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Pruning and prioritising: a case study of a pragmatic method for managing a rapid systematic review with limited resources

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Abstract

Full systematic reviews are time and resource heavy. We describe a method successfully used to produce a rapid review of yoga for health and wellbeing, with limited resources, using mapping methods. Inclusion and exclusion criteria were developed a priori and refined post hoc, with the review team blind to the study results to minimise the introduction of bias. This method allowed the review to be tailored to make use of the best available evidence and the health topics of most relevance to the commissioners, and to enable the evidence base to be disseminated to practitioners in a timely fashion.

Introduction

Well-conducted systematic reviews are a cornerstone of evidence based medicine and are used to inform policy and practice. The scientific rigour of the process can, at times, be at odds with the needs of the commissioners in terms of time, resources and political input (Rotstein and Laupacis 2004). This tension between rigour and relevance has led to the development of different approaches which reduce both the budget and the time needed to complete a review. Some such reviews are termed "rapid reviews" and employ systematic review methods, but diverge from standard methodologies in one or more stages of the review process. This may include using only one reviewer, reducing the number of databases searched, not performing quality assessment of studies, or using less comprehensive approaches to data extraction and synthesis (Ganann et al. 2010; Harker and Kleijnen 2012).

Any divergence from full systematic review methodologies comes at a price in terms of quality and robustness. For example, using only a single author can introduce errors in data extraction and study selection, whilst not performing quality assessment can lead to important issues of bias and quality in the evidence base being missed. As such, methods should be tailored to minimise the impact on the reliability of the results in light of the research question being answered. For example, if the rapid review is being conducted to provide an overview of the research available and a gap analysis,

synthesis of the results may not be as important as data extraction of study variables that enable a detailed gap analysis.

Alongside rapid reviews, other review types that employ systematic methods include mapping reviews and scoping reviews. The definitions of these three vary across the literature (Grant and Booth 2009) and the terms are often used interchangeably. Indeed a recent scoping review of scoping review methodologies included both mapping and scoping reviews (Pham et al. 2014). Both tend to address broad topic areas, aim to identify gaps in the evidence, facilitate the definition of topics and subtopics where a full systematic review is both viable and useful, and gauge the time and resources needed to complete these reviews (Grant & Booth 2009). Pham et al. quantified certain characteristics amongst the reviews they included: electronic literature searches were usual, though additional search methods were not; study selection criteria could be very broad, including most study and publication types, but selection was rarely checked by another reviewer; quality assessment was not usual; presentation was usually tabular, with less than a third presenting graphical results; and narrative summaries were presented, but meta-analyses were never done (Pham 2014). One key concept that separates the two review types is that scoping reviews may also include a summary of research findings and a consultation exercise (Arksey and O'Malley 2005), which rarely if ever appears in descriptions of mapping reviews.

In the normal scheme of a systematic review, mapping and scoping methodologies are used as well as clinical and political inputs to define the research question and scope. One key feature of systematic reviews is the a priori statement of the research question and methods in the study protocol, so these processes usually occur before the protocol is written. This minimises the potential for bias to be introduced either intentionally or unwittingly. Post hoc amendments to reviews (amendments made after the study has commenced) are generally thought acceptable where a legitimate reason is given for the change (Centre for Reviews and Dissemination 2009). And it could be argued that the point at which the change is made also impacts the potential for any such amendments to introduce bias. For example, a change in the analysis plan to include an additional analysis, made in response to impressions about the availability of evidence formed by reviewers during initial study selection (e.g. which outcome measure has been used most by researchers) may seem less problematic than a change made after the primary analysis has been performed. At the study selection stage where abstracts are consulted, the reviewer is unlikely to be forming a reliable impression of the direction of the evidence, especially where more than one outcome is being reviewed. However if an analysis is only planned and performed after the a priori analysis results are known, it could be that this was done in response to unfavourable or undesirable results in the original analysis in an attempt to mitigate any negative impact, or to replace the primary analysis altogether.

