,729       |
| 5                   | 107       | 1,209     | 1,316 (1.34%)| 0                 | 9           | 0           | 0           | 9      | 4,729    | 4,729       |
| 6                   | 107       | 1,226     | 1,333 (1.36%)| 0                 | 17          | 0           | 0           | 17     | 4,729    | 4,729       |

We extracted the 15 more relevant terms 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 topic, 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 pathogen spreading over hospital networks. Some terms were considered highly relevant by the BART models (e.g., “Worker” in the sixth position out of more than 3800 terms considered), although in a simple linear model, their effect would hardly be significant (statistic: -1.21 s.e.); these are terms that are only relevant in conjunction with other terms but not on their own, highlighting the extra predictive power achieved through the use of advanced, non-linear machine learning.

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 to perform a more extensive search to find records that may have been missed during the first search session. The resulting query was as follows:

```plaintext
(((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 define the search domain better 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 used in the first session.

The new search produced 107,294 records (Table 1), of which 48,396 (58.6%) from the EMBASE, followed by 28,811 (34.9%) from Pubmed, 17,070 (20.7%) from SCOPUS, 12,956 (15.7%) from WOS, and 61 (0.074%) from IEEE; compared with the first session, the relative weight of EMBASE and Pubmed decreased, while the level of content specificity greatly increased, as it was for SCOPUS. After removal of duplicates, 82,579 unique records were obtained. The newly collected records were joined with those from the first session and duplicates were removed. We obtained 98,371 unique records, with just 1,963 shared records between searches, which equates to 2% of the total. The percentage of records shared by two or more sources 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); however, just labelling 275 of them (the suggested 250 plus 25 already labelled for the framework hyperparameter tuning) was sufficient to reduce this number to just 190 in the subsequent round. An evaluation of the convergence (Supplemental Material S2, Fig. 1) 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 becomes more and more sceptical and even asks to review some records labelled as positive in the initial training set generated during Session 1. This behaviour can rev spot classification errors and inconsistencies. Considering both sessions, 1,333 records were manually reviewed and 107 (8.03%) confirmed positive matches were found.

Again, the evaluation of the inclusion rate of the terms showed that the engine is quite capable of internalising the concepts behind the research topic. A subsample of the relevant terms used by the model in the second session is reported in Table 2 of Supplemental Material S2.
3.3 Hyperparameter selection

As described in the methods, hyperparameters were selected by evaluating sensibility and efficiency through a grid search on a validation set of 1,200 manually labelled records. The analysis suggested that the following parameter combination performed best: an initial training set of 250 categorised records with 10x oversampling of positive matches, ten models in the ensemble, no bootstrapping and an uncertainty zone defined by the 98% predictive interval. This combination of parameters was associated with a sensitivity of 98.8% (81 / 82 positive matches found) and an efficiency of 61.5% (462 / 1200 records evaluated). The detailed results of the hyperparameter tuning analysis are reported in Table 3 of Supplemental Material S2. Fig. 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 mainly impacted 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 boundary of the record PPDs as predictor (see the Methods for details). The surrogate model showed 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).

Fig. 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 the actual number of positive records found quite well. It is striking how many more records would have required manual evaluation to find the same number of positive matches without a classification algorithm, with some positive matches found 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 95.6% [95.3%, 95.7%]. In session 2 we observed a drop in expected sensitivity, especially in the lower credibility boundary (97.3% [72.8%, 100%]: as the number of records increases, even a small probability of being a positive match can, in the worst-case scenario, lead to a relevant number of predicted positive matches (147 in this case). To ensure no obvious positive matches were missed, we evaluated 100 non-reviewed records with the highest median predicted probability 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 realisation in the dataset [trunc. PrI] (see. methods). Finally, the logistic model’s median Bayesian $R^2$ [90% CrI] is reported. PrI: Predictive Intervals; CrI: Credibility Intervals.

| Indicator                        | Session 1 | Session 2 |
|----------------------------------|-----------|-----------|
| Total records                    | 17,755    | 98,371    |
| Reviewed records (% over total records) | 766 (4.31%) | 1,333 (1.36%) |
| Expected efficiency (over random) [trunc. 90% PrI] | 95.6% [95.3%, 95.7%] | 98.6% [98.1%, 98.6%] |
| Observed positive matches (% over total records) | 101 (0.57%) | 107 (0.11%) |
| Predicted positive matches [trunc. 90% PrI] | 101 [101, 108] | 110 [107, 147] |
| Expected sensitivity [trunc. 90% PrI] | 100% [93.5%, 100%] | 97.3% [72.8%, 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 Bayesian logistic model is shown as a shaded area delimited by the 95% quantile of the PrI and by the observed number of positive matches (light blue lines). A darker blue line represents the median of the PrI.

4 Discussion

We propose a new integrated framework to help researchers collect and screen scientific publications characterised by high performance and versatility. This framework joins 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 implements standard approaches and uses ad-hoc solutions to common SRA issues. By freely sharing the tool as an open-source R package and by following a modular design, we sought 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 search engine module used by the framework can automatically collect citation data from three well-known scientific databases (i.e., Pubmed, Web of Science, and the Institute of Electrical and Electronics Engineers) and process manually downloaded results from two more sources (SCOPUS, EMBASE). In comparison, most commercial or free SRH tools rely on internal databases (e.g., Mendeley https://www.mendeley.com/) sometimes focusing only on a particular topic (Visser 2010) or a single external data source (Thomas and Brunton 2007; Poulter et al. 2008; Soto, Przybyla, and Ananiadou 2019).

Mixing different databases is essential to obtain 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 were found in only one of the different data sources, and no positive record was present in all the sources (data not shown).

The framework online search algorithms are efficient enough to manage tens of thousands of search results, using various solutions to overcome the limitations of citation databases in terms of traffic and download
quotas. The results are then automatically organised, deduplicated and arranged by “simple query ordering” in a uniform corpus. The preliminary ordering increases the positivity rate in the initial training set (Wallace, Small, et al. 2010).

For the framework’s record screening module, we developed an active machine learning protocol (Settles 2009; 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 free text into input data for machine learning. We did not include classical n-grams (Schonlau and Guenther 2017); rather, we used network analysis to find non-consecutive, frequently associated terms, a generalisation of n-grams that relaxes the term adjacency assumption. This approach can also incorporate term connections across different parts of the records, e.g., terms having a different relevance when associated with a particular author. The same technique was used with different parameters to merge redundant terms, increasing estimation efficiency and reducing noise.

The automatic classification algorithm is based on Bayesian Additive Regression Trees (BART) (Chipman et al. 2010; Kapelner and Bleich 2013). Like other boosted trees algorithms (Hastie, Tibshirani, and Friedman 2009), the BART method can explore complex non-linearities, perform variable selection, manage missing data while maintaining high predictive power.

However, the Bayesian foundation of the method provides further benefits: lower sensitivity to the choice of hyperparameters, natural regularisation through priors, and, most importantly, predictive distributions as output instead 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 overly deep trees, long tree sequences, and extreme predicted probabilities, thus reducing 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 confirmed 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 and Casella 2004), generating a shift in the probability mass towards the distribution centre and stabilising it (i.e., reducing variance without impacting bias). On the other hand, simply imposing more robust uninformative priors against extreme predictions would have reduced variance but also shifted the distribution towards a non-decision zone, increasing bias (Hansen et al. 2000).

Since the number of model replications has a significant impact on computation times, we decided to use ten replicas, the lower value after which performance stabilised, as resulted from the evaluation of the hyperparameters (Supplemental Material S2, Fig. 2).

We also investigated whether bootstrapping between replications (Breiman 1996) would improve performance; however, contrary to theory (Díez-Pastor et al. 2015), it appeared to be slightly detrimental in our case (Supplemental Material S2, Fig. 2) compared to simple ensembling.

