METHODOLOGY

Robust Statistical Stopping Criteria for Automated Screening in Systematic Reviews

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Abstract

Active learning for systematic review screening promises to reduce the human effort required to identify relevant documents for a systematic review. Machines and humans work together, with humans providing training data, and the machine optimising the documents that the humans screen. This enables the identification of all relevant documents after viewing only a fraction of the total documents. However, current approaches lack robust stopping criteria, so that reviewers do not know when they have seen all or a certain proportion of relevant documents. This means that such systems are hard to implement in live reviews. This paper introduces a workflow with robust and flexible statistical stopping criteria, which offer real work reductions on the basis of a given confidence level of reaching a given recall. The stopping criteria are shown on test datasets to achieve a reliable level of recall, while still providing work reductions of on average 17%. Other methods proposed previously are shown to provide inconsistent recall and work reductions across datasets.

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Keywords: Systematic Review; Machine Learning; Active Learning; Stopping Criteria

²⁷Background

Evidence synthesis technology is a rapidly emerging field that promises to change the practice of evidence synthesis work [1]. Interventions have been proposed at various points in order to reduce the human effort required to produce systematic reviews and other forms of evidence synthesis. A major strand of the literature works on screening: the identification of relevant documents in a set of documents whose relevance is uncertain [2]. This is a time consuming and repetitive task, and in a research environment with constrained resources and increasing amounts of ³⁴ ³⁵literature, this may limit the scope of the evidence synthesis projects undertaken. ³⁵ ³⁶Several papers have developed Active Learning (AL) approaches [3–7] to reduce the ³⁶ time required to screen documents. This paper sets out how current approaches are ³⁸ unsuitable in practice, and outlines and evaluates a small modification that would ³⁸ make AL systems ready for live reviews. ⁴⁰ Active learning is an iterative process where documents screened by humans are ⁴⁰ ⁴¹ used to train a machine learning model to predict the relevance of unseen papers [8]. ⁴¹ ⁴²The algorithm chooses which studies will next be screened by humans, often those ⁴² ⁴³ which are likely to be relevant or about which the model is uncertain, in order to ⁴³ ⁴⁴generate more labels to feed back to the machine. By prioritising those studies most ⁴⁴ 45 likely to be relevant, a human reviewer most often identifies all relevant studies - or 45

⁴⁶a given proportion of relevant studies (recall) - before having seen all the documents ⁴⁶

in the corpus. The proportion of documents not yet seen by the human when they ²reach the given recall threshold is referred to as the work saved. This represents the² ³proportion of documents that they do not have to screen, which they would have³ ⁴had to without machine learning. ⁵ Machine learning applications are often evaluated using sets of documents from ⁵ ⁶already completed systematic reviews for which inclusion or exclusion labels already ⁷exist. As all human labels are known a priori, it is possible to simulate the screening⁷ ⁸process, recording when a given recall target has been achieved. In live review ⁸ ⁹settings, however, recall remains unknown until all documents have been screened. ⁹ ¹⁰In order for work to really be saved, reviewers have to stop screening while uncertain ¹⁰ ¹¹about recall. This is particularly problematic in systematic reviews because low ¹¹ ¹²recall increases the risk of bias [9]. The lack of appropriate stopping criteria has ¹² ¹³therefore been identified as a research gap [10, 11], although some approaches have ¹³ ¹⁴been suggested. These have most commonly fallen into the following categories: • Sampling criteria: Reviewers estimate the number of relevant documents 15 16 by taking a random sample at the start of the process. They stop when this 16 17 number, or a given proportion of it, has been reached [15] 18 \bullet Heuristics: Reviewers stop when a given number of irrelevant articles are 18 19 seen in a row [6, 7]. 20 • Pragmatic criteria: Reviewers stop when they run out of time [3]. 21 - Novel automatic stopping criteria: Recent papers have proposed \mathtt{more}^{21} 22 complicated novel systems for automatically deciding when to stop screening²² [12-14]We review the first three classes of these methods in the following section and 24 ²⁵discuss their theoretical limitations. They are then tested on several previous sys-²⁵ ²⁶tematic review datasets. We demonstrate theoretically and with our experimental ²⁶ ²⁷results, that these three classes of methods can not deliver consistent levels of work²⁷ ²⁸savings or recall - particularly across different domains, or datasets with different ²⁸ ²⁹properties [2]. We also discuss the limitations of novel automatic stopping criteria, ²⁹ ³⁰which have all demonstrated promising results, but do not achieve a given level of ³⁰ ³¹recall in a reliable or reportable way. Without the reliable or reportable achieve-³¹ ³²ment of a desired level of recall, deployment of AL systems in live reviews remains³² ³³challenging. This study proposes a system for estimating the recall based on random sampling³⁴ ³⁵of remaining documents. We use a simple statistical method to iteratively test a³⁵ ³⁶null hypothesis that the recall achieved is less than a given target recall. If the ³⁶ ³⁷hypothesis can be rejected, we conclude that the recall target has been achieved ³⁷ ³⁸ with a given confidence level and screening can be stopped. This allows AL users³⁸ ³⁹to predefine a target in terms of uncertainty and recall, so that they can make³⁹ ⁴⁰transparent, easily communicable statements like "A recall of more than 95% was ⁴⁰ ⁴¹achieved with a confidence of 95%". ⁴² In the remainder of the paper, we first discuss in detail the shortcomings of existing ⁴² ⁴³stopping criteria. Then, we introduce our new criterion based on a hypergeometric ⁴³ ⁴⁴test. We evaluate our stopping criteria, and compare their performance with heuris-⁴⁴ ⁴⁵tic and sampling based criteria on real-world systematic review datasets on which ⁴⁵ ⁴⁶AL systems have previously been tested [12, 16–18].

