

Study Selection for Systematic Reviews: What We Know and Tools that can Help (A Facilitated Discussion)

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Study Identification or Study Selection or Sifting or Screening

AKA The Process of "Screaming"!



Within the Total Picture!

• "...the amount of time spent screening documents (based on title and abstract only) for relevance is estimated in the range of 10-20% of the total time it takes to plan and conduct a systematic review...." (Haddaway & Westgate, 2018)

Haddaway, N. R. and Westgate, M. J. (2018). Predicting the time needed for environmental systematic reviews and systematic maps. *Conservation Biology*, 33(2), 434-443. <u>https://doi.org/10.1111/cobi.13231</u> "In 2022, we estimated the workload of title and abstract screening for systematic reviews to range from 211,013 to 422,025 person-hours. Limiting human screening to citations which were not "ruled out" by GPT models could reduce workload by 65% and save up from 106,268 to 276,053person work hours (i.e.,66 to 172-person years of work), every year".

Tran, V. (2023). Sensitivity, specificity and avoidable workload of using a large language models for title and abstract screening in systematic reviews and meta-analyses.. https://doi.org/10.1101/2023.12.15.23300018

Study Selection requires:

 Objective criteria with good agreement between reviewers

- Clear decision rules for whether article is included, excluded or requires referral
- Rules to resolve uncertainties and disagreements e.g. additional information (full text or authors), additional reviewers' input, voting, or discussion and consensus

Study Selection - more to it than we might think!

1. Removing duplicates:

- a. Collating search results from multiple databases
- *b. Identifying and removing duplicate records*

2. Title and abstract screening:

- a. Developing screening checklist based on inclusion/exclusion criteria
- b. Pilot testing the screening checklist
- c. Independently screening titles and abstracts by at least two reviewers (sample or entire set)
- d. Comparing screening results between reviewers
- e. Resolving conflicts and disagreements through discussion or involving a third reviewer
- f. Documenting the reasons for exclusion

3. Full-text screening:

- a. Retrieving full-text articles for potentially eligible studies
- b. Independently screening full-text articles by at least two reviewers using inclusion/exclusion criteria
- c. Comparing screening results between reviewers
- d. Resolving conflicts and disagreements through discussion or involving a third reviewer

- e. Documenting the reasons for exclusion
- f. Hand-searching reference lists of included studies for additional relevant articles

g. Contacting authors for missing information or clarification, if necessary

4. Documenting the study selection process:

a. Creating a flowchart (e.g., PRISMA) to illustrate the study selection process

b. Reporting the number of studies identified, screened, eligible, and included

c. Providing a list of excluded studies with reasons for exclusion

5. Assessing inter-rater reliability:

a. Calculating inter-rater agreement (e.g., Cohen's kappa) between reviewers

b. Reporting inter-rater reliability in the systematic review

6. Updating the study selection process:

a. Rerunning the searches before finalizing the systematic review to identify newly published studies

b. Screening and incorporating any new eligible studies into the review

What is expected? – Study Selection

- Team members screen from 0.13 to 2.88 abstracts/ per minute.
- Conflict resolution–for dual screening processes–took average 5 min/conflict
- Retrieving full-text articles took 4 minutes per full text
- Full-text screening from 4.3 to 5 minutes per full text. Conflict resolution took 5 minutes per full text.

(Nussbaumer-Streit et al, 2021)

• One study showed that diagnostic test accuracy (DTA) reviews have on average 185% more workload during abstract screening and 167% more during full-text screening, because searches for DTA reviews identify many more records [51]. Shemilt et al (2014) analyzed:

(1) single screening;

(2) single screening with semiautomated software (one person screens abstracts and automatically excludes those deemed irrelevant by algorithm);

(3) dual screening (two people independently screen all abstracts and discuss conflicting decisions); and

(4) safety first (two people independently screen all abstracts, all potentially relevant abstracts included for full-text screening).

- All approaches followed by dual independent full-text screening. Based on 12477 abstracts, semi-automated approach took least time (572 hours) and cost (£ 37,860).
- Dual screening took twice as long (1089 hours) and cost nearly twice as much (£ 75,139).

What might we change?

- 1. Partial or Prioritised screening
- 2. PICo-based Title Screening
- 3. Use Single reviewer
- 4. Use large team of reviewers
- 5. Partial validation/checking
- 6. Reduced documentation i.e. only coding includes
- 7. Use data mining as a second sifter (semiautomated) [cp. PubMed 'Relevance']
- 8. Train AI to reorder relevance rankings

PICO-Based Title Screening (Rathbone et al, 2017)

- Search terms for screening generated from inclusion criteria of each review, specifically PICo elements—Participants, Interventions and Comparators.
- Used Synonyms for PICo terms, including alternatives for clinical conditions, trade names of generic drugs and abbreviations for clinical conditions, interventions and comparators.
- MeSH database, Wikipedia, Google searches and online thesauri to generate terms.
- Title-only screening by five reviewers independently in Endnote X7 using OR Boolean operator.

- Across 10 reviews, reduction in screening from 11-78% (median = 53%). In 9 SRs, recall of included studies = 100%. In one review, 4/5 reviewers missed same included study (median recall 67%).
- Rescreened using only Intervention and Comparator keywords (omitting keywords for Participants). Reduction of effort = 57%. Recall was maintained (100%).

Several RRs employed only one reviewer for title/abstract screening and data extraction (Ganann et al, 2010).

Single-reviewer abstract screening missed 13 percent of relevant studies (Gartlehner et al, 2020)

Bias may be introduced:

- selection bias
- single observer bias
- confirmation bias
- Dual study selection only if > 3 months (AHRQ, 2015)

FAQ: How Many Sifters?

