Comparison of a full systematic review versus a rapid review approaches to assess a newborn screening test for tyrosinemia type 1.

Sian Taylor-Phillips, Associate Professor*[1]
Julia Geppert, Research Associate[1]
Chris Stinton, Research Fellow[1]
Karoline Freeman, Research Fellow[1]
Sam Johnson, Academic Support Librarian[2]
Hannah Fraser, Research Associate[1]
Paul Sutcliffe, Associate Professor[1]
Aileen Clarke, Professor of Public Health and Health Services Research[1]

[1] Warwick Medical School, University of Warwick, Coventry, CV4 7AL
[2] Warwick Library, University of Warwick, Coventry, CV4 7AL

*Corresponding author: S.Taylor-Phillips@warwick.ac.uk

Abstract

Background

Rapid reviews are increasingly used to replace or complement systematic reviews to support evidence based decision-making. Not enough is known about how this expedited process affects results.

Objectives

To assess the difference between rapid and systematic review approaches for a case study of test accuracy of succinylacetone for detecting tyrosinemia type 1.

Methods

Two reviewers conducted a rapid then a systematic review. The rapid review involved narrower searches, a single reviewer with 20% checking for screening the titles/abstracts and data extraction, and quality assessment using an unchanged QUADAS-2. Two reviewers performed the systematic review with a tailored QUADAS-2. Post-hoc analysis examined rapid reviewing with just a single reviewer (basic rapid review).

Results

The rapid and systematic reviews identified the same 10 papers, although one paper was only identified in the rapid review through checking included studies' references. 2176 fewer title/abstracts and 129 fewer full-texts were screened during the rapid review. The unadjusted QUADAS-2 generated more 'unclear' ratings [29/70 (41.4%) compared to 16/70 (22.9%)], and fewer 'high' ratings [22/70 (31.4%) compared to 42/70 (60.0%)] than the systematic review using the adjusted QUADAS-2. A rapid review using a single reviewer would have missed up to four eligible studies during screening of titles/abstracts, and contained important inaccuracies in data extraction detected through use of a second reviewer.

Conclusions

Rapid reviews with 20% checking by a second reviewer appears to be an appropriate tool for policy-makers to expeditiously assess evidence. Single reviewer rapid reviews have higher risks of important inaccuracies and omissions.

Keywords: Systematic review, Rapid review, Research methods, Evidence Based Practice, Literature searching

1. Introduction

Rapid reviews (also referred to as "rapid evidence reviews" or "rapid evidence assessments", REAs) have received increased interest and recognition across Europe, North America, and Australasia, where there are demands for expedited assessment of the latest evidence to facilitate health policy decisions (Hailey 2007; Watt et al 2008; Ganann et al 2010; PHE 2015; Tsertsvadze et al 2015). As yet, there is no standard definition of or approach to REAs (Featherstone et al 2015; Polisena et al 2015). Broadly, they offer a streamlined alternative to the traditional systematic review process (Khangura et al 2012; NICE 2014). The following key differences between rapid and systematic reviews have been reported: REAs are produced more quickly, and are generally thought to require continuous engagement with the commissioning organisation throughout the document development process. They generally focus on a descriptive approach to synthesis in contrast to, for example, meta-analysis. Inferences from an REA tend to be more cautious and/or provisional (Watt et al 2008; PHE 2015).

The effect of using REA rather than systematic review methods is currently unclear. Edwards et al (2002) Edwards et al (2002)found that the average increase in the total number of randomised trials identified by using a second reviewer was 9 per cent, but that this ranged across pairs from 0 per cent (both reviewers individually identified all trials) to 32 per cent (a second reviewer identified an additional seven trials) (Edwards et al 2002). Further they found that single reviewers missed on average 8 per cent of eligible reports (range 0 to 24 per cent), whereas pairs of reviewers missed few or none (range 0 to 1 per cent). Watt and colleagues have suggested that exhaustive data searching may not greatly impact final conclusions and recommendations of a review (Watt et al 2008). In contrast, Helmer compared MEDLINE searching versus extended searching which included specialist databases, hand searching, reference list review, and personal communication with experts and found that systematic searching increased the number of studies found and decreased bias (Helmer et al 2001). Helmer suggests that the likelihood of extended searching impacting the number of items retrieved may depend on the content area and whether content is likely to be found in mainstream databases (Helmer et al 2001).