¹Methods Review ²We start by explaining the sampling and heuristic based stopping criteria and dis-² ³cussing their methodological limitations. 4 5 ⁵Sampling Based Stopping Criteria ⁶The stopping criterion suggested by Shemilt et al. [15] involves establishing the⁶ ⁷Baseline Inclusion Rate (BIR), by taking a random sample at the beginning of ⁷ ⁸screening. The BIR is used to estimate the number of relevant documents in the⁸ ⁹whole dataset. Reviewers continue to screen until this number, or a proportion of ⁹ ¹⁰it corresponding to the desired level of recall, is reached. ¹¹ However, the estimation of the BIR fails to correctly take into account sampling ¹¹ ¹²uncertainty ^[1]. This uncertainty is crucial, as errors can have severe consequences. ¹² ¹³If the estimated number of relevant documents is even one unit above the true¹³ ¹⁴value, then no work savings will be achieved. If the number of relevant documents ¹⁴ ¹⁵is underestimated, then the recall achieved will be less than 100%. ¹⁶ The number of relevant documents drawn without replacement from a finite sam-¹⁶ ¹⁷ple of documents follows the hypergeometric distribution. Figure 1a shows the dis-¹⁷ ¹⁸tribution of the predicted number of documents after drawing 1,000 documents¹⁸ ¹⁹from a total of 20,000 documents, where 500 documents (2.5%) are relevant. The¹⁹ ²⁰left shaded portion of the graph shows all the cases where the recall will be less²⁰ ²¹than 95%. This occurs 35.91% of the time. The right shaded portion of the graph²¹ ²²shows the cases where the number of relevant documents is overestimated and no²² 23 work savings could be made. This occurs 46.24% of the time. In only 17.85% of 23 ²⁴cases can work savings be achieved while still achieving a recall of at least 95%. ²⁵ Figure 1b shows the probability distribution of these errors according to the sam-²⁵ 26 ple size. Even in very large samples both types of error remain frequent, and the risk 26 ²⁷of saving no work at all remains close to 50%. This shows how baseline estimation²⁷ ²⁸inevitably offers poor reliability, both in terms of recall and in work saved. 29 30 Heuristic Stopping Criteria ³¹Some studies give the example of heuristic stopping criteria based on drawing a given³¹ 32number of irrelevant articles in a row [6, 7]. We take this as a proxy for estimating 32 33that the proportion of documents remaining in the unseen documents is low, as the 33 34probability of observing 0 relevant documents in a given sample (analogous to a34 35set of consecutive irrelevant results) is inversely related to the number of relevant35 36documents in the population. We find this a promising intuition, but argue that 1)36 37it ignores uncertainty, as discussed in relation to the previous method; 2) it lacks a³⁷ 38 formal definition; and 3) it misunderstands the significance of a low proportion of 38 39relevant documents in estimating the recall. ⁴⁰[1] Although Shemilt et al. [15] employ a method to choose a sample size based on ⁴⁰ ⁴¹uncertainty, they fail to acknowledge the potential implications for recall of their ⁴¹ ⁴²choice. Their margin of error of 0.0025 and observed proportion of relevant studies ⁴² 43 of 0.0005 translate to estimates of $_{400\pm451}$ relevant results. To reduce the margin of 44 error to $\pm 5\%$ of estimated relevant studies, they would have had to screen $638,323^{44}$ 45 out of 804,919 results. See the notebook https://github.com/mcallaghan/rapid-screening/blob/ 46 master/analysis/bir_theory.ipynb that accompanies this paper for a detailed discussion 46

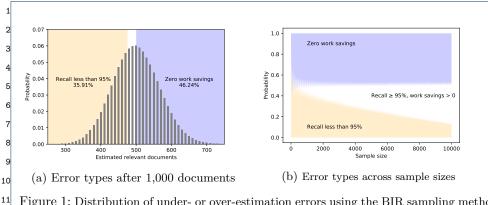


Figure 1: Distribution of under- or over-estimation errors using the BIR sampling method in a dataset of 20,000 documents of which 500 are relevant. Panel (a) shows the probag bility distribution of the estimated number of relevant documents after a sample of 1,000 documents. Panel (b) shows the probability of each type of error according to the sample