As reported in Harker and Kleijnen (2012), 47% of the rapid reviews they considered did not have a clear research or clinical question, which may indicate that little or no scoping had been performed by the commissioners of the review. When funding and time for scoping are limited, and where a question or topic is broad, it can be difficult to see clearly where available research best lends itself to evidence review and synthesis, and where the information needs of the commissioners might best be met. Whilst adaptations to systematic review methodologies to date have mainly focussed on economies in individual components of the methods used, there are some examples where economies are made across the whole review process, from scoping to completion, by selecting a different methodology for the review. Examples include the "two-stage review" process described in the EPPI Centre's methods guidelines (Anon 2010), a very similar method described in Gough et al.'s book (Gough et al. 2012) and the methods for "rapid evidence assessments" described by the UK government's Social Research Service (Civil Service 2013). In the EPPI Centre method, studies are mapped according to well defined codes and the scope of the review narrowed with regard to the results of the map. Scoping and reviewing are performed from a single iteration of searching and sifting to select appropriate literature. In the latter example (Civil Service 2013), narrowing of the review focus may take place once synthesis of study results has commenced. In this case study, we describe and discuss the strengths and weaknesses of a novel approach to shortening the timescales of a review from scoping to completion by conducting scoping/ mapping and reviewing in a three-phase process necessitating only a single iteration of searching and sifting.

The project

Our research team was commissioned by British Wheel of Yoga (BWY) in 2010 to produce an evidence summary that aimed to draw together research evidence relating to yoga in adults aged 18 to 65 years , in a broad range of common health conditions in a single resource. No limits were specified as to which health conditions to include, or which study types to focus on, though only the previous 10 years were to be searched. The output was to be suitable for a health professional and non-health-professional audience.

The commissioners required the report to be limited to approximately 40 studies in order that the report be concise for readability and to fit within their own budgetary constraints; a total of 33 days spread over several months (if necessary) were funded by the commissioners. The problem for the review team was how to deliver these requirements within the budget and time limits but without knowing the scope of the literature, without introducing bias, and whilst maintaining a broad scope to avoid excluding key topics of interest to the commissioners.

Methodological innovation within the review

The research report was to be based on the methods described by Wilkinson et al (2008) in that a set of initial inclusion and exclusion criteria would be applied to retrieved titles and abstracts, and the review team would then work with experts (who were also the commissioners) to refine the set of studies for inclusion. This constitutes a post hoc amendment to the selection criteria as set out in the study protocol. Bias could be introduced in that criteria could be altered so that their application resulted in the exclusion of studies reporting negative results, and thus bias the overall evidence base towards positive results. This could occur intentionally, or unintentionally.

We sought to reduce the potential for the introduction of such bias and sought to formalise the process of article selection to increase transparency and reproducibility, so that readers could judge for themselves whether the process and resulting criteria were likely to introduce bias. We formalised the process by mapping the available literature in two stages during the review. We were then able to tailor the focus of the review to the topics where there was available evidence and which were of most interest to the commissioners, whilst keeping within the limited resources and timescales. We sought to minimise the introduction of bias when refining the selection criteria by defining key selection criteria a priori (Table 1) in the protocol, as described in Wilkinson et al. (2008), and purposefully not extracting the results of studies during the mapping stage, in an attempt to blind those involved in refining criteria to the direction of effect. The process we undertook can be broken down in to a three-phase process as follows (see Figure 1):

Phase 1: First phase of mapping the topic

1a) Initial selection criteria were defined with input from the commissioners (Table 1)

1b) A broad, systematic and comprehensive search of the topic was performed

1c) Studies were selected according to the initial selection criteria (Table 1).

1d) Studies selected were tagged in the reference management software according to key characteristics (kept to a minimum to avoid unnecessary work) ascertained from the title or abstract. In this case study, this included only the health topic (e.g. asthma, cancer, arthritis etc.) and the study design (e.g. systematic review, randomised controlled trial [RCT] etc.). A matrix of the studies by the key characteristics was constructed to provide an overview of the literature (Table 2).