A low proportion of relevant matches (class imbalance) is typical for literature reviews (Sampson, Tetzlaff, and Urquhart 2011; Wallace, Trikalinos, et al. 2010; O’Mara-Eves et al. 2015), and a strong imbalance between positive and negative records can affect sensitivity (Khoshgoftaar, Van Hulse, and Napolitano 2010; Chawla, Japkowicz, and Kotcz 2004).

To overcome this problem, we oversampled (Batista, Prati, and Monard 2004) the positive records ten times before model fitting. The hyperparameter analysis showed that the oversampling rate, together with model ensembling, was the parameter with the most significant impact on performance.

A known risk with positive oversampling is the misclassification of negative records (Ramezankhani et al. 2016). However, since all predicted positives in our approach are reviewed manually, we are always guaranteed to achieve 100% specificity/positive predictive value: the only price for the increased sensitivity due to oversampling is a larger number of records to be reviewed.

An alternative to oversampling would be to apply different weights and/or costs to the classes (Abd Elrahman and Abraham 2013; Díez-Pastor et al. 2015), but the BART implementation we used did not have this feature; furthermore, using simple oversampling allows for a broader compatibility with different modelling engines (Galar et al. 2011; Roshan and Asadi 2020).
Finally, sorting the records by query term frequency (simple query ordering) produces a much higher rate of relevant records in the initial training set (17.2%) compared to the overall data (0.11%), which boosts the sensitivity of the model.

One of the key innovations we have introduced is the concept of “uncertainty zone”, the implementation of which is possible thanks to the Bayesian foundation of the classification model. This construct guides the selection of records to be manually reviewed and gets dynamically updated and reduced after each CR iteration, as more uncertain predictions are evaluated (Supplemental Material S2 Fig. 1).

The use of a dynamic uncertainty zone overcomes the usual requirement of dataset-specific hard thresholds in active machine learning and allows to review multiple items at once between iterations (Laws and Schütze 2008; Miwa et al. 2014; Zhu et al. 2010). The hyperparameters required by our algorithm are general and non-task-specific, like the PPD intervals underlying the uncertainty zone and the maximum number of iterations without positive matches after which a session is concluded; the evaluation of the classification model hyperparameters shows that the algorithm is robust against variations in these parameters, and we expect the default values to perform well on most datasets.

Since researchers are asked to review both records predicted as surely relevant 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 assessed performance as the ability of the screening procedure (automatic classification plus manual review) to find the largest number of relevant records while requiring manual reviewing for as few of them as possible (i.e., sensitivity × efficiency).

We avoided the classical out-of-sample approaches such as 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 the same on average in every possible 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 active learning algorithm, resulting in a lower positivity rate in the unlabelled records (Fig. 2). Moreover, 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.

To estimate overall sensitivity, we instead applied simple Bayesian regression (surrogate model) to the manually reviewed data to abstract the classification model predictions and generate 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 only the lower 98% PrI boundary of the PPDs as predictor, indicating predictive consistency in the classification model. The posterior predictive distribution of the surrogate model could be used to explore worse case scenarios in terms of sensitivity.

Our framework achieves very high sensitivity by screening only a very small fraction of all records, bringing a meaningful 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 as only a few new positive matches were found; however, given the large number of records, uncertainty about sensitivity increased, as expected.

Both results are above the usual performance in this field (O’Mara-Eves et al. 2015) and are in line with the average sensitivity of 92% estimated after human-only screening (Edwards et al. 2002). In one interesting case, the model detected a human-caused misclassification error, demonstrating its robustness and value as a second screener, a role already suggested for SRA tools in previous studies (Frunza, Inkpen, and Matwin 2010; Bekhuis and Demner-Fushman 2012, 2010). Although “simple query ordering” concentrated most relevant matches in the first 20-25 thousand records, without the tool support, the remaining relevant records would have been missed without manually screening almost the entire dataset.

The model required ~5-20 minutes per iteration to perform the 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, 2014 laptop; 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). This is a considerable time saving compared to the several months
typically 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) was larger than what is typical of most SRA studies (O’Mara-Eves et al. 2015; Olorisade et al. 2016), highlighting the scalability of the tool in real-world scenarios.

The last module of our framework is an algorithm for data-driven search query generation. Generating an efficient and effective search query is a complex task (Lefèvre et al. 2011; Hammerstrøm et al. 2010); 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 be reviewed. Our solution combines 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 new search query to help find records missed in the first search session. The generated query did indeed retrieve a few more relevant records not found in session 1 but at the cost of significantly increasing the number of documents.

An interesting aspect of this feature 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, served also as a means for machine learning explainability (Bhatt et al. 2020; Burkart and Huber 2021), useful for understanding and detecting biases 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 dataset 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 when applied to an unbounded search domain and may return an unmanageable amount of irrelevant 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 mentioned early, our framework builds on modularity. We have designed so that each module can become fully independent in future iterations; it will be possible for users to add custom features such as citation search and parsing for other scientific databases, alternative text processing algorithms or machine learning modules. We consider such interoperability to be extremely relevant: the main strength of our tool lies in the composition of many independent solutions, such as the idea of Bayesian active machine learning and the exploit of the derived uncertainty in defining the records needing human review.

Each component could benefit considerably from the recent improvements in text mining and machine learning.

For example, the text processing approach based on the “boolean bag-of-words” paradigm is quite simple and could be improved by more nuanced text representations. It might be considered whether feature transformations such as TF-IDF (Baeza-Yates, Ribeiro-Neto, et al. 1999; Ananiadou and McNaught 2006) could be advantageous, although we hypothesise that tree-based classification algorithms like BART are robust enough not to require such operations. Instead, it might be worth exploring the application of word embedding: this technique transforms terms into semantic vectors derived from the surrounding text (Turian, Ratinov, and Bengio 2010; Bollegala, Mahara, and Kawarabayashi 2015; Minaee et al. 2021) and could be used to reduce noise by merging different terms that are semantically similar or enhance signal by distinguishing identical terms with different meaning given the context. Another option would be to employ unsupervised learning models like Latent Dirichlet Analysis and Latent Semantic Analysis, (Pavlinek and Podgorlec 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 that expand the feature space.

Our classification algorithm is applicable with any Bayesian supervised machine learning method that provides full PPDs; therefore, alternative classification models, such as Gaussian Processes, known for their flexibility (Jayashree and Srijith 2020; S.-H. Chen et al. 2015), could be evaluated. It would be even more interesting to test advanced learning algorithms that go beyond the bag-of-words approach and take into consideration higher-level features in the text such as 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), provided that a Bayesian implementation of such algorithms is available (for example C. Chen, Lin, and Terejami (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 evolving rapidly, and we anticipate an increasing use of such technologies to address the accelerating pace of scientific production. We believe it is encouraging that a wide variety of tools are being made available to let researchers and policymakers find the approach that best fits their needs.

We contribute to this field with an innovative framework that provides excellent performance and easy integration with existing systematic review pipelines. The value of this work lies not only in the framework itself, which we make available as open-source software, but also in the set of methodologies we developed to solve various SRA issues and which can also be used to improve existing solutions.
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Study protocol

2022-02-22

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Research Protocol

This document will guide the reader along the steps to utilise the framework and reproduce our results.

To use the framework, clone it from https://github.com/AD-Papers-Material/BART_SystReviewClassifier and run the R project in the repository.

The first step is loading the code base using:

```
source("R/Setup.R") # Load the framework
```

Acquisition of the citation data

Once the framework is loaded, the user defines an initial search query which needs to be as specific as possible while still generating a sufficient number of positive matches:

```
# Initial query to be built on domain knowledge. It accepts OR, AND, NOT boolean
# operators and round brackets to group terms.
query <- '(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))'
```

# Year filter. The framework converts it to the API-specific format seamlessly.
# common logical comparators can be used, i.e. <, <=, >, >=, while dashes
# denotes inclusive date intervals. A single year restricts results to one year
# period.
year_filter <- "2010-2020"

The query is passed to the `perform_search_session()` function, together with an unique identifier for the search session and search query. A session identifies a homogeneous group of complementary or alternative queries (e.g. sometimes it is easier to create two different simpler queries than a complex and long one that returns the same results).

In our implementation, we define as sessions the group of actions that includes the utilisation of a search query and the subsequent iterations of manual review and automatic classification of its results.

`perform_search_session()` will use common scientific research database APIs (at the moment Pubmed/Medline, Web of Science and IEEE) to acquire records related to the query. The records will be stored as CSV files in folders with the following structure: Records/session_id/query_id, created automatically. The function also creates or updates a journal (a CSV or Microsoft Excel file) with all the information about sessions, queries and search results.

Some services (IEEE, WOS) requires an API key to access the data; the API key is not mandatory for Pubmed but can help avoid quota limitations in case of a large number of results. IEEE can also be searched without API: the framework will utilise a web scraping approach if the API key is missing, but the success of this method is not guaranteed. See Supplemental Material S3 for detailed instruction on preliminary steps and tips for each supported search engine. These keys can be stored in the secrets.R file in the working directory, with the following example code (the “baysren” prefix identifies the package specific options):

```
options(baysren.ieee_api_key = "your-ieee-api-key")
options(baysren.ncbi_api_key = "your-ncbi-api-key")
options(baysren.wos_api_key = "your-wos-api-key")
```

This file will be automatically parsed from the framework.

If previously downloaded results are already available, users can create the Record folder manually and put the data there (the file names need to contain the source name as written in the sources argument of
perform_search_session(). The function will acquire and parse them into a standard format, even if split into multiple files due to download limits (e.g. Pubmed1.nbib, Pubmed2.nib, etc.). In addition to Pubmed, WOS and IEEE, this method permits to import also EMBASE and SCOPUS records, for which an API search was not yet implemented.

Using the actions argument, users can choose whether to perform an API search, parse already downloaded results, or both.

Note that for Pubmed, the API search may return slightly fewer records than a manual one due to a different query expansion algorithm (this divergence should be solved in April 2022) between the two services, therefore is advisable to perform also a manual search and put the resulting .nbib files in the Session/Query folder. perform_search_session() will acquire them seamlessly.

If a user wants to perform the API searches and results parsing manually for each source, the framework exposes the individual functions to search for records using the APIs and parsing bibliography files.

```r
# This call will perform an API search on Pubmed, WOS and IEEE services and/or
# (based on the `actions` argument) parse already downloaded data (in our case,
# additional Pubmed results and Embase and Scopus results) manually added to the
# Session1/Query1 folder.

journal <- perform_search_session(
  query = query, year_query = year_filter,
  session_name = "Session1", query_name = "Query1",
  records_folder = "Records",
  journal = "Session_journal.csv"
)
```

Once the records are stored, they must be read and merged into an annotation file, where the initial manual review will be performed. A series of functions are available to prepare the data for manual evaluation:

```r
# Extract the file paths of records. Arguments can be used to filter by session
# query, source. Only parsed files will be returned, not the raw ones downloaded
# manually.

Annotation_data <- extract_source_file_paths(journal) %>%
  # Read record files. Will parse them if raw data is downloaded manually (not
  # necessary in this case). Return a list of records, one per file.
  read_bib_files() %>%
  # Join a list of records into one dataframe, solving duplicated records
  join_records() %>%
  # Order records by the relative frequency of the query terms in the title +
  # abstract text. Increases the chance of encountering relevant records at the
  # beginning of the manual classification.
  order_by_query_match(query) %>%
  # Add fields for the manual classification
  mutate(
    Rev_manual = NA, # Where the first manual classification will be made
    Rev_prediction = NA, # Column for the evaluation of the predicted classes
    .before = DOI
  )
```

All these steps are joined into the `create_annotation_file()` function. This function can take as input either the path to record files, or the parsed records (one data frame or a list of data frames) or a group of folders where to look for files into. In addition, it also allows adding the new records to a previous annotation file (arg `prev_records`). This is useful when performing consecutive search sessions with different queries; previous manual classifications can be imported using `prev_classification`.

If the original query is passed to the `reorder_query` argument, the records are reordered according to the
frequency of query terms in the title and abstract (simple query ordering); this approach lets increase the rate of relevant matches at the beginning of the dataset.