Some reviews have found little difference in the conclusions of an REA when they are compared with those of full health technology assessment reports or systematic reviews on the same topic (Watt et al 2008; Watt et al 2008; Ganann et al 2010; Scott and Harstall 2012). However, authors of these reviews report that REAs have a narrower scope, less depth, and

provide less detail in their recommendations. A scoping review of rapid reviews, found that in 84 papers, 50 different approaches to reviewing were used (Tricco et al 2015). There was a lack of reporting of duration of review in 73%, reference checking in 40%, limitations by language in 34%, and number of reviewers in 29% of rapid reviews. In addition, few REAs discussed limitations associated with or bias introduced by, the streamlining process. According to Scott & Harstall (Scott and Harstall 2012) the degree to which the individual components of a review influence its results is unclear. It is also unclear what the minimum essential elements might be for accurate and reliable results. Comparisons to date have been largely between multiple reviews conducted by different research teams, which may be biased due to potential systematic differences between teams of researchers who conduct rapid and systematic reviews.

The aim of this research was to compare the findings of a traditional SR with REA conducted by the same research team. We used a case study of a review of test accuracy of the succinylacetone (SUAC) newborn blood spot screening test for tyrosinemia type 1 (TYR1).

2. Methods

The protocol for the rapid review is registered at PROSPERO (registration number CRD42015026912) and was developed in close collaboration with the UK National Screening Committee, the commissioner. The rapid and systematic review approaches employed different search strategies, and methods for screening of titles and abstracts, assessment of full text articles, extraction, and quality appraisal. The same two reviewers conducted both reviews, first the REA and then the full review with no changes of staffing. No task other than quality appraisal was repeated twice by any individual reviewer, i.e. the data extraction from the REAs was also used in the SR. The methods used for the REA and systematic review approaches are shown in Table 1.

+Table 1+

2.1 Identification and selection of studies

Both the REA and SR searches were conducted in Medline, Pre-Medline, Embase, and the Cochrane Library. The REA also included Web of Science (WoS) Core Collection, while the SR included WoS All Databases. The search strategy for the REA was narrower, for example

the search terms for the condition were related to tyrosinemia type 1 only, whereas in the SR the terms were broadened to ‘tyrosinemia type 1’ or ‘inborn errors of metabolism’. In the REA approaches search terms for the screening test were related to ‘succinylacetone’ only, whereas in the systematic review this was broadened to include ‘tandem mass spectrometry’ and ‘newborn screening’ (see Supplement 1 and Supplement 2 for complete search strategies). Searches for both reviews were performed on 26th January.

2.2 Eligibility criteria

English language journal articles which investigated screening for TYR1 by tandem mass spectrometry (TMS) analysis of SUAC from dried blood spots in newborn infants were included in all reviews. The reference standard for both reviews was urine testing for SUAC and/or subsequent clinical detection of TYR1. Outcomes included test accuracy from cross-sectional test accuracy, case-control, or cohort studies. Exclusion criteria were non-human studies, papers not available in the English language, letters, editorials and communications, grey literature, conference abstracts, and studies published before 2004 (the year the first study of TMS measurement of SUAC in dried blood spots was published).

2.3 Screening and data extraction

Screening of titles and abstracts of all retrieved records, and subsequently of full texts was conducted by one reviewer for the REA, with a second reviewer checking a random 20% full independent screening by two reviewers for the SR. Data extraction was performed by a single reviewer, with 20% and 100% checked by a second reviewer for the rapid and systematic reviews, respectively.

2.4 Quality appraisal

For the REA, quality appraisal was undertaken by one reviewer using QUADAS-2 which, contrary to the guidelines for use of the tool, was not tailored to the question. 20% of assessments were independently assessed by a second reviewer. For the full systematic review, risk of bias was assessed using the QUADAS-2 which was tailored to the research as recommended, and assessment was undertaken independently by two reviewers for all studies. Tailoring of the QUADAS-2 tool included adding a topic-specific signalling question and defining appropriate reference standards and cut-offs for participant exclusions as well as guidance on how many positive signalling questions are required for an overall positive rating in terms of bias and applicability concerns. The systematic review was conducted after

the REA, so that the reviewers undertaking untailored QUADAS-2 could not benefit from their experience using a tailored QUADAS-2.

2.5 Data summary and synthesis

Descriptive statistics were calculated for the differences between the REA and systematic review in the number of papers found in each search, the number of full-texts accessed, and the number of papers included. Narrative comparisons were made between the data extracted and the quality appraisal scores between the two reviews.