"Use (at least) two people working independently to determine whether each study meets the eligibility criteria. Ideally, screening of titles and abstracts to remove irrelevant reports should also be done in duplicate by two people working independently (although it is acceptable that this initial screening of titles and abstracts is undertaken by only one person). It is essential, however, that two people working independently are used to make a final determination as to whether each study considered possibly eligible after title/abstract screening meets the eligibility criteria based on the full text of the study report(s)" (**Cochrane Handbook**)

Double Sift – All Stages?
Double Sift – Either Ti, Ab or Full Text?
Single Sift – Plus Random Sample (10% or 20%)?
Double Sift during Pilot, Single once Inter-Rater Reliability is Acceptable?
Text Mining as a Second Sifter?

Reflection: Are we doing this as a tick box exercise

or a training exercise?

Methodological Shortcuts (Pham et al, 2016)



Figure 1. Overview of study process for evaluating the potential implications of methodological shortcuts on the outcome of rapid reviews. This process was repeated for each of three original systematic review meta-analyses.

Pham et al (2016) SINGLE SCREENING A = 89%-100% SINGLE SCREENING B = 42%-83%

How reliable?

"Single-reviewer screening of abstracts by two investigators missed 4.4% and 5.3% of relevant studies, respectively" Stoll, C., Izadi, S., Fowler, S. A., Green, P., Suls, J., & Colditz, G. A. (2019). The value of a second reviewer for study selection in systematic reviews. *Research Synthesis Methods*, 10(4), 539-545. <u>https://doi.org/10.1002/jrsm.1369</u>

"A systematic **review** detected **four studies of the accuracy of single-reviewer compared with dualreviewer screening** (Waffenschmidt et al, 2019). Across these publications, the median proportion of **missed studies** was 5%."

Waffenschmidt, S., Knelangen, M., Sieben, W., Bühn, S., & Pieper, D. (2019). Single screening versus conventional double screening for study selection in systematic reviews: a methodological systematic review. BMC Medical Research Methodology, 19(1). <u>https://doi.org/10.1186/s12874-019-0782-0</u>

A false negative during screening is effectively lost, as an inappropriately excluded article joins a possibly large pool of articles that the research team believes to be irrelevant; **a false negative in this case is a needle thrown back into the haystack**. The absence of false negatives by DistillerAI is promising compared to the 0.6–16.6% rate of false negatives by single human assessors reported in a systematic review by Waffenschmidt and colleagues (Waffenschmidt et al, **2019**) and the 2.5% rate of false negatives observed with pairs of human assessors (Gartlehner et al, <u>2020</u>)...."

<u>Using an artificial intelligence tool can be as accurate as human assessors in level one screening for a systematic</u> <u>review. Burns¹, Etherington², Cheng-Boivin³ et al.</u> 2021 <u>Health Info Libraries J</u> "Two reviewers performed a **complete dual review** of 15,000 search results (**throughout the title/abstract and full-text stages**) and a **limited dual review** of 15,000 search results (**through full text stage only**). The number of relevant studies mistakenly excluded by highly experienced reviewers in the complete dual review was compared to the number mistakenly excluded during the full-text stage of the limited dual review.

In the complete dual review approach, an additional 6.6% to 9.1% of eligible studies were identified during the title/abstract stage by using two reviewers, and an additional 6.6% to 11.9% of eligible studies were identified during the full-text stage by using two reviewers *(i.e. cumulative 13.2% to 20%)*. In the limited dual review approach, an additional 4.4% to 5.3% of eligible studies were identified with the use of two reviewers *(i.e. cross-sectional)*.

Using a second reviewer throughout the entire study screening process can increase the number of relevant studies identified for use in a systematic review. Systematic review teams should consider using a complete dual review process to ensure all relevant studies are included in their review."

Stoll CRT, Izadi S, Fowler S, Green P, Suls J, Colditz GA. The value of a second reviewer for study selection in systematic reviews. *Res Synth Methods*. 2019;10(4):539-545. doi:10.1002/jrsm.1369

How long?

"An experienced reviewer takes **from 30 seconds to several minutes** to evaluate a citation [Wallace et al 2010]. The work involved in screening 10,000 citations is considerable (and screening burden in some reviews is considerably higher...)" (see also [Allen & Olkin, 1999]). In: O'Mara-Eves et al, 2015.

Wallace B, Trikalinos T, Lau J, Brodley C, Schmid C: **Semi-automated screening of biomedical citations for systematic reviews.** *BMC Bioinformatics* 2010.,**11**(55): Allen I, Olkin I: **Estimating time to conduct a meta-analysis from number of citations retrieved.** *JAMA* 1999,**282**(7):634–5. 10.1001/jama.282.7.634

Research suggests that adopting automation can reduce the need for manual screening by at least 30% and possibly more than 90%, although sometimes at the cost of up to a **5% reduction** in sensitivity (<u>O'Mara-Eves et al 2015</u>).

How much Time?

"...The Cochrane Handbook for Systematic Reviews of Interventions suggests that title and abstract screening can be completed at a rate of 120 papers per hour [16]...."

"With well-defined criteria, in our experience a trained screener can screen up to 200 titles and abstracts per hour, but not for more than 2 hours per day...." [Leenaars, C., et al. (2020). Reviewing the animal literature: how to describe and choose between different types of literature reviews. Laboratory Animals, 55(2), 129-141. <u>https://doi.org/10.1177/0023677220968599</u>]

"...An SLR with 5000 abstracts can take an experienced reviewer between 60 to 85 h to complete abstract screening (approximately 500-800 abstracts per working day, which we assume to be 8 h) [8, 10], compared with 5 min for the algorithm...." [Popoff, E., et al. (2020). Aligning text mining and machine learning algorithms with best practices for study selection in systematic literature reviews. Systematic Reviews, 9(1). <u>https://doi.org/10.1186/s13643-020-01520-5</u>]

The Cochrane "Rule of Thumb"

"Article abstracts identified through a database search can usually be screened very quickly to ascertain potential relevance. At a conservatively estimated reading rate of one or two abstracts per minute, the results of a database search can be screened at the rate of 60–120 per hour (or approximately 500–1000 over an 8-hour period!!), so the high yield and low precision associated with systematic review searching may not be as daunting as it might at first appear in comparison with the total time to be invested in the review".