```r
# This function extracts the appropriate file paths from a session journal. By default it passes all stored files, otherwise it can be filtered by session, query, and source (i.e. Pubmed, WOS, IEEE)
record_files <- extract_source_file_paths(journal)

# create_annotation_file() accept a great variety of inputs:

# either record file paths
input <- record_files

# or specific record file folders
input <- file.path("Records", "Session1", "Query1")

# or parent folders, since it searches for files recursively
input <- "Records"

# or the already parsed files
input <- read_bib_files(record_files)

# We can then call create_annotation_file() with one of the above input
Annotation_data <- create_annotation_file(input, reorder_query = query)
```

Once created, the annotations need to be stored in an Annotation Session folder, which will contain all information necessary for the automatic classification. This operation is streamlined by the create_session() function, which also helps manage duplicated sessions.

```r
# Create the first session
create_session(Annotation_data, session_name = "Session1")
```

The file annotation file is called Records_{date}, situated in the folder Sessions/{session_name}; date here represents the session creation timestamp, and session_name the name passed by the user.

Notice that the framework is “pipe” friendly, and the precedent steps can be all performed in one call:

```r
# First, create the Records/Session1/Query1 folder and put the manually downloaded records there if there are any. Otherwise, the folders will be created automatically during the API search
Records <- perform_search_session(
  query = query, year_query = year_filter,
  session_name = "Session1", query_name = "Query1",
  journal = "Session_journal.csv"
) %>%
  extract_source_file_paths() %>%
  create_annotation_file(reorder_query = query) %>%
  create_session(session_name = "Session1")
```
Manual classification and prediction

Now the annotation file is ready for the manual classification of records. The annotation file has a column called `Rev_manual`, where records need to be classified as “positive” (y) or “negative” (n) matches by the user. We suggest manually labelling an initial training set of 250 records, which should be sufficient if a minimum ~20% positivity rate is present (see the section about model hyperparameter optimisation). The function `plot_classification_trends()` can help visualise the trend of the positives/negatives as the number of classified records increases.

Once the initial training set is prepared, the function `enrich_annotation_file()` will use a Bayesian Additive Regression Trees (BART) machine learning model to learn the labelling pattern given the record data and predict the positive match probability for the unlabelled records. `enrich_annotation_file()` comprises two steps. The first step is the creation of a Document Term Matrix (DTM):

- The framework extracts features from text data (i.e., title and abstract) of the records, authors, keywords and MESH terms (see Methods for more info). Before features extraction, text data terms are lemmatised, stop-words are removed, and only nouns, adjectives, verbs and untagged terms are retained. Authors, keywords, and MESH are kept unmodified. Co-occurrent features (non-consecutive ngrams, nc-grams) are identified and redundant features (i.e. terms always appearing together) are joined to decrease the feature space noise.
  - For each term, a 0/1 label specifies if it is present in each record.
  - Positive records are oversampled 10x to improve sensitivity;
  - An extra feature is added for each record field (title, abstract, authors, keywords, MESH terms), reporting the number of terms in each.

The second step is the modelling of the labelling pattern and the predictions of the relevancy/positivity probability:

- The function models the labelling pattern using the manually labelled records (**learning phase**);
- After the model is created, it assigns a posterior predictive distribution (PPD) of the probability of positivity to each record (**prediction phase**). If the number of records is large, the prediction step is broken into chunks of 5000 records, to avoid memory saturation; the users can set up this parameter according to their machine capabilities;
- This process is repeated 10 times, and the predictive distributions are averaged to induce shrinkage of the estimates (i.e., decrease the chance of outliers), creating a prediction ensemble;
- The resulting distributions are summarised by the 98% interval [98% PrI] and the median; the [98% PrI] identifies the uncertainty zone;
- The unlabelled records will receive a positive/negative predicted label; the label is positive if its lower [98% PrI] bound is higher than the uncertainty zone upper bound and negative if the upper [98% PrI] bound is lower than the uncertainty zone lower bound. All other records are labelled as uncertain (unk);
- If a predicted label does not match an existing one, the record will be labelled as check.

The user can define parameters like the positives’ oversampling rate, the number of models to average in the ensemble, whether to apply bootstrap resampling before each repetition, the prediction chunk size, and the PPD quantiles used to build the uncertainty zone. To use the function, the only input needed is the session name; the function will automatically identify the relevant file to make predictions on.

```r
New_annotations <- enrich_annotation_file("Session1")
```

This function will produce a new file which is stored in the Annotations folder of the session; the file will contain a column called `Rev_prediction_new` where the user will find records that require manual review marked by “*”. These are records with uncertain labelling (“unk”), but also positive predictions and “y”/“n” predictions that contradict previous manually input labels (labelled as “check”). This approach increases the accuracy of the predictions since the machine learning system is set up to favour sensitivity at the expense of specificity. If the function finds prelabelled records (`Rev_previous` column) added when the Annotation file
was created or passed as a further input to `enrich_annotation_file()` (prev_classification argument), they will be used to label the records marked in the Rev_prediction_new column.

The function adds a number of diagnostic pages to the annotation file:

- A results summary page reporting the number of records in need of manual review (which includes “unk”, “check”, and unreviewed “y” records), the changes, and the final distribution of the labels. It also shows other information like the number of features in the DTM, the path to the parent file from which the annotation file was created, the replication number (see later). The content of this summary is also saved as a CSV file in the Results folder.
- A variable importance page that lists the used terms in order of the fraction of BART trees in which they were used. If an ensemble of models is used, a more robust score is computed, taking the ratio of the mean of the term tree fraction among models and its standard deviation.
- The arguments passed to the function if different from the default, useful for reproducibility.
- A classification performance summary. The procedure may take some time and requires `cmdstan` installed on the machine. Therefore the `compute_performance` argument is set to `FALSE` by default.

Finally, the PPD ensemble produced by the Bayesian engine is stored on the disk. This feature can be turned off to save disk space, but at least the samples of the last annotation are necessary to build a new search query (see next section).

The process of predicting labels and manually reviewing them is what defines a “classification-review (CR) iteration”.

Once the manual review of the generated annotation file is completed, the user can **rerun exactly the same function**. The last reviewed annotation file will be automatically picked up and used for a new CR iteration. If the number of records requiring manual review is excessively high (e.g. > 250), the function needs to be called with the stop_on_unreviewed argument set to `FALSE` to allow the classification to proceed even in the presence of unreviewed records in the Rev_prediction_new column.

If there are no positive results (after manual review) in a CR iteration, the framework records the next iteration as a replication. After four replications with no new positive matches, the function will prevent another iteration and send a warning message, indicating that the session is complete. The user can change the replication limit; furthermore, it is possible to prevent the function from starting if either a specified number (or a fraction) of the total records has been manually reviewed or if a certain number of positive matches have been found. These three parameters are passed to the limits argument, which has the following structure and defaults:

```r
list(
  stop_after = 4,
  pos_target = NULL,
  labeling_limit = NULL
)
```

The result files in the Results folder are generated after the Classification step but before the Review one, and therefore show preliminary results with still unreviewed records. The function `consolidate_results(session_name)` will update the results files to show the results after the manual review. The original result data is stored anyway in the respective annotations files.

**New search query generation**

It is not uncommon for the first query used in a systematic review not to be the most effective one, especially in terms of sensitivity. We tried to address the problem by creating an automatic query generation system. The solution is experimental and greatly favours sensitivity at the expense of specificity. The general query generation mechanism is the following:

- We fit a partition tree 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 \textit{AND/NOT} “conditions” made of terms/authors/keywords/MESH tokens, which together identify a group of records.

- For each rule, negative conditions (i.e., \textit{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 \textit{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.

The steps for generating a query are the following:

```
# The last annotation file and posterior sample matrix in the session are found; # the most predictive terms (based on the importance score) are chosen and # simple prediction trees are used to map the terms to a random subset of the # posterior probabilities (extracted from the posterior sample matrix saved # during the last CR iteration). The score threshold and the number of trees can # be chosen by the user; less stringent values can significantly increase # computation time. By default results will be saved in a file called # rule_data.rds in the session folder.

candidate_queries <- extract_rules("Session1")
```
Once the manual selection is performed the selected rules can be further simplified in order to remove redundant rules and terms. The aim is to provide the shortest rule without loss of sensitivity.

```r
simplified_rules <- file.path("Sessions", "Session1", "Selected_rules.xlsx") %>%
    import_data() %>%
    simplify_ruleset(target_vec = Target, target_data = SpecificDTM) %>%
pull(rule)
```

# Finally, the selected rules need to be joined in a query that scientific search engines can understand

```r
ew_query <- rules_to_query(simplified_rules)
```

One last thing to keep in mind is that the query generation algorithm is only aware of the literature collected inside the annotation file. So it will only be able to pick term combinations that identify positive matches inside the already specific set of documents while not necessarily being discriminant from the rest of the global literature.

Once the final query is generated, it is advisable to add a constraining subquery with an AND logic to restrict the results to the scope of the topic of interest.

In our case, we added “AND ((antimicrobial resistance) OR (healthcare infection))” since, being the global topic of our research, those terms were understandably not selected by the query generation algorithm as discriminant.

The final query was the following:

```r
query <- "(((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))"
```

Another advantage of this technique is an at-glance view of relevant concepts for the topic of interest.

**New search session**

Once the new query was generated, a second search session can be created, using the strategy described in the protocol. Due to the lower specificity of the second query, particular care may be needed in managing the far larger amount of records: multiple record files may need to be downloaded from Pubmed, EMBASE, and SCOPUS since they have limits on the number of records one can download at once; also, the machine learning classification processing time increases proportionally with the number of records to label.

It is advisable to consider removing the less specific rules iteratively from the new query until a manageable number of records is obtained, considering that the probability of finding new positive matches not already found by more specific queries drops exponentially.

Once the new records are collected are manually collected for databases without an API access, a new search
can be performed. The following code describe how to create a new session. Keep in mind to give the session a new name and to import the annotated records from the last session when generating the new annotation file.

```r
# Repeat the search with the new query, giving a different Session name not to # override previous records
journal <- perform_search_session(
  query = query, year_query = year_filter,
  session_name = "Session2", query_name = "Query1",
  records_folder = "Records",
  journal = "Session_journal.csv"
)

# Get the path to the annotated data in the previous session
previousAnnotations <- get_session_files("Session1")$Annotations %>% last()

# Create the new annotation file passing the folder with the new records
Annotation_data <- create_annotation_file(
  file.path("Records", "Session2"),
  reorder_query = query,
  prev_records = previousAnnotations
)

# Create the new annotation session
create_session(Annotation_data, session_name = "Session2")
```

Once the new annotation session data is created, `enrich_annotation_file('Session2')` can be used to perform the CR iterations. It may be possible that during the first CR iteration, the number of records requiring manual review is very large: we suggest evaluating still not more than 250, setting the `stop_on_unreviewed` argument to `FALSE` to proceed in the automatic classification even if some uncertain records from the precedent iteration are still unreviewed.

### Result analysis and reporting

Several tools are available to evaluate the classification performance. As said before, a result file is stored in the session folder for each CR iteration with information on the classification status; remember to run `consolidate_results(session_name)` if you want these results to account for the manual review of the predicted labels.

- `compute_changes()` is a simple function that take an annotation data frame and computes the changes from the last annotation iteration.
- The primary function for result analysis is `estimate_performance()`. The function uses a Bayesian model to estimate how the probability of a positive match drops as records’ lower boundary of the posterior predictive interval decreases. This model computes the cumulative distribution of possibly missed positive records, which permits extraction of the engine’s theoretical sensitivity and efficiency to lower 90% credibility boundaries (i.e., the indicators are better than these values with 95% probability). These boundaries depict worse case scenarios since it is impossible to know the actual number of missed positive records. The function returns the Bayesian $R^2$ of the model (values below 85-90% would invalidate the model analysis), the posterior sensitivity, the observed and theoretical number positives, the expected efficiency given the theoretical number of positives (that is 1 - the number of reviewed records above the number of records to review find the predicted number of positives according to a negative hypergeometrical distribution). Also, the function plots the cumulative distribution of the observed and theoretical number of positive records.
- `extract_var_imp()` extract the most relevant terms used by the BART model and summarises their relevancy for the machine learning model and their statistical relevancy in a simple generalised linear model.

A discrepancy between these two scores indicates a non-linear effect/interaction between terms.
Finally, several functions are available (check R/Reporting.R) to summarise and format the results. For example, `summarise_by_source()` and `summarise_sources_by_session()`, respectively for one session or a group of sessions, report the scientific databases the records are collected from, while `get_source_distribution()` describes the distribution of the number of common sources per record. `summarise_annotations()` and `summarise_annotations_by_session()` describe the results of the CR iterations, again for one or multiple sessions, reporting the number of positive and negative matches, the number of terms (features) used by the machine learning model, and how the labels change between iterations. `format_performance()` and `format_var_imp()` format the output of the similarly named analyses. `plot_predictive_densities()` plots how the mixture of posterior predictive distributions of records, divided into positive, negative and to-review records, change between iterations.

Model parameters’ optimisation

As with many complex machine learning models, the choice of hyperparameters can significantly influence the framework performance. For a systematic review helper algorithm, we define the performance as the ability to find as many positive matches by manually labelling/reviewing as few records as possible:

\[
\text{Sensitivity} \times \text{Efficiency} = \frac{\text{Pos. found}}{\text{Tot. pos.}} \times \left(1 - \frac{\text{Records reviewed}}{\text{Tot. records}}\right)
\]

We utilise a “grid search” algorithm to find the best set of hyperparameters. We tested these tools on our dataset, manually labelling the first 1200 records, ordered by simple query match. The first step is to create a folder to store the file required for the search, for example, `Grid Search`. Then a fully reviewed record file in the format generated by `create_annotation_file()` is necessary; this file can be put in the `Grid Search`, but it is not mandatory.

Once the folder and the file are ready, run:

```r
Grid_search <- perform_grid_evaluation(
  records,
  sessions_folder = "Grid_Search",
  prev_classification = records,
  ## Model parameters (can be changed by users)
  resample = c(FALSE, TRUE),
  n_init = c(50, 100, 250, 500),
  n_models = c(1, 5, 10, 20, 40, 60),
  pos_mult = c(1, 10, 20),
  pred_quants = list(
    c(.1, .5, .9),
    c(.05, .5, .95),
    c(.01, .5, .99)
  )
)
```

with `records` being the fully annotated data frame or a file path. `prev_classification` can be used to import a different classification file with prelabeled records.

The hyperparameters are the following:

- `resample`: whether to use bootstrapping to increase variability between the ensemble models;
- `n_init`: number of initially labelled records to train the machine learning model on;
- `n_models`: the number of models to fit and then average to decrease the variance due to random sampling in the MCMC algorithm. It reduces the likelihood of extreme predictions;
- `pos_mult`: the oversampling rate of positive records. Useful to improve sensitivity;
• pred_quants: the distribution quantiles of each PPD, used to define the uncertainty zone and identify records to review.

Once the grid search is performed, the function `analyse_grid_search()` can be used to analyse its results. This function uses a partition tree analysis to identify hyperparameter sets (performance clusters) with different mean performance scores. As said before, the default score is Sensitivity x Efficiency; alternatives are the Positive Rate (i.e., positive records found over total records) or Positive Rate x Sensitivity. The function produces a data frame with the score and the cluster of reference of each combination of parameters, the best hyperparameter set (i.e., the most efficient set among the most sensitive in the best cluster) and the best set for each cluster. Also, the function creates a plot to visualise each hyperparameter’s impact on performance.

Once chosen, the hyperparameters can be passed to `enrich_annotation_file()`. Mind that the grid search process may take many days, even on powerful computers, so we suggest sticking to the default set of parameters when using `enrich_annotation_file()`. 
S2. Additional figures and table

2022-02-21

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Posterior predictive distributions

Figure 1 show the mixture of the PPD of the probability of a positive match, grouped by label (only records manually labelled or reviewed are considered). The posterior samples for each record were extracted and joined into a global distribution; on this distribution, density was computed on the logit scale and then logistic transformed for displaying on a $[0 - 1]$ scale. The purple ridges show the distribution of the still unlabelled records.

For each iteration, the thresholds which define the “uncertainty zone”, i.e, the lower and the upper range of 98% PrI for the positive and negative records respectively, are shown. Records whose 98% PrI intersects the uncertainty zone requires manual review.

Notice how the positive and negative record densities tend to increasingly overlap at each iteration; meanwhile the distribution of the records to be reviewed shrinks and shifts towards the negative side, as positive records get found and labeled.

In Session 2, as the number of negative records reviewed increased, also already positively labelled records re-entered the uncertainty zone; this is due to the baseline positivity rate decreasing as the number of negatives in the training data increased, forcing to review dubious records that may have been mislabelled.

List of terms relevant for prediction

In table 1 and 2 are listed the 50 more relevant terms used by the BART algorithm to discriminate between positive and negative records, for Session 1 and 2 (see Methods). Term importance (“Inclusion rate” in the tables) is defined as the ensemble average inclusion rate in posterior trees over 10,000 total term inclusions, while the Inclusion Stability (IS) is the ratio of the average inclusion rate over its standard deviation among the ensemble models. The symbol “|” in the terms indicate redundant terms while “&” indicate nc-grams. The component in which the term was used is reported in the leftmost column.

For each term, we added its linear association with a positive label estimated through a Poisson regression, reporting it as Relative Risk (RR) and its statistical significance (Statistic) measured as number of standard errors, s.e., of the terms. A strong BART score with a Statistic close to zero identify terms whose effect is highly non-linear (e.g., they are relevant only in the presence of other terms).

Hyperparameters grid search

Table 3 shows the best hyperparameter set and their performance for each cluster while figure 2 displays the conditional impact on performance of each hyperparameter.

The most influential parameters were the number of models in the ensemble, the oversampling rate and the initial training set size. The use of bootstrap resampling seemed detrimental.