Figure 2 illustrates this third point. We show two scenarios with identical low 17 ¹⁸proportions of relevant documents observed in the unseen documents. In the top ¹⁸ ¹⁹figure, machine learning (ML) has performed well, and 74% of the screened docu-¹⁹ ²⁰ments were relevant. In the bottom figure, ML has performed less well, and only ²⁰ $^{21}26\%$ of the screened documents were relevant. In both cases, only 2% of unseen doc- 21 ²²uments are relevant, but 2% of a larger number means more relevant documents are ²² ²³missed. Recall is not simply a function of the relevance of unseen documents, but²³ ²⁴also of the number of unseen documents. This also means that where ML has per-²⁴ ²⁵ formed well (as in the top figure), a low proportion of relevant documents in those ²⁵ ²⁶ not yet checked is indicative of lower recall than where ML has performed less well. ²⁶ 27 Likewise, where the proportion of relevant documents in the whole corpus is low, a^{27} ²⁸ similarly low proportion of relevant documents is likely to be observed, even when ²⁸ ²⁹true recall is low. This shows us that even a perfect estimator of the relevance of ²⁹ 30 unseen documents is insufficient on its own to provide sufficient information about 30 ³¹when to stop screening. 32

³³Pragmatic stopping criteria

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³⁴Wallace et al. [4] develop a "simple, operational stopping criterion": stopping after ³⁴ ³⁵half the documents have been screened. Although the criterion worked in their ex-³⁵ ³⁶periment, it is unclear how this could be generalised, and its development depended ³⁶ 37 on knowledge of the true relevance values. Jonnalagadda and Petitti [6] note that 37 38"the reviewer can elect to end the process of classifying documents at any point, 38 ³⁹recognizing that stopping before reviewing all documents involves a trade-off of ³⁹ ⁴⁰lower recall for reduced workload", although clearly the reviewer lacks information ⁴⁰ ⁴¹about probable recall. 42

⁴³Novel automatic stopping criteria

⁴⁴Two examples come from the information retrieval literature. Di Nunzio [13]⁴⁴ ⁴⁵presents a novel automatic stopping criterion based on BM25, although recall re-⁴⁵ ⁴⁶ported is "often between 0.92 and 0.94 and consistently over 0.7". Yu and Menzies 46

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¹[12] also present a stopping criterion based on BM25 which allows the user to target ² a specific level of recall. However, reviewers are not given the opportunity to specify ³ a confidence level, and for two of the four datasets in which they tested their crite-³ ⁴ria, the median achieved recall at a stopping criteria targeting 95% recall was below ⁵ 95%. In each case, the reliability of the estimate is dependent on the performance ⁵ of the model.

Finally, Howard et al. [14] present a method to estimate recall based on the the number of irrelevant documents D observed in a list of documents since the δth^8 previous relevant document. They reason that this should follow the negative binomial distribution based on the proportion of remaining relevant documents p, and use this information to estimate \hat{p} , and with this, the total number of relevant articles and the estimated recall. However, since screening is a form of sampling without replacement, the negative hypergeometric distribution should be preferred to the negative binomial. Further the authors do not give sufficient information to software), nor an equation for \hat{p} . Additionally, the criterion requires a tuning patherizameter δ , which users may have insufficient information to set optimally. Lastly, their method does not quantify uncertainty, but can only claim that the method then the tends to result in a conservative estimate of recall (emphasis ours).

These last examples are promising developments, but fail to take into account the needs of live systematic reviews, where the reliability of and ease of communication about recall are paramount, and the results are independent of model performance.

In the following, we explain our own method, which provides clearly communicable estimates of recall, which manage uncertainty in a way robust to model performance.

26 26 Methods 27

28 A Statistical Stopping Criterion for Active Learning

In our screening setup, we start off with N_T documents that are potentially relevant. $_{29}$ $_{30}\rho_{tot}$ of these documents are actually relevant, but we don't know this value *a priori*. $_{31}$ As we screen relevant documents we include them, so ρ_{seen} represents the number $_{31}$ $_{32}$ of relevant documents screened, and τ recall is given by

$$\tau = \frac{\rho_{seen}}{\rho_{tot}} \tag{1}_{35}$$

We set a target recall τ_{tar} and a confidence level α . We want to keep screening³⁷ ³⁸until $\tau \geq \tau_{tar}$, and devise a hypothesis test to estimate whether this is the case with³⁸ ³⁹a given level of confidence. We do this based on interrupting the active-learning³⁹ ⁴⁰process and drawing a random sample from the remaining unseen documents. We⁴⁰ ⁴¹first describe this test, before showing how a variation on the test can be used to⁴¹ ⁴²decide when to begin drawing a random sample.

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⁴⁴Random Sampling

 $^{^{45}}$ At the start of the sample, N_{AL} is the number of documents seen during the active 46 learning process, and N_s is the number of documents remaining, so that