1e) The review team and commissioners chose key characteristics as selection criteria to select the studies of most relevance to their needs. In this case study, this entailed selecting the

health topics of greatest relevance to the commissioners and where there was evidence, and the study designs of highest quality according to the hierarchy of evidence (National Institute for Health and Clinical Excellence 2012). The characteristics that were selected are listed in Table 1.

Phase 2: Second phase of mapping the topic

2a) A preliminary data extraction was carried out in which key study characteristics were extracted from included abstracts (or full text where necessary). Crucially, study results were not extracted so that users were blind to the direction of effect (within the confines of the data extraction database). The fields that were extracted in this case study are listed in Table 1.

2b) A list of potential further selection criteria were developed by the review team with reference to the preliminary data extraction and presented for discussion to the commissioners. The list of potential selection criteria for this case study are listed in Table 1.

2c The commissioners and the review team discussed and selected the inclusion criteria that both improved relevance to the commissioners, and resulted in the review remaining within resource constraints. The list of final selection criteria for this case study are listed in Table 1.

Phase 3: Main review (rapid or systematic)

3a) In this case study, we performed a rapid review. The rapid review was "rapid" in that study selection and data extraction were conducted by one reviewer, and formal data synthesis was not performed. The preliminary data extraction was expanded into full data extraction to minimise duplication of effort. However, it should also be possible to conduct a full systematic review with any form of synthesis (e.g. narrative, meta-analysis).

3b) The main output was a report to the commissioners which comprised structured abstracts for each study, with a commentary on study quality and conclusions. A further important output of this particular review was a set of research recommendations, and identification of gaps in the literature.

The flow of studies through the review is represented in a modified PRISMA diagram in Figure 2.

Discussion

In this case study, we have described a pragmatic method for managing a review within limited resources, by incorporating the mapping/scoping stage of a review within the review process, and devising a method of developing selection criteria post hoc in a blinded fashion to limit the

introduction of bias. Mapping the review comprised two phases – one to map two key characteristics of the literature (in this case, health condition and study type) and narrow the potential includable studies according to the interests of the commissioners and availability of high quality evidence, and a further phase to data extract details about additional key characteristics of the studies and further refine the selection criteria.

As discussed in the introduction, any deviation from standard systematic review methods has the potential to impact on the quality of the results and the impartiality of the analysis and interpretation of the review. In comparison to other review methodologies that could have been employed for this review, the method we have outlined here has both strengths and weaknesses. We have compared the method against seven other review methodologies in Table 3. The key factors considered in this Table include: whether a separate scoping review was first performed; whether comprehensive literature searches were conducted; whether the selection criteria were designated a priori; whether critical appraisal was conducted; whether two reviewers were involved in the review process; what type of synthesis was performed. The final column in Table 3 lists some of the key strengths and weaknesses of each method for easy comparison. As can be seen from this Table, each method entails its own unique trade-off between rigour and relevance. At one end of the spectrum, we have systematic reviews which can reach the highest standards of rigour, but are resource-heavy and as such can lack relevance to the commissioners e.g. where they have inadequate funds. All other methods sit somewhere below these high standards in no empirically determined order, or serve a somewhat different objective, e.g. scoping reviews do not intend to be comprehensive.

In comparison to a full systematic review, our method sacrifices some of the rigour; the refinement of some inclusion criteria was performed post hoc and could introduce bias. This sacrifices buys the method greater relevance/utility to the commissioners, in that the review can be produced in a timely fashion with minimal resource use and tailored to their interests. An alternative to this method would have been to conduct a rapid review without the mapping phases. Rapid reviews tend to trade off different aspects of rigour, for example, completeness of searching, or data extraction validation, to achieve the same reduction in demand on resources. However, this would have necessitated a separate scoping or mapping stage to narrow the review a priori. In this case study, a rapid review methodology was employed to gain yet further economies, but more robust full systematic review methods could have been employed in these later stages. The methods for writing the UK government's Social Research Service rapid evidence assessments (Civil Service 2013) are divided into "a priori" reviews and "iterative" reviews on the basis of the detail in the protocol. Our method employs aspects of both the a priori and iterative approaches, but has the advantage over the iterative approach of being predictable in terms of demands on resources, and the advantage over the "a priori" approach of not involving narrowing of the review focus at the synthesis stage, where it could be argued there is a greater likelihood of bias being introduced.