Figure 1. Mixture predictive distribution of the probability of a positive match, grouped by labelling status.
Figure 2. Score clusters and impact of single hyperparameters on engine performance. The performance is measured as Sensitivity x Efficiency. Each cluster is color coded. Mods: number of models in the ensemble; Quant: PrI quantiles for the uncertainty zone; Resamp: bootstrap resampling between models; Init: size of initial training set; Multi: oversampling rate of positive records.
| Component | Term | Inclusion rate | IS  | RR  | Statistic |
|-----------|------|----------------|-----|-----|-----------|
| Keyword   | Patient Transport | 61.2 | 3.77 | 99.1 | 21.3 |
| Abstract  | Transfer | 57 | 3.93 | 22.5 | 15.4 |
| Title     | Network | 56.5 | 2.91 | 18.0 | 14.2 |
| Abstract  | Network & Patient | 54.2 | 4.66 | 26.3 | 15.2 |
| Author    | Donker T | 53.5 | 4.56 | 159.0 | 16.5 |
| Abstract  | Worker | 50 | 3.33 | 0.421 | -1.21 |
| Keyword   | Hospitals | 49.8 | 4.31 | 27.8 | 16.5 |
| Abstract  | Movement | 47.8 | 2.7 | 27.2 | 15.0 |
| Title     | Spread | 46.6 | 2.25 | 16.2 | 12.1 |
| Abstract  | Facility | 45 | 2.22 | 19.6 | 14.8 |
| Keyword   | Orange County | 44.3 | 3.19 | 199.0 | 17.2 |
| Abstract  | Patient | 42.6 | 3.7 | 27.6 | 7.23 |
| Abstract  | Perform | 41.9 | 2.38 | 0.342 | -2.55 |
| Title     | Hospital | 39 | 1.95 | 12.5 | 12.5 |
| Abstract  | Regional | 38.9 | 3.08 | 21.7 | 14.9 |
| Abstract  | Agent | 38.1 | 2.74 | 4.36 | 6.28 |
| Abstract  | California | 37.3 | 2.6 | 38.0 | 12.6 |
| Title     | Transfer | 36.6 | 3.54 | 27.0 | 11.8 |
| Keyword   | Patient Transfer | 36.6 | 2.16 | 164.0 | 2.0 |
| Abstract  | Finding | 33.2 | 1.78 | 0.372 | -2.35 |
| Title     | Outbreak | 32.9 | 2.91 | 3.4 | 3.51 |
| Abstract  | Collect | 32.4 | 1.82 | 0.408 | -1.95 |
| Title     | Regional | 32.3 | 2.68 | 44.2 | 14.2 |
| Abstract  | Network | 31.6 | 3.09 | 12.8 | 11.7 |
| Abstract  | Resistant | 31.6 | 2.55 | 11.0 | 11.2 |
| Abstract  | Outcome | 31.2 | 1.93 | 0.178 | -2.95 |
| Abstract  | Discharge | 31.1 | 3.79 | 9.99 | 9.02 |
| Abstract  | 2014 | 30.3 | 1.84 | 0.588 | -1.04 |
| Abstract  | Practice | 29.5 | 2.47 | 0.508 | -1.33 |
| Abstract  | Culture | 28.9 | 2.14 | 0.378 | -1.66 |
| Abstract  | Positive | 28.8 | 1.96 | 0.346 | -2.52 |
| Abstract  | Gene | 28.3 | 1.29 | 0.000000024 | -0.0415 |
| Abstract  | Disease | 28 | 2.42 | 0.365 | -3.7 |
| Keyword   | Enterococci | 27.5 | 1.92 | 25.2 | 6.33 |
| Abstract  | Month | 27.3 | 1.86 | 0.288 | -2.44 |
| Abstract  | Healthcare & Facility | 26.2 | 1.74 | 34.9 | 17.0 |
| Abstract  | Prevalence | 26.2 | 1.71 | 4.08 | 6.69 |
| Abstract  | Effort | 26.1 | 1.22 | 6.64 | 8.57 |
| Abstract  | Length | 25.3 | 1.44 | 6.1 | 6.45 |
| Keyword   | System | 25 | 1.43 | 11.9 | 3.47 |
| Abstract  | Laboratory | 24.4 | 1.38 | 0.442 | -1.39 |
| Keyword   | Resistant Staphylococcus Aureus | 24.4 | 1.59 | 22.8 | 11.2 |
| Abstract  | Clinical | 23.6 | 2.1 | 0.421 | -2.9 |
| Abstract  | Dataset | 22.5 | 1.47 | 8.75 | 6.5 |
| Abstract  | Development | 22.2 | 1.22 | 0.148 | -2.68 |
| Abstract  | Hand | 22 | 1.97 | 0.822 | -0.335 |
| Keyword   | Pathogen Transmission | 21.8 | 3.26 | 67.9 | 7.2 |
| Keyword   | Cross Infection & Humans & Transmission | 21.7 | 1.4 | 31.1 | 15.5 |
| Abstract  | Flow | 21.4 | 1.14 | 4.07 | 4.56 |
Table 2. Term importance at the end of Session 2.

| Component | Term | Inclusion rate | IS | RR | Statistic |
|-----------|------|----------------|----|----|-----------|
| Keyword   | Patient Transport | 61.2 | 3.77 | 99.1 | 21.3 |
| Abstract  | Transfer | 57 | 3.93 | 22.5 | 15.4 |
| Title     | Network | 56.5 | 2.91 | 18 | 14.2 |
| Abstract  | Network & Patient | 54.2 | 4.66 | 26.3 | 15.2 |
| Author    | Donker T | 53.5 | 4.56 | 159 | 16.5 |
| Abstract  | Worker | 50 | 3.33 | 0.421 | -1.21 |
| Keyword   | Hospitals | 49.8 | 4.31 | 27.8 | 16.5 |
| Abstract  | Movement | 47.8 | 2.7 | 27.2 | 15 |
| Title     | Spread | 46.6 | 2.25 | 16.2 | 12.1 |
| Abstract  | Facility | 45 | 2.22 | 19.6 | 14.8 |
| Keyword   | Orange County | 44.3 | 3.19 | 199 | 17.2 |
| Abstract  | Conduct | 42.6 | 3.7 | 0.221 | -2.57 |
| Abstract  | Patient | 42 | 3.61 | 27.6 | 7.23 |
| Abstract  | Perform | 41.9 | 2.38 | 0.342 | -2.55 |
| Title     | Hospital | 39 | 1.95 | 12.5 | 12.5 |
| Abstract  | Regional | 38.9 | 3.08 | 21.7 | 14.9 |
| Abstract  | Agent | 38.1 | 2.74 | 4.36 | 6.28 |
| Abstract  | California | 37.3 | 2.6 | 38 | 12.6 |
| Title     | Transfer | 36.6 | 3.54 | 27 | 11.8 |
| Keyword   | Patient Transfer | 36.6 | 2.16 | 164 | 2 |
| Abstract  | Finding | 33.2 | 1.78 | 0.372 | -2.35 |
| Title     | Outbreak | 32.9 | 2.91 | 3.4 | 3.51 |
| Abstract  | Collect | 32.4 | 1.82 | 0.408 | -1.95 |
| Title     | Regional | 32.3 | 2.68 | 44.2 | 14.2 |
| Abstract  | Network | 31.6 | 3.09 | 12.8 | 11.7 |
| Abstract  | Resistant | 31.6 | 2.55 | 11 | 11.2 |
| Abstract  | Outcome | 31.2 | 1.93 | 0.178 | -2.95 |
| Abstract  | Discharge | 31.1 | 3.79 | 9.99 | 9.02 |
| Abstract  | 2014 | 30.3 | 1.84 | 0.588 | -1.04 |
| Abstract  | Practice | 29.5 | 2.47 | 0.508 | -1.33 |