2.6 Post hoc analyses

The approach to REA used here is arguably more comprehensive than some other REA processes. To explore the impact of this we conducted a post hoc comparison of the effect that using an REA with fewer resources would have had on the findings. This approach comprised a single reviewer, a search limited to electronic databases only, no checking of references, and the use of the unadjusted QUADAS-2 (see table 1 for full details). We refer to this approach as a 'basic REA', to contrast it our more comprehensive REA (which we refer to as an 'enhanced REA'). Single reviewer data were available from reviewer A and reviewer B, giving us two 'case studies' of this approach.

3. Results

3.1 Searching and screening of titles/abstracts

Figure 1 provides the PRISMA flow diagram for the REA and systematic review searches. We identified 310 unique records in each of the basic and enhanced REAs, and 1274 in the systematic review. After screening titles and abstracts, 19 (6%) were retained for the basic REA by reviewer A and 25 (8%) by reviewer B, 19 (6%) for the enhanced REA, and 76 (6%) for the systematic review. In the basic REA nine (47%) of the full text articles were included after assessment against inclusion criteria by reviewer A and 5 (20%) by reviewer B. In the enhanced REA, nine (47%) of the full text articles were include with one additional paper (Lund et al 2012) identified by reference checking of a previous review (Bazian 2014). In the systematic review, 10 (13%) of 76 full-text articles were included in the review. The same 10 articles were included after screening the titles/abstracts in each of the enhanced REA and systematic review approaches. See Supplement 3 for a list of excluded studies with reasons.

+Figure 1+

In the basic REA, there was no difference in the number of titles/abstracts that were screened by reviewers A and B. However, reviewer B examined 13 more full texts than reviewer A. There were no differences between either of the basic REAs and the enhanced REA in terms of the number of titles/abstracts that were screened. Compared to each of the basic REAs, the enhanced REA resulted in the examination of 0 (reviewer A) or 13 (reviewer B) additional full texts. The systematic review resulted in an extra 964 titles/abstracts being screened compared to each of the basic REAs, and an extra 964 titles/abstracts by reviewer A and 1212 by reviewer B (as reviewer A only screened 20% of the titles/abstracts in the rapid review) compared to the enhanced REA. The systematic review resulted in the assessment of an additional 57 or 51 full texts compared to reviewers A and B, respectively, in the basic REA, and the examination of an additional 57 full texts compared to the enhanced REA. The additional study identified by reference checking in the enhanced REA was an observational cohort describing screening of 504,049 newborns in Denmark, the Faroe Islands and Greenland (Lund et al 2012). It was missed by the REA electronic searches as this article did not contain the search terms for 'tyrosinemia' in title or abstract, only broader terms like 'inborn errors of metabolism' or 'IEM'.

3.2 Quality assessment

Using unadjusted QUADAS-2 in the basic and enhanced REAs resulted in more 'unclear' and fewer 'high risk' ratings for both risk of bias and applicability concerns than the adjusted QUADAS-2 used in the full systematic review. Using an adjusted QUADAS-2 tool with guidance notes reduced the total number of 'unclear' ratings from 26 of 63 (41%) in the basic REA by reviewer A and 10 of 35 (29%) by reviewer B, and 29 of 70 (41.4%) in the enhanced REA to 16 of 70 (22.9%) in the systematic review. The number of 'high' ratings increased from 21 of 63 (33%) in the basic REA by reviewer A and 12 of 35 (34%) by reviewer B, and 22 of 70 (31.4%) in the enhanced REA to 42 of 70 (60.0%) in the systematic review. This difference is particularly apparent in the reference standard domain, where in the rapid review 1/10 studies were rated high risk of bias and 0/10 had high applicability concerns, whereas in the systematic review 7/10 were rated high risk of bias and 7/10 had high applicability concerns. Results by domain for the basic REA, enhanced REA, and SR are given in Table 2.

+Table 2+

3.3 Data extraction and review findings

Differences were found within the basic REAs (reviewer A versus reviewer B), and between the basic REAs and the enhanced REA/SR approaches, in relation to data extracted and review findings. Using the basic REA methods (which did not include screening reference lists) would have resulted in missing one relevant paper (Lund et al 2012). The use of a single reviewer would have affected our results at every stage of the review process. At the screening the titles/abstracts stage, a single reviewer would not have missed any records (if it was reviewer A) but one record if reviewer B was the only reviewer. Full text assessment by a single reviewer would have either missed no records (if it was reviewer A) or 4 eligible records (if it was reviewer B). Most importantly, using single data extraction would have resulted in extracting and calculating inappropriate sensitivity, specificity and negative predictive values for six prospective studies (if it was reviewer A). These studies reported sensitivity and specificity, but it was inappropriate to use due to lack of follow up of negative cases with any reference standard test. The inclusion of a second reviewer (20% for the REA or 100% for the SR) prevented these omissions and errors occurring.