Lefebvre C, Glanville J, Briscoe S, et al. Chapter 4: Searching for and selecting studies. In: Higgins JPT, Thomas J, Chandler J, Cumpston M, Li T, Page MJ, Welch VA (editors). *Cochrane Handbook for Systematic Reviews of Interventions* version 6.4 (updated October 2023). Cochrane, 2023. Available from www.training.cochrane.org/handbook.

Study selection: rationale

To identify relevant studies in a way which is:

- Systematic
- Replicable
- Free from bias

What strategies are reported within systematic literature reviews of software engineering and computer science? (K. Petersen and N. B. Ali, "Identifying Strategies for Study Selection in Systematic Reviews and Maps," *2011 International Symposium on Empirical Software Engineering and Measurement*, Banff, AB, Canada, 2011, pp. 351-354, doi: 10.1109/ESEM.2011.46).

Thirteen different strategies for inclusion/exclusion have been identified.

Three are used to assure objective inclusion/exclusion criteria to reduce bias, three to resolve disagreements and uncertainties due to bias, and seven defined decision rules on how to handle disagreements/agreements

Thirteen different strategies for inclusion/exclusion (Petersen & Ali 2011)

Table I STRATEGIES OBJECTIVE CRITERIA

ID	Code	Descr.	No. of citations
01	Objective Criteria Assessment (pre- inclusion/exclusion)	Test sub-sets of articles with two or more persons before starting the actual inclusion/exclusion process, high agreement indicate objective criteria	5
02	Objective Criteria Assessment (post- inclusion/exclusion)	Reviewers measure their agreement after completing inclusion/exclusion to determine level of objectivity on all or a sample set of studies (can also be	10
03	Objective Formu- lation of Criteria	done on a sub-set) Require objective statements (e.g. is X stated, Yes/No)	1

Table II STRATEGIES RESOLVE DISAGREEMENTS/UNCERTAINTIES

ID	Code	Descr.	No. of citations
RI	Another person check	Additional reviewer(s) is/are con- sulted to support in the decision of inclusion or exclusion by review- ing/assessing the result	14
R2	Second vote on uncertain and ex- clude	Only for papers rated as either un- certain or exclude a second vote is obtained.	1
R3	If disagreement or uncertainty in decision then discuss	If a set of researchers is in dis- agreement, then a decision for the next step is taken after discussion to resolve the disagreement.	18

Table III DECISION RULES TO DIRECTLY ARRIVE AT A DECISION

ID	Code	Descr.	No. of citations
Dl	Majority Vote	The group of researchers take a vote on the article and the decision of the majority is followed	1
D2	At least one "in- clude" then in- clude	If one of the reviewers includes the paper then it is considered for being an included primary study	1
D3	All "include" then include	Only if all reviewers include the article then it is included, otherwise it is excluded	1
D4	At least one "un- certain" then in- clude	If one of the reviewers is uncertain regarding the paper, it is considered in the next step of the systematic review	11
D5	One "exclude" and one "uncertain" then exclude	If one of the reviewers says exclude the other uncertain then the paper is excluded	1
D6	All researchers vote "uncertain" then include	If all researchers vote uncertain, then the paper is included	1
D7	All "exclude" then exclude	If all reviewers agree that the pa- per should be excluded then it is excluded, otherwise it is included	1

Study Selection Process

Step 1: Apply Inclusion/Exclusion Criteria to Titles and Abstracts

- Step 2: **Eliminate** Studies That Clearly Meet One or More Exclusion Criteria ("**Ruling Out**")
- Step 3: Retrieve the Full Text of the Remaining Studies
- Step 4: Evaluate the Remaining Studies for Inclusion and Exclusion
- Step 5: **Include** Studies That Meet All Inclusion Criteria and No Exclusion Criteria ("**Ruling In**")
- Step 6: Exclude Studies From Systematic Review With Reasons
- Step 7: Accept Studies for Systematic Review



Fig. 1. Overview of the selection process

Questions So Far?

Study Selection Software

Aka: Eros, Parsifal or Bert? – Who will you entrust yourself to?



Reviewer perspective (Easy study selection)

Versus

"**Review Manager**" perspective (Setting up double screening; Adding accounts; Monitoring progress)

Versus

"**Review Administrator**" (Costing model e.g. per Reviewer, Per Review etc, Licenses, Institutional Agreements)



Friend (admiring small car). "VERY NICE; BUT HOW DO YOU-ER-MANAGE TO GET INTO IT ?" Large Owner. "I don't. I just slip it on."

Software to support selecting studies can be classified as:

- systems that support the study selection process, typically involving multiple reviewers (see Section <u>4.6.6.1</u>); and
- tools and techniques based on text mining and/or machine learning, which aim to semi- or fully-automate the selection process (see Section <u>4.6.6.2</u>).

Software to support the selection process can be identified using the **Systematic Review Toolbox** (45 tools – Sutton (2024) *Personal Communication*). The Systematic Review Toolbox is a community driven, web-based catalogue of tools to support systematic reviews (Marshall and Brereton 2015).

Software for managing the selection process

Software for managing the selection process

Managing selection can be challenging, particularly in a large-scale systematic review that involves multiple reviewers.

Basic productivity tools can help (such as word processors, spreadsheets, and reference management software)

Several purpose-built systems that support multiple concurrent users are also available that offer support for the study selection process. See Systematic Review Toolbox.

Compatibility with other software tools used in the review process (such as RevMan) may be a consideration when selecting a tool to support study selection. Covidence and EPPI-Reviewer are Cochrane-preferred tools, and are likely to have the strongest integration with RevMan.