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 $H_0: K > K_{tar}$

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 $(8)^{46}$

1 1 2 2 $N_s = N_T - N_{AL}$ $(2)^3$ We refer to the number of relevant documents seen during active learning as ρ_{AL} , ⁶ and the number of remaining relevant documents as K. We do not know the value ⁶ ⁷of K but know that it is given by the total number of relevant documents minus⁷ ⁸the number of relevant documents seen during active learning. 9 10 10 $(3)^{11}$ 11 $K = \rho_{tot} - \rho_{AL}$ 12 With each draw, n is the number of documents drawn and k is the number of 13 ¹⁴relevant documents drawn. The number of relevant documents seen is updated by ¹⁴ ¹⁵adding the number of relevant documents seen since sampling began to the number ¹⁵ ¹⁶of relevant documents seen during active learning. 17 18 18 19 $(4)^{19}$ $\rho_{seen} = \rho_{ML} + k$ 20 We proceed to form a null hypothesis that the true value of recall is less than our²¹ 22 target recall: 23 24 24 $(5)^{25}$ 25 $H_0: \tau < \tau_{tar}$ Because we are sampling without replacement, we can use the hypergeometric²⁷ 28 distribution to find out the probability of observing k relevant documents in a^{28} ²⁹sample of n documents from a population of N documents of which K are relevant.²⁹ 30 ³⁰We know that k is distributed hypergeometrically: 31 31 32 32 33 $X \sim Hypergeometric(N, K, n)$ $(6)^{33}$ 34 We introduce a hypothetical value for K, which we call K_0 . This represents the³⁵ ³⁶minimum number of relevant documents remaining at the start of sampling com-³⁶ ³⁷patible with our null hypothesis that recall is below our target. 38 38 39 39 $K_0 = \lfloor \frac{\rho_{seen}}{\tau_{tar}} - \rho_{AL} + 1 \rfloor$ $(7)^{40}$ Our null hypothesis can thereby reformulated: the true number of relevant docu- $^{42}\,$ $^{43}\mathrm{ments}$ in the sample is greater than our hypothetical value. 44

¹ We test this by calculating the probability of observing k or fewer relevant from ¹
² the hypergeometric distribution given by K_{tar} , using the cumulative distribution
³ function.
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$p = P(X \le k), \text{ where } X \sim Hypergeometric(N_s, K_{tar}, n) $ (9)
⁸ Because $P(X \leq k)$, is strictly decreasing for values below K_{tar} , this gives the ⁸ maximum probability of observing k for all values of K compatible with our null ⁹ hypothesis.
If the maximum probability of obtaining our observed results given our null hy- 1 Pothesis is below our $1-\alpha$, then we can reject our null hypothesis and stop screening. We can report the likelihood that we achieve a recall below our target as being less 1 than $1-\alpha$.
Nonranaom sampung
In order to decide when to begin a random sample, we employ nonrandom sampling, where we treat previously screened documents as random samples. The distribution of relevant documents among previously screened documents is clearly not random, as documents predicted to be relevant are prioritised. It is reasonable to assume, though, that the density of relevant documents is greater among previously screened documents than among remaining unseen documents. This would make the following estimates conservative. After reviewing each document, N_{seen} documents have been screened, and N_{unseen} documents are yet to be seen. We treat $i=1\dots N_{seen}$ of the previously screened documents as separate random samples, and calculate p , using the method above, for each sample. We take the minimum across all samples p_{min} as the lowest probability that our null hypothesis is correct. If p_{min} is less than $1-\frac{\alpha}{2}$, we switch to random sampling. This method is an imprecise heuristic used to decide when to start random sampling. However, we also test its performance as a separate stopping criterion. We calculate p_{min} for the remaining documents as if we had not switched to random sampling and record the recall and work saved when $p_{min} < 1-\alpha$. We present these sampling and record the recall and work saved when $p_{min} < 1-\alpha$. We present these sampling and record the recall and work saved when $p_{min} < 1-\alpha$.
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Evaluation
We evaluate each of the criteria discussed on real world test data, operationalising
¹⁸ the heuristic stopping criteria with 50, 100, and 200 consecutive irrelevant records.
¹⁹ We run 100 iterations on each dataset and record the following measures.
• Actual Recall: The recall when the stopping criteria was met
• WS-SC: Work saved when the stopping criteria was met
• Additional Burden: the work saved when the criterion was triggered sub- tracted from the work saved when the recall target was actually achieved. For simplicity, we use a basic SVM model [19, 20], with 1-2 word n-grams taken
¹⁵ from the document abstracts used as input data. We start with random samples ¹⁶ of 200 documents (we do not employ Shemilt et al's methods for identifying the ¹⁸

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1		dataset	data_source	N	r_docs	р
2	0	UrinaryIncontinence	cohen	284	68	0.24
•	1	Antihistamines	cohen	287	90	0.31
3	2	Estrogens	cohen	349	79	0.23
4	3	NSAIDS	cohen	358	83	0.23
	4	OralHypoglycemics	cohen	475	135	0.28
5	5	Triptans	cohen	594	205	0.35
6	6	ADHD	cohen	803	83	0.10
	7	AtypicalAntipsychotics	cohen	1030	333	0.32
7	8	CalciumChannelBlockers	cohen	1103	257	0.23
8	9	ProtonPumpInhibitors	cohen	1210	227	0.19
0	10	SkeletalMuscleRelaxants	cohen	1348	30	0.02
9	11	COPD	copd_pb	1443	179	0.12
•	12	Kitchenham	fastread	1700	45	0.03
.0	13	Opiods	cohen	1769	43	0.02
.1	14	BetaBlockers	cohen	1872	270	0.14
	15	ACEInhibitors	cohen	2234	168	0.08
.2	16	Statins	cohen	2743	152	0.06
.3	17	ProtonBeam	copd_pb	4108	240	0.06
-	18	Radjenovic	fastread	5999	47	0.01
.4	19	Wahono	fastread	7002	62	0.01
.5	20	Hall	fastread	8911	104	0.01

Table 1: Dataset properties

"optimal" sample size, as we showed these in the methods section to be unhelpful). 19
20 Subsequently, we "screen", that is, we reveal the labels of, batches of the 20 docu20 ments with the highest predicted relevance scores, retraining the model after each
21 batch. The batch size could be adjusted to retrain more or less frequently, which is
22 a trade-off between computational time spent training, and the speed at which the
23 algorithm can "learn". Each criterion is evaluated after each document is "screened".
24 For our criteria, we set the target recall value to 95% and the confidence level to
25 95%.