| Abstract  | Culture | 28.9 | 2.14 | 0.378 | -1.66 |
| Abstract  | Positive | 28.8 | 1.96 | 0.346 | -2.52 |
| Abstract  | Gene | 28.3 | 1.29 | 0.00000024 | -0.0415 |
| Abstract  | Disease | 28 | 2.42 | 0.365 | -3.7 |
| Keyword   | Enterococci | 27.5 | 1.92 | 25.2 | 6.33 |
| Abstract  | Month | 27.3 | 1.86 | 0.288 | -2.44 |
| Abstract  | Healthcare & Facility | 26.2 | 1.74 | 34.9 | 17 |
| Abstract  | Prevalence | 26.2 | 1.71 | 4.08 | 6.69 |
| Abstract  | Effort | 26.1 | 1.22 | 6.64 | 8.57 |
| Abstract  | Length | 25.3 | 1.44 | 6.1 | 6.45 |
| Keyword   | System | 25 | 1.43 | 11.9 | 3.47 |
| Abstract  | Laboratory | 24.4 | 1.38 | 0.442 | -1.39 |
| Keyword   | Resistant Staphylococcus Aureus | 24.4 | 1.59 | 22.8 | 11.2 |
| Abstract  | Clinical | 23.6 | 2.1 | 0.421 | -2.9 |
| Abstract  | Dataset | 22.5 | 1.47 | 8.75 | 6.5 |
| Abstract  | Development | 22.2 | 1.22 | 0.148 | -2.68 |
| Abstract  | Hand | 22 | 1.97 | 0.822 | -0.335 |
| Keyword   | Pathogen Transmission | 21.8 | 3.26 | 67.9 | 7.2 |
| Keyword   | Cross Infection & Humans & Transmission | 21.7 | 1.4 | 31.1 | 15.5 |
| Abstract  | Flow | 21.4 | 1.14 | 4.07 | 4.56 |
Table 3. Hyperparameter clusters and best cluster subsets. For each cluster, the defining rules and mean Sens. x Eff. is shown, followed by the per-cluster best set results.

| Cluster (mean score) | Num. iterations | Positive matches | Reviewed records | Sensitivity | Efficiency | Score (Sens. x Eff.) | Num. ensemble models | Uncertainty interval | Resampling | Num. initial training records | Positives oversampling multiplier |
|----------------------|------------------|------------------|------------------|-------------|------------|---------------------|----------------------|---------------------|-----------|-----------------------------|---------------------------------|
| 1. Mult = 10,20 & Mods = 10,20,40,60 & Resamp = n & Init = 100,250 (0.64) | 7 | 81 / 82 | 462 / 1200 | 98.8% | 61.5% | 0.608 | | | | | |
| 3. Mult = 10,20 & Mods = 10,20,40,60 & Resamp = n & Init = 50,500 (0.5) | 6 | 81 / 82 | 589 / 1200 | 98.8% | 50.9% | 0.503 | | | | | |
| 4. Mult = 10,20 & Mods = 1,5 & Resamp = n & Mods = 5 (0.47) | 8 | 82 / 82 | 1123 / 1200 | 100% | 6.42% | 0.0642 | | | | | |
| 5. Mult = 10,20 & Mods = 10,20,40,60 & Resamp = y & Init = 250,500 & Mods = 10 (0.39) | 5 | 82 / 82 | 685 / 1200 | 100% | 42.9% | 0.429 | | | | | |
| 6. Mult = 10,20 & Mods = 1,5 & Resamp = n & Mods = 10,20,40,60 & Init = 250,500 (0.31) | 5 | 82 / 82 | 874 / 1200 | 100% | 27.2% | 0.272 | | | | | |
| 7. Mult = 10,20 & Mods = 10,20,40,60 & Resamp = y & Init = 50,100 (0.23) | 7 | 82 / 82 | 948 / 1200 | 100% | 21% | 0.21 | | | | | |
| 8. Mult = 10,20 & Mods = 1,5 & Resamp = n & Mods = 1 & Init = 50,100 (0.078) | 6 | 82 / 82 | 1139 / 1200 | 100% | 5.08% | 0.0508 | | | | | |
| 9. Mult = 1 & Init = 250,500 & Mods = 20,40,60 (0.21) | 60 | 82 / 82 | 806 / 1200 | 100% | 32.8% | 0.328 | | | | | |
| 10. Mult = 1 & Init = 50,100 (0.054) | 5 | 82 / 82 | 832 / 1200 | 100% | 30.7% | 0.307 | | | | | |
| 11. Mult = 1 & Init = 250,500 & Mods = 1,5,10 (0.025) | 10 | 82 / 82 | 981 / 1200 | 100% | 18.2% | 0.182 | | | | |
Databases’ search guide

2022-02-19

WOS

- activate your institution proxy or log in in some way (it is only needed to get the API key, then you can log off the proxy)
- go to https://apps.webofknowledge.com/
- extract the API key from the URL, is denominated by SID
- put the key in the secrets.R file and source it or source Setup.R
- perform the search using the API

IEEE

- get your API key and put it into the secrets.R
- source the file source Setup.R
- perform the search using the API

Scopus

- change the NOT in the query to AND NOT
- go to https://www.scopus.com/search/form.uri?display=basic#basic
- log in with your institution credential
- set up the year range
- search within: title, abstract, keywords, authors
- select years in order to have just less than 2000 records
- download csv (Excel). Choose “Citation information” and “Abstract & keywords”
- call the files Scopus#.csv with # being a sequential number
- perform_search_session() will find and parse the files

Pubmed

- put your API key in the secrets.R file and source it or source Setup.R
- perform the search using the API
- put the same query into pubmed and download records in the Pubmed format.
- only 10k records can be downloaded at once, so use the year filter to partition the records
- call the file Pubmed#.nibib with # being a sequential number
- perform_search_session() will find and parse the files

Embase

- Login from within the institution or activate remote access
- go to https://www.embase.com/#advancedSearch/default
- set your date range
- in the mapping field select all apart from “Limit to terms indexed in article as ‘major focus’”
- in “Sources” select Embase
- select records in blocks of 10K
- chose format: Csv - fields by column, output: Full record (may take a while to prepare the download and you can download only one batch at time)
• call the files Embase#.csv with # being a sequential number