A similar process to our three-phase approach is described in the EPPI Centre's methods guide (2010) and is termed a "two-stage" review, where studies are first coded according to key characteristics, and the scope narrowed on the basis of this map. Another very similar method is described in Gough et al.'s book (2012), where an initial map is used as the basis for discussion with commissioners in narrowing the review question. The method we describe here was developed independently, and differs in some aspects. Rather than coding all key characteristics in all potentially includable studies, we selected two key characteristics (health condition and study type) and used these to short-list studies for further consideration, according to the interests of the commissioners and availability of evidence. This equates roughly to the first stage described by the EPPI Centre. To then capture the level of detail about studies as is achieved by the EPPI Centre method through their use of a "standard and well defined set of keywords", our method performs a preliminary data extraction on the remaining studies. This is likely to take longer than simply tagging studies. However, our approach recoups some of this time in that a) we used only two tags per study, and completed the initial sift very quickly, b) we did not have to invest time in the identification, definition and redefinition of a standard set of keywords, but rather we generate these keywords inductively through the process of data extraction and c) fewer studies are involved in this greater level of coding. In addition, the flexibility of and detail captured during data extraction may reduce the chances of missing important details that can be lost in abstraction when using pre-defined key word tags. And finally d) the time spent on data extraction is in part recouped as this data extraction will be expanded into a full data extraction for those studies that are eventually included in the review.

Both our approach and the EPPI Centre and Gough et al. approaches have differential demands on resources, and reviewers should consider which methods to use with reference to the specifics of their own review. For example, our approach may have advantages where the review team and commissioners are new to the topic and do not have the required breadth of familiarity to identify and code the heterogeneity of key study characteristics such as population, setting, outcomes and intervention from the outset. Conversely, where commissioners or reviewers are highly familiar with the field, coding data from the outset may represent an economy as studies will only have to be visited once, and data extraction performed only on included studies. Our method may also be more useful where there are initially a very large number of includable studies; in cases where the EPPI Centre method would require coding of all these studies, and the number of codes is large, our approach would allow an initial triaging of studies so that only those of most interest are considered in detail.

The method we have outlined here carries certain limitations, and will certainly not be a suitable approach in all situations. However, when it is used carefully and appropriately it can offer a number of advantages. We believe this method will be most useful where budgets are tight and contracts impose restraints in terms of resources or timescales. It could be especially useful if a commissioner's scope is initially unclear or overoptimistic. The review team will then be able to maximise the

strengths of the available research, balancing this against the utility of the finished review to the end user, and the requirements of the commissioners. We expect that these types of conditions are most likely to occur where reviews are commissioned by bodies with limited resources, such as charities, or where the commissioners are interested in a broad topic, but are unsure where best to focus their efforts, such as in policy units.

There are important limitations to this method and instances where its application would be inappropriate. The unbiased choice of post hoc selection criteria relies on blinding to study results being achieved by not extracting this data. However, this blinding could be subverted in cases where those involved in choosing the selection criteria are already familiar with the literature, or refer to the original journal articles when refining the selection criteria. This could lead to the conscious or subconscious introduction of bias in the choice of selection criteria. This may be a particular problem where commissioners have a vested interest in the results of the review, for example, pharmaceutical manufacturers. Even in our case study this could have been problematic, though fortunately the commissioners were not themselves familiar with the literature. Several practical considerations may also determine whether this method will be appropriate. Where the scope of a review is clear and focussed, such as for reviews of intervention A versus intervention B, this method would confer no advantages and would be unnecessary. Where searches lead to very large numbers of citations being retrieved and carried forward to the second phase of mapping the topic, there is a risk that many studies will needlessly undergo preliminary data extraction, only to be later rejected. In this case, the approach may still be useful, but instead of performing a preliminary data extraction on all studies, the reviewers could use characteristics presented in existing reviews to help inform the choice of inclusion and exclusion criteria, though this may unduly limit which characteristics are considered. The resource implications of a review can be ascertained at protocol stage by running a search in one key bibliographic database e.g. Medline, and methods selected appropriately. Finally, where commissioners are not able or willing to contribute to the process, this method may still be viable if the commissioners are happy for the review team to finalise the inclusion and exclusion criteria. In this case, it may also be possible to substitute the commissioners with other independent reviewers.