The systematic review datasets used for testing are described in table 1. We
tuse the seminal collection of systematic reviews used to develop machine learning
applications for document screening by Aaron Cohen and co-authors in 2006 [16],
along with the widely used Proton Beam [17] and COPD [18] datasets, and computer
science datasets used to test FASTREAD [12]. Testing on datasets with different
properties and from different domains is key to establishing criteria appropriate for
seneral use. Choosing as broad as possible data also prevents us from being able
to "tune" our machine learning approach in ways that may work well for specific
datasets but not generalise well. Work savings, even maximum work savings are
therefore below the state of the art recorded for each of these datasets. In this way
we can show how well the criteria perform even when the model performs badly.

All computational steps required to reproduce this analysis are documented online at https://github.com/mcallaghan/rapid-screening.

⁴⁰Results

⁴¹Figure 4 shows the actual recall and work savings achieved when each stopping ⁴² criteria has been satisfied. For comparison, we also include the results that would ⁴² ⁴³ have been achieved with *a priori* knowledge of the data, that is, the work saved when ⁴³ the 95% recall target was actually reached. In a live systematic review, reviewers ⁴⁴ would never know when this had been reached, but these are the work savings most ⁴⁵ often reported in machine learning for systematic review screening studies.

¹ Both the random sampling and the pseudorandom sampling criteria achieve the ¹ ²target threshold of 95% in more than 95% of cases. In fact, the pseudorandom² ³sampling criterion outperforms the random sampling criterion with respect to both³ ⁴recall and work savings, saving a mean of 17% of the work compared to 15%, ⁴ ⁵and missing the target in only 0.95% compared to 3.29% of cases. In theory, the ⁵ ⁶pseudorandom sampling criteria is conservative if the assumption holds that doc-⁶ ⁷uments chosen by machine learning are not less likely to be relevant than those ⁷ ⁸chosen at random. Based on our experiments, this assumption seems reasonable, ⁸ ⁹and accounts for the higher recall. Because the pseudorandom sampling criterion ⁹ ¹⁰can flexibly choose its sample, whereas the random criterion has to wait for a ran-¹⁰ ¹¹dom sample to be triggered, the criterion is also triggered earlier, as it can make ¹¹ ¹²use of more data. This accounts for the higher work savings. 13 The baseline sampling criteria (Figure 4c) misses the 95% recall target in $39.67\%^{13}$ ¹⁴of cases, while the most common work saving is 0%. This is in line with our ex-¹⁴ ¹⁵pectations that, due to random sampling error, the expected number of documents ¹⁵ ¹⁶will often be over-estimated or under-estimated, resulting in zero work savings or ¹⁶ ¹⁷poor recall. ¹⁸ The Heuristic stopping criteria, both for 50 consecutive irrelevant results (Figure¹⁸ ¹⁹4d - IH50), and for 200 irrelevant results (Figure 4e) also perform unreliably. Al-¹⁹ ²⁰though the mean work saved for IH50 is 41%, the target is missed in 39% of cases. ²⁰ ²¹The cases below the horizontal grey line indicate instances where work has been ²¹ ²²saved at the expense of achieving the recall target. In figure 5 we rescale the x axis, calling it additional burden, which is simply the 23 ²⁴work saved when the criterion is triggered minus the work saved when the recall²⁴ ²⁵target was actually achieved. This measure indicates whether the stopping criterion²⁵ ²⁶was triggered too early (negative values), or too late (positive values). The figure ²⁶ ²⁷directly highlights the tradeoffs involved in deciding when to stop screening: For our ²⁷ ²⁸criteria, there is mostly a small additional burden which comes with the necessity to ²⁸ ²⁹make sure the desired recall target has been reached and reject the null hypothesis²⁹ ³⁰that this has not been the case. For the other criteria, there are many cases in which ³⁰ ³¹additional burden is negative, i.e. the criterion has been triggered too early. In these³¹ ³²cases, however, the desired recall is hardly ever reached. To help explain the different work savings that were observed in our experiments,³³ ³⁴we show the distribution of work savings from our pseudorandom criterion for each ³⁴ ³⁵dataset in figure 6. In general, higher work savings are possible when the total num-³⁵ ³⁶ber of documents is larger. However, in datasets with a low proportion of relevant ³⁶ ³⁷documents, many documents need to be screened to achieve a high confidence that ³⁷ ³⁸there are only few relevant documents remaining in the unseen ones. Therefore, ³⁸ ³⁹smaller work savings are possible. Figure 7 shows the recall and the probability of the null hypothesis for the best⁴⁰ ⁴¹performing iteration of four datasets. Although the 95% recall target is achieved ⁴¹ ⁴²very quickly in the Radjenovic dataset, the null hypothesis cannot be excluded until ⁴² ⁴³much later. This is because the dataset has only 47 relevant documents out of a⁴³ ⁴⁴population of 5,999. After the 95% recall target was achieved, 45 out of 47 relevant ⁴⁴ ⁴⁵documents had been seen and 5,029 documents remained. The null hypothesis was ⁴⁵ ⁴⁶therefore that 3 or more of these 5,029 documents were relevant, which requires a lot ⁴⁶

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¹of evidence to disprove. The burden of proof was smaller in the case of the Proton ¹ ²Beam dataset: at the point that the 95% recall threshold was reached, the null² ³hypothesis to disprove was that a minimum of 13 out of 3,369 remaining documents³ ⁴were relevant.