We found no differences between the enhanced REA and SR approaches to data extracted or overall review findings. Using enhanced REA and SR approaches, ten studies were identified that provided data on test accuracy of the SUAC newborn blood spot screening test for TYR1. Five studies reported results from prospective newborn screening programmes (Sander et al 2006; la Marca et al 2011; Morrissey et al 2011; Lund et al 2012; Zytkovicz et al 2013), and four reported results from case-control studies used stored DBS samples from confirmed TYR1 patients and healthy controls (Allard et al 2004; la Marca et al 2008; Turgeon et al 2008; Dhillon et al 2011; Metz et al 2012). It was not possible to calculate sensitivity, specificity, or negative predictive values due to lack of follow up of screen negative results. Positive predictive values (PPV) from four prospective screening studies using SUAC as primary marker was 100% (6 true positive cases out of 717,501 people screened) in three studies (Sander et al 2006; Lund et al 2012; Zytkovicz et al 2013), and 67% (2 true positive cases and 1 false positive case out of ~500,000 people screened) in one study (Morrissey et al 2011). PPV could not be calculated in two studies (la Marca et al 2011; Metz et al 2012).

4. Discussion

4.1 Summary of findings

We conducted a case study to compare the impact of reducing or eliminating some of the steps of a full SR by undertaking a REA. We investigated the number of relevant studies missed and the frequency of errors in a review of a newborn screening test for TYR1. This comparison suggests that for this particular topic, a REA with 20% checking by a second reviewer and checking references of included studies identified the same number of relevant studies as did a broader SR search but with a reduced amount of effort, time, and resources.

A key difference between the SR and REA approaches was the use of an adjusted (SR) and unadjusted (basic and enhanced REA) QUADAS-2 tool. This has a substantial impact on the interpretation of the review findings. Using an unadjusted QUADAS-2 resulted in risks of bias/applicability concerns that could not be qualified in a large proportion of the studies (29 – 41% of total ratings). Conversely, tailoring the QUADAS-2 tool in the SR resulted in clearer estimates of risk of bias/applicability. In particular, in the reference standard domain there were more papers in the SR with high risk of bias and applicability concerns. Overall, 60% of total ratings were for high risks of bias/applicability concerns, and only 23% were for unclear risks of bias/applicability concerns. Therefore, the adjusted QUADAS-2 (used in the SR) improved our understanding of the risks of bias/applicability concerns within the studies, and indicated that, due to the large proportion of studies with potential biases, we should exercise caution in the interpretation of results.

Post hoc analysis indicated that a less thorough REA (that searched fewer sources, and used a single reviewer only) may have generated substantial errors that would have impacted on test accuracy estimates produced.

4.2 Strengths and limitations

Previous comparisons of REA and SR have typically looked at reviews that were conducted by different review teams. In these instances we cannot know if disparities in the results are caused by variation in the methodologies used or difference in the abilities and experience of the reviewers. In our study the same two reviewers conducted the rapid review, then the systematic review analysing an identical review question. This reduced the confounders introduced by variation between reviewers in comparisons between rapid and systematic reviewing methods. The reviewers both had significant systematic reviewing experience at the beginning of the study, and some experience of test accuracy reviewing. Our post hoc

analyses indicated that using a single reviewer showed significant potential errors could arise from this. This result is likely to be generalisable to others because of the level of experience of the reviewers, and because the main error (reporting sensitivity and specificity directly from the studies without checking for follow up of negative results to an adequate reference standard) was also made in a previous review on this topic (Bazian 2014), which used a single reviewer method.

Our review has a number of limitations. First, our systematic review was not carried out in a blinded manner; as the REA was conducted first, the reviewers had prior knowledge of what evidence was available and had made an assessment about the studies and their quality before the SR began. We cannot be sure that the results of the SR would have been exactly the same if the SR had been conducted independent of the REA. Second, the decision to examine the impact of a more basic review that did not include a second reviewer (and to compare reviewer A versus reviewer B) was made after the reviews had been completed. The results of these elements of our review should be considered to be exploratory. More generally, a key limitation to this approach is that this is only a single case study. Caution must be exercised in making generalisations to other reviewers, or topics. For this topic both the condition (TYR1) and the test (TMS measurement of SUAC) are rare. Refining search terms and screening of titles/abstracts are both relatively straight forward in this topic area so the findings demonstrate the difference between REAs and full systematic reviews very clearly. However it may not be appropriate to generalise to more complex topics or to those with a larger evidence base. Finally, the included studies did not include adequate information to populate the 2x2 tables, so we could not calculate whether omission of the paper by (Lund et al 2012), which would have been missed in a more minimal search would have changed the study conclusions.