Bramer and colleagues have reported a method for using EndNote X7 for managing the screening process (<u>Bramer et al 2017</u>). (Cochrane Handbook)

Automating the selection process

Tools and techniques for reviewers to consider fall within three main areas: a) those that operate across reviews; b) those that use crowdsourcing to reduce reviewer workload; and c) those that operate within individual reviews.

a) **Tools that operate across reviews** are machine learning models (or 'classifiers') that can be built where sufficient data are available e.g. the 'RCT Classifier' can identify reports of randomized trials based on titles and abstracts. The classifier is built on a large dataset of hundreds of thousands of records screened by <u>Cochrane Crowd</u>, Cochrane's citizen science platform, where contributors help to identify and describe health research (<u>Marshall et al 2018, Noel-Storr et al 2021a</u>, <u>Thomas et al 2021</u>). See <u>Cochrane Information Specialists'</u> <u>Handbook</u> (Cochrane Information Specialist Support Team 2021c).

Tools built on 'Large Language Models' (LLMs) (e.g. ChatGPT) may help in automating study selection. These models promise 'zero shot learning', where records can be classified automatically without the need for any specific training. Such approaches may offer substantial benefits, though, at the time of writing in mid-2023, no sufficiently large and valid evaluations are available.

b) Crowdsourcing tools: Cochrane has also implemented a screening workflow called Screen4Me (<u>Noel-Storr et al 2021b</u>).

This workflow incorporates the RCT Classifier and Cochrane Crowd, to identify the RCTs found in authors' search results. As of July 2023, Screen4Me has been used in over 200 Cochrane intervention reviews. An unpublished, evaluation (AN-S) showed a mean reduction in screening workload of 53% (range 26% to 84%).

c) Active learning by machine learning models that "learn" how to apply eligibility criteria within individual reviews.

Semi-automates study selection by continuously promoting records most likely to be relevant to the top of the results list (<u>O'Mara-Eves et al</u> <u>2015</u>). Requires authors to prespecify "stopping rules" to stop screening and allow some records to be eliminated automatically without manual assessment. Recent work suggests it is possible to estimate how many relevant records remain to be found based on the sample already screened (<u>Sneyd and Stevenson 2019</u>, <u>Callaghan and Muller-Hansen 2020</u>, <u>Li and Kanoulas 2020</u>). More work needed to develop and validate safe 'stopping rules'. However can be used to prioritize records for screening so that authors identify potential includes much earlier in the screening process.

Recent developments have seen increased support for living systematic reviews. Two case studies found that automation to support living reviews in COVID-19 were accurate and saved manual effort (<u>Shemilt et al 2021</u>, <u>Marshall et al 2023</u>).

Tools are available that use natural language processing to highlight sentences and key phrases automatically (e.g. PICO elements, trial characteristics, details of randomization) (**cp Rayyan**) to support the reviewer whilst screening (<u>Tsafnat et al 2014</u>).

What are the Choices?

Study Selection - Generic

Excel

Google Sheets

Study Selection – Function Specific

Abstrackr (Rathbone J, Hoffmann T, Glasziou P. Faster title and abstract screening? Evaluating Abstrackr, a semi-automated online screening program for systematic reviewers. *Syst Rev.* 2015;**4**:80. doi: 10.1186/s13643-015-0067-6.)

Study Selection – Systematic Review Software

Covidence (Covidence. Better systematic review management. <u>https://www.covidence.org</u>)

Study Selection – Systematic Review Software with Prioritization

Colandr (Kahili-Heede, M. and Hillgren, K. (2021). Colandr. Journal of the Medical Library Association Jmla, 109(3). https://doi.org/10.5195/jmla.2021.1263)

Distiller-AI. Gartlehner, G., Wagner, G., Lux, L. J., Affengruber, L., Dobrescu, A., Kaminski-Hartenthaler, A., ... & Viswanathan, M. (2019). Assessing the accuracy of machine-assisted abstract screening with Distiller AI: a user study. *Systematic Reviews*, 8(1). https://doi.org/10.1186/s13643-019-1221-3

EPPI Reviewer (EPPI-Centre. EPPI-Reviewer. http://eppi.ioe.ac.uk/cms/Default.aspx?alias=eppi.ioe.ac.uk/cms/er4. Accessed 20 Oct 2022.)

RobotAnalyst (Przybyła, P., Brockmeier, A. J., Kontonatsios, G., Pogam, M. L., Elm, E. v., Nolan, K., ... & Ananiadou, S. (2018). Prioritising references for systematic reviews with RobotAnalyst: a user study. Research Synthesis Methods, 9(3), 470-488. https://doi.org/10.1002/jrsm.1311)

Rayyan (Ouzzani M, Hammady H, Fedorowicz Z, Elmagarmid A. Rayyan: a web and mobile app for systematic reviews. *Syst Rev.* 2016;5:210. doi: 10.1186/s13643-016-0384-4. and Yu, F., Liu, C., & Sharmin, S. (2022). Performance, usability, and user experience of Rayyan for systematic reviews. *Proceedings of the Association for Information Science and Technology*, 59(1), 843-844. https://doi.org/10.1002/pra2.745)

Study Selection – Home Made

Catchii (Halman, A. (2023). Catchii: empowering literature review screening in healthcare. *Research Synthesis Methods*, 15(1), 157-165. <u>https://doi.org/10.1002/jrsm.1675</u>)

WebTSDB (Study selection by means of a web-based Trial Selection DataBase (webTSDB) <u>https://abstracts.cochrane.org/2011-madrid/study-selection-means-web-based-trial-selection-database-webtsdb</u>



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Use of social media associated with poorer sleep quality and low selfesteem in teenagers.

The need to be available constantly on social media is causing depression, anxiety and poor sleep quality in teenagers, new study results suggest.

Nursing standard (Royal College of Nursing (Great Britain) : 1987) Sep 2015;30(4):15



Covidence – Screening Tool used by Cochrane

Progress Reporting



PRISMA 2020 flow diagram template for systematic reviews.