⁵ The Statins and Triptans datasets show how the criterion performs when the⁵ ⁶machine learning model has performed poorly in predicting relevant results. In each⁶ ⁷case, 95% recall is achieved with close to 20% of documents remaining. With fewer⁷ ⁸documents remaining, it takes fewer screening decisions to rule out the possibility⁸ 9that the number of relevant documents left is incompatible with the achievement9 ¹⁰of the recall target.

12 Discussion

13Our results show that it is possible to use machine learning to achieve a given level 13 140f recall with a given level of confidence. The tradeoff for achieving recall reliably 14 15is that the work saving achieved is less than the maximum possible work saving.15 16However, for large datasets with a significant proportion of relevant documents, 16 17the additional effort required to satisfy the criterion will be small compared to the 17 18 work saved by using machine learning. This makes the approach well suited to broad 18 19topics with lots of literature. In other words, it is precisely where machine learning 19 20 will be most useful that the additional effort will be small.

21 Different use cases for machine learning enhanced screening may also carry dif-21 22 ferent requirements for recall, or different tolerances for uncertainty. These can be 22 23 flexibly accommodated within our stopping criterion. Importantly, the ability to 23 $_{24}$ make probabilistic statements about the chance of achieving a given recall target $_{24}$ ₂₅makes it possible to clearly communicate the implications of using machine learning₂₅ ₂₆enhanced screening to readers and reviewers who are not machine learning special-₂₆ ₂₇ists. This is extremely important in live systematic reviews.

Our criteria have the further advantage that they are independent of the choice or 28 performance of the machine learning model. If a model performs badly at discerning relevant from irrelevant results, the only consequence will be that the work saved will be low. With other criteria this may result in poor recall. When using machine $\underset{32}{\text{learning for screening, poor recall can result in biased results, while low work savings}_{32}$ represent no loss to the reviewer as compared to not using machine learning.

So far, systematic review standards have no way of accommodating screening with $_{_{\mathbf{2}A}}$ machine learning. We hope that the reliability and clarity of reporting offered by our stopping criteria make them suitable for incorporation into standards, so that machine learning for systematic review screening can fulfil its promise of reducing workload and making more ambitious reviews tractable. 38 38

39 Conclusion ⁴⁰This paper demonstrates the unsuitability of existing stopping criteria for machine ⁴⁰ ⁴¹learning approaches to document screening, and proposes a simple method that ⁴¹ ⁴²delivers reliable recall, independent of machine learning approach or model perfor-⁴² ⁴³mance. Our robust statistical stopping criteria allow users to easily communicate ⁴³ ⁴⁴the implications of their use of machine learning, making machine learning enhanced ⁴⁴ ⁴⁵screening ready for live reviews. 46 Callaghan and Müller-Hansen Page 11 of 18