Conclusion

All adaptations of the systematic review process involve a trade-off between quality and resource use, both of which are aspects of rigour and relevance. Our research report for the BWY successfully used post hoc refinement of inclusion and exclusion criteria to select studies for inclusion that we believe avoided major bias and maintained a degree of scientific rigour comparable to other rapid review methods. We were able to satisfy the requirements of the commissioners by producing a review with high relevance to them and with a broad scope, whilst keeping within the budget and resource constraints of the review and maximising the strengths of the available research. We believe our

three-phase approach has wider applications as a pragmatic method of mapping and reviewing the literature within a limited timescale where the scope is broad or unknown.

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Figure 1. Flow chart of the review methods

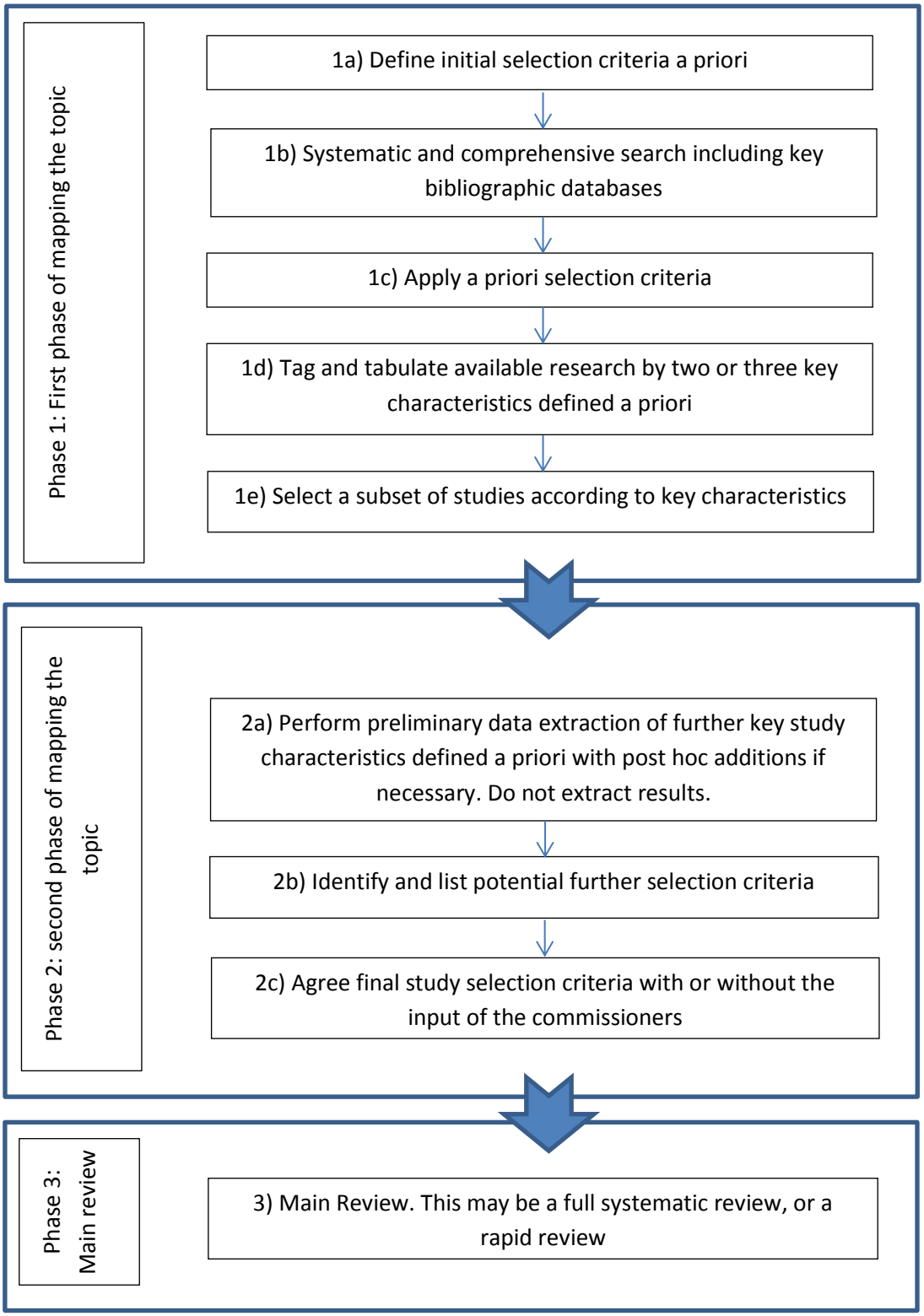

Figure 2. Modified PRISMA flow diagram representing flow of studies through the review