Matthew J Page et al. BMJ 2021;372:bmj.n71



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Superconducting critical fields of alkali and alkaline-earth intercalates of MoS2 Date: 1976-01-01 Woollam, J A; Somoano, R B	Abstract:
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Human Frequency Response as a Function of Visual Feedback Delay Date: 1968-01-01 Leslie, J M; Thompson, D A	have the highest critical temperatures, critical fields, and critical boundary slopes. In all cases the hexagonal materials have critical fields exceeding the paramagnetic limiting fields. The second group, consisting of the nonhexagonal Na, Sr, and Ca intercalates, have lower critical temperatures and fields, and the paramagnetic limit is not exceeded. The American Physical Society.
Electrical Conductivity Studies Over a Tectonically Active Area in Eastern Canada. Date: 1974-01-01 Bailey, R C; Edwards, R N; Garland, G D; Kurtz, R; Pitcher, D	Publication Types: Journal Article.
Magnetic Lines of Force Deformed by Motion of a Conducting Fluid (II) Date: 1969-01-01 Hagiwara, Y	Topics: Alkalies. Authors:
Some dynamic properties of linear, hyperbolic and sigmoidal multi-enzyme systems Date: 1974-01-01 Walter, C F	Woollam, J A, Somoano, R B. Journal: Physical Design R = Veloce 12 January 0, pp. 2242-2252 - published 1076-01-01
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The geotectonic cycle and the new global tectonics Date: 1970-01-01 Coney, P J	Include ? Maybe X Exclude with Reasons Icabel Image: Add note Image: Add note Image: Add note

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Abstract:

Congenital cytomegalovirus (cCMV) infection is a leading cause of hearing loss and neurological disabilities in children, with the disease burden and disabilities due to cCMV greater than many other well recognized childhood conditions. A minority of infants with cCMV will have symptoms at birth. Infants with symptomatic cCMV are at higher risk for sequelae than those born without symptoms. The majority of infants with cCMV are asymptomatic at birth, but 10%-15% will develop hearing loss. Although clinical symptoms can help predict which infants will have sensorineural hearing loss, among asymptomatic cCMV there are currently no predictors of adverse outcome. The identification of a biomarker to identify those at highest risk of sequelae is highly desirable to target interventions to those who could potentially benefit. Because there is increasina rationale for establishing both targeted and universal screening programs for cCMV in the United Labels

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Congenital Cytomegalovirus Infection. Date: 05-3-2020

? Bashar new label

Congenital cytomegalovirus infection diagn... Date: 01-10-2022

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Hearing loss and congenital CMV infection: a... Date: 01-11-2014

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Cytomegalovirus infection during pregnancy... Date: 01-9-2020

An Example



Systematic Review Accelerator

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Potential Inclusion Criteria (Harrison et al, 2020)

- 1. Software is currently accessible
 - i. The website hosting this software tool must currently exist
 - ii. It is possible either to access this tool online or download and install the tool
- 2.It is possible to test the software for free
 - i. This requires that either the tool is free to use or there is a free trial available
 - ii. Where a free trial was not automatically available, then the company or organization hosting the application was contacted to request one
- 3. The software has reasonable system requirements
 - i. The user is not required to provide specific computing infrastructure (such as an SQL server) in order to use the software
- 4. Provide basic screening functionality for SRs
 - i. The tool can be used to screen references (at least by title and abstract). Requires additional functionality above what is provided by a reference manager
- 5. The software is working (it is possible to carry out a test project)
 - i. The user must be able to carry out basic tasks (such as importing references) with the tool

Tasks (Harrison et al, 2020 modified)

- 1. Creating an account
- 2. Creating a systematic review project
- 3. Importing references
- 4. Inviting collaborators to join the project
- 5. Assigning records to screeners
- 6. Carrying out T&Ab screening on the references
- 7. Comparing Verdicts
- 8. Comparing Inter-Rater Reliability
- 9. Exporting the screened references
- 10. Finding and using the help section

Some comments on Covidence (Acknowledgment to Sue Harnan):

Consensus

• Annoying process to change decisions, both reviewers have to go back in to change decisions (unless there is a way around this that I couldn't find)

Coding

• Tagging is a bit time-consuming

Search functions

- Can't use Boolean Logic in Covidence to retrieve records
- Search facility does very poorly when searching for authors with names that either occur frequently, or are small words such as Xu. Probably better to use title.

Import/Export

• Can't export whole sift easily, have to do it in batches according to what decision was made at which point in the sift (e.g. can export all title-abstract excludes, then separately all full text includes etc). I'd personally like more access to the back-end database and a better search facility.

Value Added facilities

• PRISMA flow chart generation is a great feature.

Feature Analysis (Harrison et al, 2020)

Themes	Features	Weighting ^a
Economic	The tool does not require financial payment to use.	HD
Ease of Introduction	The tool has straightforward system requirements	HD
and Setup	There is an installation guide (where applicable)	D
	There is a tutorial/help section	D
	The software does not require user to code	HD
	There is an app for mobile/tablet	D
Systematic Review	Supports deduplication	D
Support	Supports title and abstract screening	-
	Supports full text screening	D
	Supports data extraction	N
	Supports other stages of the review	N
Process Management	Support for multiple users	М
	Support for multiple projects	D
	Choice of single or double screen before progression	HD
	Work Allocation	HD
	Management of roles	D
Reference	Import of References	-
Management	Export of References	Μ
	Export of Decisions	Μ
	Import of .pdfs	D
Workflow	The tool is flexible to varying workflow	HD
	Short User Set-up (before screening can begin)	D
	Progress is monitored and fed back to user	HD
Screening Features	Include/Exclude Option	-
	Key word highlighting (or similar)	D
	Can filter citations by category	D
	Can search citations (i.e. search engine)	D
	Further categorize/label references	HD
	Blind screeners to decisions of others.	HD
	Conflict Resolutions	HD
	Citation classification/ranking tool (clustering/ML)	N
Security	Insecure website	HD

a. Abbreviations:M Mandatory HD Highly Desirable D Desirable N Nice to Have

I Irrelevant

Comparison (Harrison et al, 2020)