	ics approval applicable.	1			
³ Cor	sent for publication	3			
4Not	applicable.	4			
5 Av a	ilability of data and materials	5			
	computational steps required to reproduce this analysis are documented online at	6			
ntt 7	ps://github.com/mcallaghan/rapid-screening.	7			
	npeting interests	8			
I he	e authors declare that they have no competing interests.	9			
Aut	hor's contributions	10			
14 has	designed the research and conducted the experiments. FMH contributed to the development of the statistical is for the stopping criterion. Both authors wrote and edited the manuscript.				
		11			
	nowledgements « Callaghan is supported by a PhD scholarship from the Heinrich Böll Foundation. Finn Müller-Hansen	12			
	acknowledges funding from the German Federal Ministry of Research and Education within the Strategic Scenario				
14Ana	lysis (START) project (grant reference: 03EK3046B).	14			
¹⁵ Aut	hor details	15			
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	sdam, Germany.	18			
¹⁹ Ref	erences	19			
20 1.	Westgate MJ, Haddaway NR, Cheng SH, McIntosh EJ, Marshall C, Lindenmayer DB. Software support for	20			
21 2.	environmental evidence synthesis. Nature Ecology & Evolution. 2018;2:588–590. O'Mara-Eves A, Thomas J, McNaught J, Miwa M, Ananiadou S. Using text mining for study identification in	21			
	systematic reviews: A systematic review of current approaches. Systematic Reviews. 2015;4(1):1–22.	22			
	Miwa M, Thomas J, O'Mara-Eves A, Ananiadou S. Reducing systematic review workload through certainty-based screening. Journal of Biomedical Informatics. 2014;51:242–253. Available from:				
23	http://dx.doi.org/10.1016/j.jbi.2014.06.005.	23			
24 4.	Wallace BC, Trikalinos TA, Lau J, Brodley C, Schmid CH. Semi-automated screening of biomedical citations for systematic reviews. BMC Bioinformatics. 2010;11.	24			
25 5.	Wallace BC, Small K, Brodley CE, Trikalinos TA. Active learning for biomedical citation screening.	25			
26	2010;(July):173.	26			
27 6.	Jonnalagadda S, Petitti D. A new iterative method to reduce workload in systematic review process. International Journal of Computational Biology and Drug Design. 2013;6(1/2):5.	27			
28 7.	Przybyła P, Brockmeier AJ, Kontonatsios G, Le Pogam MA, McNaught J, von Elm E, et al. Prioritising	28			
29	references for systematic reviews with RobotAnalyst: A user study. Research Synthesis Methods. 2018;9(3):470–488.	29			
	Settles B. Active Learning Literature Survey. University of Wisonsin-Madison; 2009.	30			
9. 31	Lefebvre C, Glanville J, Briscoe S, Littlewood A, Marshall C, Metzendorf MI, et al. Cochrane Handbook for Systematic Reviews of Interventions. In: Higgins J, Green S, editors. Cochrane Handbook for Systematic	31			
32	Reviews of Interventions. version 5. ed. The Cochrane Collaboration; 2011. Available from:	32			
	www.handbook.cochrane.org. Bannach-Brown A, Przybyła P, Thomas J, Rice ASC, Ananiadou S, Liao J, et al. Machine learning algorithms				
34	for systematic review: reducing workload in a preclinical review of animal studies and reducing human screening	3 ₂₁			
	error. Systematic Reviews. 2019;8(1):1–12.				
3511.	tools in research synthesis. Systematic Reviews. 2019;8(1):1-10.	35			
³⁶ 12.		36			
37 13.	Applications. 2019;120:57–71. Available from: https://doi.org/10.1016/j.eswa.2018.11.021. Di Nunzio GM. A study of an automatic stopping strategy for technologically assisted medical reviews. In: Pas	37 si			
38	G, Piwowarski B, Azzopardi L, Hanbury A, editors. Lecture Notes in Computer Science (including subseries	38			
39	Lecture Notes in Artificial Intelligence and Lecture Notes in Bioinformatics). vol. 10772 LNCS. Cham: Springer International Publishing; 2018. p. 672–677.	39			
4014.	Howard BE, Phillips J, Tandon A, Maharana A, Elmore R, Mav D, et al. SWIFT-Active Screener : Accelerated	40			
41	document screening through active learning and integrated recall estimation. Environment International. 2020;138(April 2019):105623. Available from: https://doi.org/10.1016/j.envint.2020.105623.	41			
42 ¹⁵ .	Shemilt I, Simon A, Hollands GJ, Marteau TM, Ogilvie D, O'Mara-Eves A, et al. Pinpointing needles in giant	42			
43	haystacks: Use of text mining to reduce impractical screening workload in extremely large scoping reviews. Research Synthesis Methods. 2014;5(1):31–49.	43			
44 ¹⁶ .		44			
45	automated citation classification. Journal of the American Medical Informatics Association.	45			
40 17.	2006;13(2):206–219. Terasawa T, Dvorak T, Ip S, Raman G, Lau J, Trikalinos T. Review Annals of Internal Medicine Systematic				
40	Review: Charged-Particle Radiation Therapy for Cancer. Annals of Internal Medicine. 2009:(5).	46			

¹ 18.	Castaldi PJ, Cho MH, Cohn M, Langerman F, Moran S, Tarragona N, et al. The COPD genetic association compendium: A comprehensive online database of COPD genetic associations. Human Molecular Genetics.	1 2
³ 19.	2009;19(3):526–534. Cortes C, Vapnik V. Support-Vector Networks. In: Machine Learning; 1995. p. 273–297.	3
₄ 20.	Pedregosa F, Varoquaux G, Gramfort A, Michel V, Thirion B, Grisel O, et al. Scikit-learn: Machine Learning Python Fabian. Journal of Machine Learning Research. 2011;12:2825–2830.	in ₄
5	Tython Fabian. Journal of Machine Learning Research. 2011,12.2023–2030.	5
6		6
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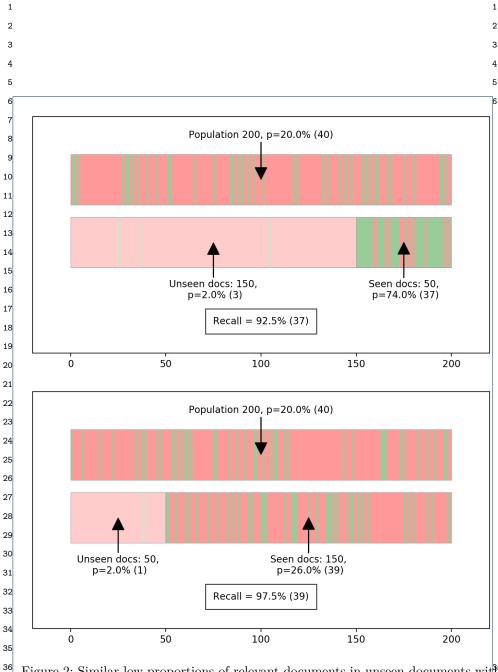
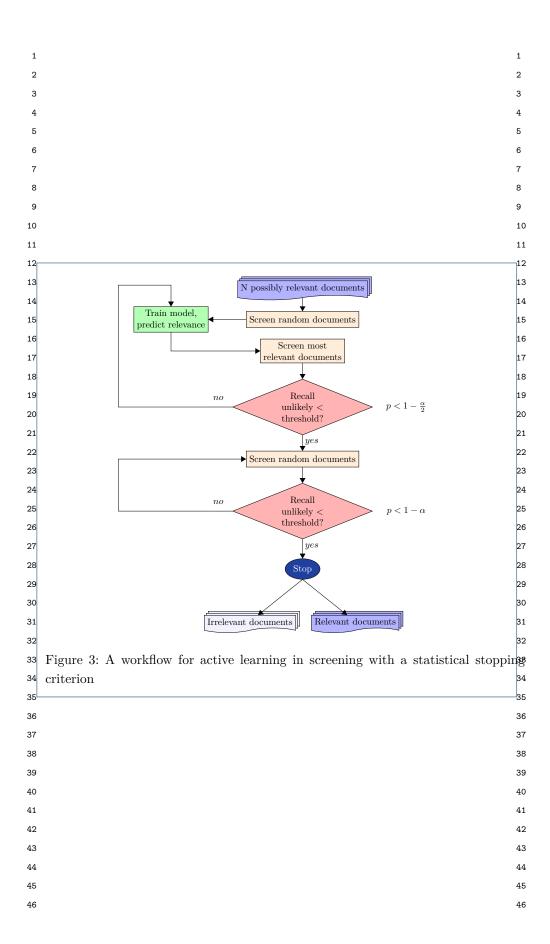