4.3 Comparison to other studies

Our research is broadly consistent with prior studies which have compared REA and SR approaches. Similar to Edwards et al (2002), we found evidence that single reviewers missed more eligible papers than pairs of reviewers; in our study this was dependent upon which reviewer was being evaluated. This suggests that individual reviewer characteristics are an important contributor to review accuracy when a single reviewer only is used. Further, while there were clear differences between our REA and SR in terms of the scope and depth of searches, like Ganann et al (2010); Scott and Harstall (2012), and (Watt et al 2008; Watt et al 2008) these did not affect our overall conclusions.

4.4 Implications for policy and practice

The appropriateness of deciding to use a rapid rather than systematic review may depend on purpose and context. REA and SR are not equal or interchangeable, rather they often serve different purposes. The purpose of an REA is not to provide a definitive answer to the effectiveness of a particular intervention but, for example, to provide sufficient information on which to decide next steps, including whether, and what, further work is required before a decision can be made, e.g. if different types of evidence are required (primary research, cost-effectiveness studies) or if a full systematic review is warranted. Depending on the findings of the REA, a policy makers might see a justification for a systematic review when a body of evidence (of sufficient volume and quantity) is identified that could be explored using meta-analysis to better characterise, for example, treatment effects, or a need to more thoroughly investigate issues of bias is presented. While a SR would answer questions about where evidence is lacking, it would do so at the price of time and resources with no guarantees that it would provide useful additional information that an REA would not. Indeed, there is evidence of this in our own review; in the context of a 3-yearly update review developed in consultation with stakeholders and experts, to guide policy-makers on whether this topic merits further investigation and future research questions, the conclusions from the REA were not altered by the SR. This research thus provides some evidence that rapid reviewing (with 20% checking by a second reviewer) may be an effective method of triage, to determine whether there is a need for a full systematic review. REAs may also be of value in the identification of focused topics requiring SRs and the provision of an estimate of the volume of literature which might be expected and some of the quality issues likely to be encountered. However, even for the rare disease and uncommon test evaluated here, use of a full adjusted QUADAS-2 increased the estimates of bias in comparison to a rapid unadjusted QUADAS-2, indicating that REA is not a replacement for a full systematic review, particularly if the rapid review recommends implementation of a new treatment, test or screening programme. This review also provides some evidence that rapid reviewing with a single reviewer only may not be an effective approach to reviewing the evidence, and that there may be significant benefit from 20% checking by a second reviewer.

REA and SR are not the only approaches to reviewing for policy makers. An alternative is a scoping review. Typically, scoping reviews do not seek to answer specific research questions using narrow criteria, nor do they include formal appraisal of the quality of literature, or a synthesis of evidence (Arksey and O'Malley 2005; Peters et al 2015). A range of uses of scoping reviews have been reported, including mapping of key concepts,

identifying the extent evidence, determining if systematic reviews are justified, summarising and disseminating the findings of research, and identifying gaps in the literature (Arksey and O'Malley 2005). This approach may be more appealing to policy makers than REAs as it requires fewer resources and is less likely to 'miss' relevant literature. Like REAs, scoping reviews currently lack uniform terminology, methods, and reporting standards (Colquhoun et al 2014).

5. Conclusions

For this rare disease with a single uncommonly used test, comparison of SR with REA methods did not affect our findings in terms of search yield or data extraction, but tailoring the QUADAS-2 tool in the SR resulted in higher estimates of risk of bias. A full SR approach dramatically extended the work load of searching and screening of titles/abstracts. REA involving only one reviewer with no checking by a second reviewer may have resulted in missed studies, and significant inaccuracies in data extraction. We consider that REAs are likely to have their place in the armamentarium for those providing information to policy makers and working closely with commissioners. A thorough preceding rapid review may point to the need for a further judgement as to where, how, when and in what circumstances a full systematic review might be the better option.

Competing interests

The authors have no conflicts of interest.

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Abbreviations

CRD	Centre for Reviews and Dissemination
NICE	National Institute for Health and Care Excellence

REA	Rapid evidence assessments
SR	Systematic review
SUAC	Succinylacetone
TMS	Tandem mass spectrometry
TYR1	Tyrosinemia type 1
UK NSC	UK National Screening Committee
WoS	Web of Science

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