<u>5 software tools to support your systematic review processes (ifis.org)</u> [Jan 2021]

Function comparison of 5 software tools to support the systematic review process

Functions	Covidence	Rayyan	EPPI-Reviewer	CADIMA	DistillerSR
Protocol development	×	~	×		~
Database integration	×	×	Only PubMed	×	PubMed
Ease of import & export	~	×	×	×	×
Duplicate removal	~	×	×	×	×
Article screening	Inc. full text	Title & abstract	Inc. full text	Inc. full text	Inc. full text
Article screening Critical appraisal	Inc. full text	Title & abstract	Inc. full text	Inc. full text	Inc. full text
Article screening Critical appraisal Assist with reporting	Inc. full text	Title & abstract	Inc. full text	Inc. full text	Inc. full text
Article screening Critical appraisal Assist with reporting Meta-analysis	Inc. full text	Title & abstract	Inc. full text	Inc. full text	Inc. full text

Rayyan provides a very powerful solution to completing title, abstract and full text screening using the concept of stages while leaving you in complete control over which data to add to full text screening. To add a Full Text Screening stage to your review, follow these steps:

- 1. Complete Title and Abstract Screening on your review
- 2. Click on the Full Text Screening tab on your review

Tayyan Teams+ Full Text Screening Overview Review Data Screening Full Text Screening

3. Click Enable to add Full Text Screening to your review

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Some Practical Tips

- 1. Title and Abstract sifting is for Ruling Out, Full Text is Ruling In
- 2. Consider pairing "Hawks" with "Doves"
- 3. Pilot Eligibility Criteria on X titles and abstracts (Cochrane Rapid Review Group)
- 4. Use an overlapping 20% as a Training Set
- 5. Use Kappa formatively rather than summatively
- 6. Make sure the 20% is a Random Sample (Information Specialists will create a Database Selection effect)
- 7. Decide whether to use Include/Exclude (e.g. Systematic Reviews), Include/Exclude/Full Text (e.g. Rapid Reviews) or Study PICO Codes for Categories of Includes (e.g. Mapping Reviews -> First Round Data Extraction)
- 8. Decide whether obvious types of records can be excluded in blocks e.g. pigs and mice!
- 9. Decide on Two Round (Title/Abstract; Full Text) versus Three Round (Title; Abstract; Full Text) for large reviews to rule out.

Cochrane rapid review methods recommendations - Study selection

Title and abstract screening

Use standardized title and abstract form, conduct pilot exercise **using same 30–50 abstracts** for entire screening team to calibrate and test review form.

•Use two reviewers for dual screen of **at least 20% (ideally more) of abstracts**, with conflict resolution.

•Use one reviewer to screen remaining abstracts and a second reviewer to screen all excluded abstracts, and if needed resolve conflicts. (R14)

Full-text screening

•Use standardized full-text form, conduct pilot exercise **using 5–10 full-text articles** for entire screening team to calibrate, and test review form.

•Use one reviewer to screen all included full-text articles and second reviewer to screen all excluded full-text articles. (R15)

Some comments on Covidence (Acknowledgment to Sue Harnan):

Training the system

Train the AI using known includes as well as known excludes and some that you don't know a priori are either (I've not tried this, I picked it up from a presentation from York or Cochrane I think)

Managing Consensus

Problems with double checking (if using two reviewers) the first 100 (to train each other) if you have the learning algorithm on as you both end up looking at different records - not sure if this is a problem with all the AIs, or just Covidence, and not sure how to get around it if you want to train the AI as well as train each other!

Review complexity

Problems with difficult reviews, or where there are multiple questions. Train the AI on examples of includes/excludes from all review questions? (again not tried this, but might work)

In my review, where we used the AI naively without training as above, and when reviewing multiple diagnostic technologies, there were includes right up to the end.

All of these can be fixed, but highlight the need to train the AI and train the humans before they dive in!

Decisions, decisions! 1. Select best for a specific function (e.g. study selection) or 2. Select best for overall review management?

3. Ease of Selection (e.g. Swipe Right!) or
4. Facility to Manage References (e.g. Having to Open Reference Management records – for Coding) or
5. Dragging and Dropping (Folders) Some things to consider: 1. Should we document which ESGs use which software? 2. Should we create virtual user groups/fora for the "leading brands"? 3. Should we have an annual update session on "what has changed since last year"? 4. Should the "AI for Study Selection" angle be covered differently from "Routine Study Selection"?

Appendix (Thanks to Anthea Sutton & Systematic Review Toolbox)

CADIMA - supports the conduct of systematic reviews and evidence/systematic maps by the provision of a freely available online tool that: 1. guides review authors through the evidence synthesis process, 2. facilitates the coordination of cooperating team members, 3. eases steps with considerable workload and 4. guarantees for its thorough documentation. The evidence synthesis tool was established and is further developed in a close collaboration between the Julius Kühn-Institut and the Collaboration for Environmental Evidence

Review Stages: Search, Screen, Data Extract, Quality Assess, Synthesis, Report, Reference Management

Cientopolis Scolr: supports collaboration in the process of conducting open, literature reviews.

Review Stages: Protocol, Search, Screen, Data Extract, Quality Assess, Synthesis, Report, Reference Management, Stakeholder Engagement

Colandr: An open access, machine-learning assisted tool for conducting evidence synthesis. This tool uses machine learning, natural language processing, and text-mining functions to partially automate finding relevant citations and extract desired data from PDF articles.

Review Stages: Search, Screen, Data Extract

Covidence - A web-based software platform that streamlines the production of systematic reviews, including Cochrane Reviews. Citation screening, Full text review, Risk of Bias assessment, Extraction of study characteristics and other study data, Export of data into RevMan. Nonprofit organization, open source software

Review Stages: Screen, Data Extract, Quality Assess

DistillerSR - Summary: A web based reference screening, data extraction and reporting solution for systematic reviews.