Figure 2: Similar low proportions of relevant documents in unseen documents with different consequences for recall. The top bar shows a random distribution of relevant documents (green) and irrelevant documents (red) at a given proportion of relevance. The bottom bar shows distributions of relevant and irrelevant documents in hypothetical sets of seen (right) and unseen (left - transparent) documents.

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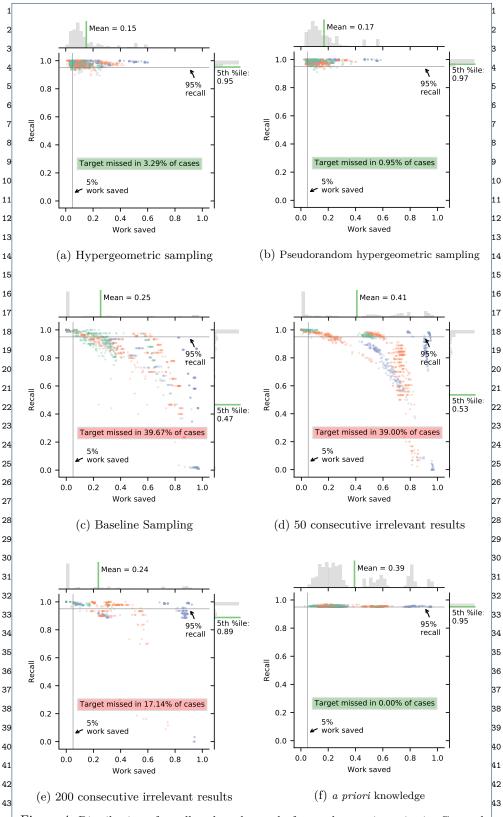


Figure 4: Distribution of recall and work saved after each stopping criteria. Green does show results for datasets with less than 1,000 documents, orange dots show datasets with 1,000 - 2,000 documents, and blue dots show datasets with more than 2,000 documents.

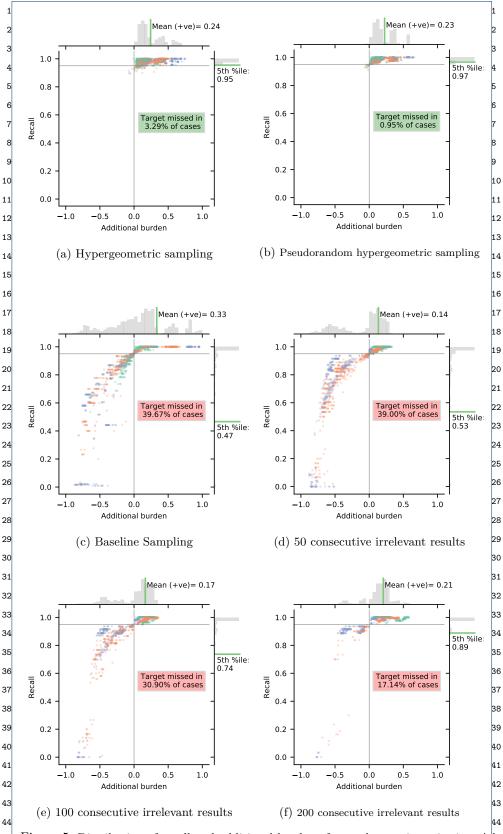


Figure 5: Distribution of recall and additional burden after each stopping criterion. Adjutional burden is the work saved when the criterion was triggered minus the work saved when the target was reached. Coloring of data points as in Fig. 4.

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