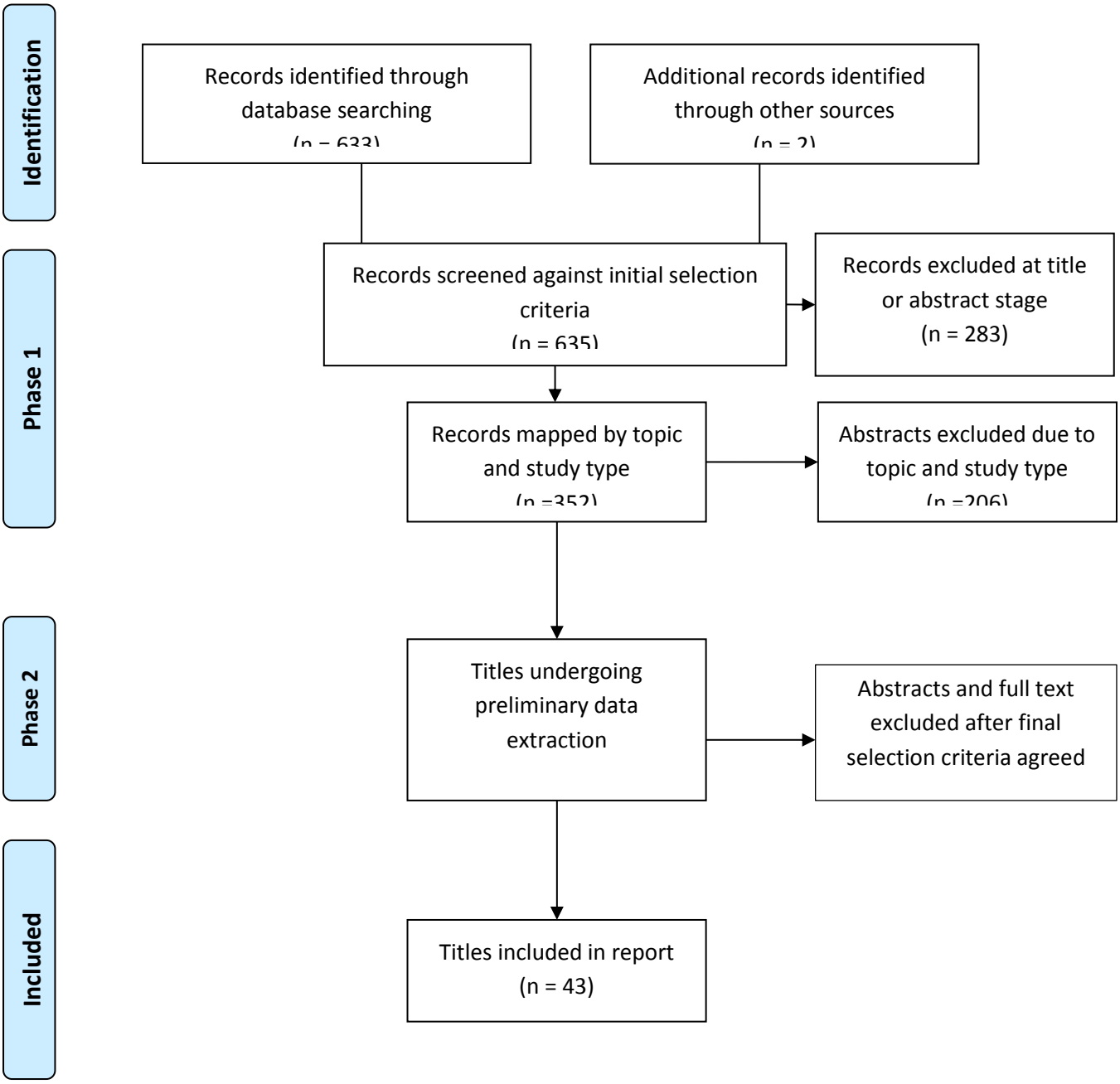

Table 1 Development of selection criteria.

Initial selection criteria	Selection criteria following first mapping phase	Data extraction fields used in second mapping phase	Final* selection criteria following second mapping phase and preliminary data extraction
Population: Adults aged 18 to 65 years old. **Intervention:** Any yoga intervention including yoga meditation, relaxation, postures and breathing techniques. **Comparator:** Any. **Outcomes:** Any health outcome. **Study design:** Any. Excluded laboratory-based studies.	**Health Topics** • Arthritis • Asthma • Back pain • Cancer • Cardiovascular disease • Depression and Anxiety • Diabetes • Menopause • Pregnancy • Healthy adults **Study designs** • SR • RCT • Qualitative • Mixed methods	• Author, date of publication • Country of origin • Study design • Condition or population • Outcomes measured • Elements of yoga practised • N of studies included (within SRs) • N of participants (primary research) • Intervention description • Scope of SR (within SRs)	**Exclude** • Qualitative • studies of meditation-based stress reduction (MBSR) • studies where yoga is delivered with other interventions • dissertations if not published in peer-reviewed journal • studies of specific groups eg archers • studies of specific (non health) outcomes eg glossolalia • studies using breathing devices • studies judged to have sub-therapeutic exposure to yoga • interventions not replicable in UK e.g. Yoga camps. If systematic review only lists one RCT, the RCT itself is selected instead. Exclude RCTs if included in a systematic review, or if only a protocol