Review Stages: Protocol, Screen, Data Extract, Quality Assess, Synthesis, Report, Reference Management

EPPI-Reviewer: Web-based tool for managing and analysing data in systematic reviews. Review Stages: Screen, Data Extract, Synthesis

JBI-SUMARI: A System for the Unified Management, Assessment and Review of Information containing a suite of tools to support various aspects of the systematic review process. JBI run short courses on using the software: <u>http://joannabriggs.org/education/short-courses</u>

Review Stages: Protocol, Screen, Data Extract, Quality Assess, Synthesis, Report, Reference Management **Nested-Knowledge** is transforming the process and impact of systematic review with automations and augmentations of the review process and visualizing the results in interactive web-based meta-analyses. Two tools in one platform. AutoLit covers every step—search, screen, tag, and extract—using best practices in a single platform. Synthesis presents qualitative breakouts of the common concepts from studies, quantitative meta-analysis of patient outcomes, and narrative summaries of your findings. Synthesis is updated in real time, serving as a one-stop portal to your living systematic review. Perview Stages: Search, Screen, Data Extract, Quality Assess, Synthesis, Report, Reference Management

Review Stages: Search, Screen, Data Extract, Quality Assess, Synthesis, Report, Reference Management **Parsifal:** Web-based tool which supports multiple stages of a systematic review in software engineering. URL: Link

Review Stages: Protocol, Search, Screen, Data Extract, Quality Assess, Reference Management

Review Management Packages that Include Screening

PICO Portal: Web-based tool for citation screening, full text review, data extraction and quality assessment that facilitates an efficient systematic review. Aims to combine the strengths of a modern user interface and cutting-edge machine learning functionality. It offers a highly accurate deduplication that was benchmarked against published papers.

Review Stages: Screen, Data Extract, Quality Assess

Rax: Personalized AI-powered research assistant. It adapts to your changing needs as you move through various stages of your research project's life-cycle.

Review Stages: Search, Screen, Data Extract, Quality Assess, Synthesis, Report, Reference Management **ReLiS:** Highly configurable tool to conduct systematic reviews collaboratively and iteratively on the cloud. Review Stages: Protocol, Screen, Data Extract, Quality Assess

SESRA: Web application to support the Systematic Literature Review process for researchers and practitioners in the software engineering domain. SESRA uses the guidelines proposed by Kitchenham and Charters (2007). Review Stages: Protocol, Screen, Data Extract, Quality Assess, Report

SLR-Tool: A freely-available tool to support each stage of the SR process in software engineering.

Review Stages: Protocol, Screen, Data Extract, Quality Assess, Synthesis, Report

SRDB.PRO: Commercial software for managing and aiding systematic reviews.

Review Stages: Protocol, Search, Screen, Data Extract, Quality Assess, Synthesis, Report, Reference Management

StArt: State of the Art through systematic review (StArt) aims to provide support for each stage of the SR process in software engineering.

Review Stages: Protocol, Search, Screen, Data Extract, Quality Assess, Report, Reference Management

SyRF: Systematic Review Facility: Fully integrated online platform for performing systematic reviews of preclinical studies. SyRF provides a secure screening database, repository and analysis applications. It also provides educational resources on how to conduct and report a systematic review and guidance on preclinical systematic review and meta-analysis.

Review Stages: Screen, Data Extract, Synthesis

Sysrev: A web-based platform, Sysrev facilitates a range of document-review types, from pure data curation projects to systematic reviews. Sysrev's Label system enables flexibility in how users screen, tag, annotate, & sort documents and extract data. Articles can be uploaded via .XML, .RIS, and .PDF file types or via Sysrev's native Pubmed and ClinicalTrials.gov searches. Sysrev BASIC is a 100% Free subscription that enables users to create unlimited Public projects with as many reviewers as their projects require. Sysrev BASIC users also have access to Sysrev's machine learning models. Additional functionality available for a small fee.

Review Stages: Protocol, Search, Screen, Data Extract, Quality Assess, Synthesis, Reference Management

Systematic Review Accelerator: Free software developed at Bond University which includes a validated deduplication tool for faster deduplication of search results, a word frequency analyser to help with search strategy development, a search translator to speed up translation of searches from PubMed/Ovid MEDLINE to other major databases, and a hotkey tool to make screening articles in EndNote easier.

Review Stages: Protocol, Search, Screen, Reference Management

TaskExchange: Cochrane TaskExchange is a collaboration tool that connects people who need help or advice on health evidence projects with people who have the time and expertise to assist. People needing help post a task and people wanting to help out respond to tasks of interest. Most common tasks types are data screening, data extraction, consumer tasks and translation. Anyone can sign up to use the platform.

Review Stages: Screen, Data Extract, Quality Assess, Stakeholder Engagement

Thoth: A web-based support tool developed to support the SLR process in software engineering.

Review Stages: Protocol, Screen, Data Extract, Quality Assess, Report

Screening Specific Tools

Abstrackr: An online tool for the task of citation screening for systematic reviews.

Review Stages: Screen

AntConc: A freeware corpus analysis toolkit for concordancing and text analysis.

Review Stages: Screen

ASReview LAB is free and open source machine learning software, dedicated to accelerate the screening of textual data. Software comes with a modern user-friendly interface for screening texts, as well as an extensive simulation toolkit. Use simulations to explore how much work you can save by using ASReview LAB over manual screening. The software is installed locally, making sure that your data stays your own.

Review Stages: Screen

BioReader (Biomedical Research Article Distiller): A tool that enables users to perform classification of scientific literature by text mining-based classification of article abstracts. The tool is trained by uploading article corpora for two training categories - e.g. one positive and one negative for content of interest - as well as one corpus of abstracts to be classified and/or a search string to query PubMed for articles. The corpora are submitted as lists of PubMed IDs and the abstracts are automatically downloaded from PubMed, preprocessed, and the unclassified corpus is classified using the best performing classification algorithm out of ten implemented algorithms. BioReader is freely available as a web service.