RCT, randomised controlled trial; SR, systematic review; N, number			

Table 2. First phase of mapping the topic. Matrix number of studies of each study design, by health topic. Note: some studies appear in more than one category.

	Total	Systematic review	Review	RCT	Qualitative	Other	Unclear	Mixed methods	Research report
Healthy	67	1		11	23	29	2	1	
Depression/anxiety	38	11		10		9	4	2	2
Stress	37	4		7	3	19	3		1
Cancer	35	6	2	11	2	6	6	1	1
Cardiac	32	4	2	10	2	9	5		
Back pain	22	6		9	1	1	5		
Menopause	19	5	2	6	1	3	2		
Mental health	16	1		7		3	3	1	1
Diabetes type 2	16	2	2	2	3	5	2		
Arthritis	11	5		2		2	2		
Asthma	10	3	1	5			1		
Pregnancy	10	2		3	1	4			
Carpal tunnel syndrome	7	7							
Pain	6	1				3	2		
BMI	6					2		3	1
Substance abuse	6	1		1	3		1		
Multiple sclerosis	6	1	1	3		1			
Epilepsy	6	3	1	1		1			
Abuse recovery	5				1		2	1	1
Eating disorders	5			1		2		1	1
Fatigue	5	2		1		2			
Fibromyalgia	5	1		2		2			
Stroke	4	2					2		
ADHD	3	2				1			
COPD	3			1			2		
HIV/AIDS	3			1		2			
Insomnia	3	1				2			
Athletes	3					1	1		
IBS	2	1			1				
Palliative care	2		1		1				
	2			1			1		
Musicians	2			1		1			
Dry eyes	2						1		
Headache	1	1							
Hyperkyphosis	1			1					
Lung health	1					1			
Lymphoedema	1						1		
Managers	1				1				
Migraine	1			1					