Review Stages: Screen

Concept Encoder: A neural network–based artificial intelligence engine that analyses text by converting sentences into vectors. extract and learn each vector component as a feature value, identify similar vectors as indicators of the similarity of sentence content, and perform a rapid search for similar sentences. As such, it can be used to reduce the screening workload in systematic reviews. Note that the research study was performed in systematic reviews for clinical guidelines and the authors state that further investigation is needed to assess the effectiveness for meta-analyses of epidemiological studies.

Review Stages: Screen

DBPedia: A resource description framework repository to support automated selection of primary studies.

Review Stages: Screen

Disputatron: Tool designed to automatically detect screening disagreements between reviewers of a systematic review, who screened in EndNote.

Review Stages: Screen

DoCTER: ICF's Document Classification and Topic Extraction Resource, is a free web-based software application that helps explore and prioritize documents — such as abstracts from the scientific literature or social media messages — for expert review. Using algorithms from the domains of natural language processing and machine learning, DoCTER improves efficiency in tasks that involve large volumes of text.

Review Stages: Screen

EMB automated PICO identification tool: An end-to-end Entity Recogniser that identifies the PICO within medical publications. It identifies sentences within medical literature that contains the PICO, and answers the questions stated within the PICO. The dataset was manually annotated by medical practitioners.

Review Stages: Screen

R.ROSETTA: an interpretable machine learning framework: An R package for the construction and analysis of machine learning rule-based classifiers. It provides additional functions for statistics and visualisation of data.

Review Stages: Screen

Screening Specific Tools

Rayyan: Web based, collaborative application to support undertaking systematic reviews. Also includes a mobile app for screening studies on the go.

Review Families: Systematic, Rapid, Qualitative, Scoping, Mapping, Mixed Method, Other

Review Stages: Screen

Research Screener : A cloud-hosted Web application and algorithm that semi-automates abstract screening for systematic reviews. The algorithm applies deep learning and natural language processing methods to represent abstracts as text embeddings.

Review Stages: Screen

Screenatron: A tool designed to allow fast abstract screening when performing a systematic review.

Review Stages: Screen

SWIFT-Active Screener: A web-based, collaborative systematic review software application. Active Screener was designed to be easy-to-use, incorporating a simple, but powerful, graphical user interface with rich project status updates. What makes Active Screener special, however, is its behind-the-scenes application of state-of-the-art statistical models designed to save screeners time and effort by automatically prioritizing articles as they are reviewed, using user feedback to push the most relevant articles to the top of the list.

Review Stages: Screen

Screening Specific Tools

revtools: Software package currently being developed by Martin Westgate, a postdoc at the Australian National University in Canberra. Aims to produce interactive visualisation of patterns in bibliographic data to support systematic reviews and meta-analyses, allowing users to quickly get a feel for patterns in collections of articles, and to select those entries of most interest to them. New software but more updates planned. Downloadable from Github.

Review Stages: Screen

RobotSearch: Front-end for a machine learning model that identifies reports of randomized controlled articles (RCTs), from the developers of RobotReviewer

Review Stages: Screen

SWIFT-Review: An acronym for "Sciome Workbench for Interactive computer-Facilitated Text-mining"), is a freely available interactive workbench which provides numerous tools to assist with problem formulation and literature prioritization.

Review Stages: Screen

Syras: A flexible web-based product with easy to use solutions for abstract deduplication, abstract screening, and exporting of abstracts for systematic reviews. Suitable for small scale and large scale teams completing reviews. Made by passionate tech veterans "at a price-point that an academic would be happy to pay for".

Review Stages: Search, Screen

BeCAS: the Biomedical Concept Annotation System, is a web application, application programming interface (API) and widget for biomedical concept identification. BeCAS aims to help researchers, healthcare professionals and developers in the identification of over 1,200,000 biomedical concepts in text and PubMed abstracts using text-mining capabilities. Details of how to use the web app are available in the help section of the tool. If you want to integrate BeCAS annotation services in your own text-processing pipeline, this can be done via the API. Web developers can augment websites with text annotation and highlighting capabilities using the BeCAS widget. BeCAS is freely available for non-commercial use.

Review Stages: Search, Screen, Data Extract

BERT: (Bidirectional Encoder Representations from Transformers) is a tool developed by researchers at Google AI Language. BERT is a machine learning tool that can be used in a wide variety of NLP tasks, including Question Answering, Natural Language Inference, and others.

Review Stages: Screen, Data Extract

Buhos: A web-based systematic literature software that supplies the necessary functionalities for managing the complete literature review process, covering the stages of searching, screening, data extraction and reporting for social sciences.

Review Stages: Search, Screen, Data Extract, Report

EROS: Early Review Organizing System: Web-based software designed to assist with the initial phases of a systematic review: reference management, screening, and quality assessment.

Review Stages: Search, Screen, Quality Assess

FastText: an open-source, free, lightweight library that allows users to learn text representations and text classifiers. It works on standard, generic hardware. Models can later be reduced in size to even fit on mobile devices.

Review Stages: Screen, Synthesis

- 1. CADIMA (<u>https://www.cadima.info/</u>)
- 2. Colandr (<u>https://www.colandrcommunity.com/</u>)
- 3. Covidence (<u>https://www.covidence.org/</u>)
- 4. Distiller-SR <u>https://www.distillersr.com/products/distillersr-systematic-review-software</u>
- 5. EPPI-Reviewer (EPPI-Reviewer: systematic review software (ioe.ac.uk)
- 6. PICO Portal (PICO Portal)
- 7. Rayyan <u>https://www.rayyan.ai/</u>
- 8. SR Accelerator (Screenatron & Disputatron) <u>https://sr-accelerator.com/#/screenatron</u>
- (Full list of URLS available as Word document but only while Systematic Review Toolbox is migrating servers. After that better to use the live version; searching under "Screen")

Any Questions?

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