Musculoskeletal	1					3			
Osteoporosis	1					1			
Pancreatitis	1	1							
Piriformitis	1					1			
Post partum	1				1				
Preterm labour	1						1		
Prisoners	1					1			
PTSD	1			1					
Sexual function	1						1		
Smoking cessation	1			1 (ongoing)					
Trauma	1		1						
RCT, randomised controlled trial; PTSD, post-traumatic stress disorder; BMI, body mass index; COPD, chronic obstructive pulmonary disease.									

Table 3. **Comparison of review methodologies.** Scores indicate a generalisation, and there may be exceptions in all cases.

Review type	**Separate scoping review?**	**Searches comprehensive?**	**a priori selection criteria?**	**CA performed?***	**Two reviewers?****	**Usual method of synthesis***	**Key strengths and weaknesses**
Systematic review (including qualitative, quantitative, meta analysis and mixed methods)*	Y	Y	Y	Y	Y	Usually comprehensive and integrative, tabular with narrative, sometimes graphical	**Weaknesses:** Time consuming and expensive **Strengths:** Comprehensive search strategy - most/all literature should be found; minimises sources of bias ; allows evidence based conclusions to be drawn
Scoping review*	NA	Y or tailored to resources	Y	N	Opt	Tabular with some narrative	**Weaknesses:** If searches not comprehensive, may miss studies; if no CA, precludes robust analysis of results; searches and sifting may need to be conducted for both scoping and reviewing stages **Strengths:** Quick, cheap; if searches comprehensive, most/all literature should be found; selection bias avoided; ensures subsequent review, reviews or primary research is viable and relevant
Mapping review*	Opt	Y or tailored to resources	Y	N	Opt	Tabular or graphical	
Rapid review*	Opt	Y or tailored to resources	Y	Opt	Opt	Tabular and narrative	**Weaknesses:** as for scoping and mapping review **Strengths:** As for mapping and scoping reviews plus if CA conducted, allows evidence based conclusions to be drawn
Rapid evidence assessment – a priori(Civil Service 2013)	U	Y or tailored to resources	Y	Y	Opt	May be iterative and time consuming. Usually comprehensive and	**Weaknesses:** If searches not comprehensive, may miss studies; May be time consuming in comparison to a rapid review or three-phase review; Bias may be introduced in

Review type	Separate scoping review?	Searches comprehensive?	a priori selection criteria?	CA performed?*	Two reviewers?**	Usual method of synthesis*	Key strengths and weaknesses
Rapid evidence assessment – iterative(Civil Service 2013)	U	Y or tailored to resources	Opt	Y	Opt	integrative. May be tabular and/or narrative	iterative processes at any stage in review, including synthesis. **Strengths:** If searches comprehensive, most/all literature should be found; Iterative synthesis allows focus to be tailored according to availability of evidence and areas of most interest
EPPI Centre two-stage review(2010)	N	Y	Partial	Y	Y	Usually comprehensive and integrative, tabular with narrative	**Weaknesses:** May be time consuming in comparison to a rapid review or three-phase review **Strengths:** Comprehensive search strategy - most/all literature should be found; Removes need for separate scoping stage to save time and resources; Ensures review focus is viable and relevant
Three-phase review	N	Y or tailored to resources	Partial	Opt	Opt	Any of the above	**Weaknesses:** As for mapping, scoping and rapid reviews, plus bias may be introduced in focussing selection criteria **Strengths:** As for rapid reviews, plus: Removes need for separate scoping stage to save time and resources; Ensures review focus is viable and relevant; Only one search and sift required across scoping and reviewing stages; Subsequent review phase can be rapid or comprehensive, according to the resources available

* some definitions adapted from Grant and Booth (2009)
** to check or independently perform sifting, data extraction and/or CA
CA, critical appraisal; Y, yes; N, no; Opt, optional; NA, not applicable; U, unclear