

Stakeholder involvement in systematic and rapid reviews of health services research

Protocol version 2

1. Background

Description of full systematic reviews (SR) and rapid reviews (RR)

According to the Cochrane handbook (1), a traditional systematic review is a review that “attempts to collate all empirical evidence that fits pre-specified eligibility criteria in order to answer a specific research question. It uses explicit, systematic methods that are selected with a view to minimizing bias, thus providing more reliable findings from which conclusions can be drawn and decisions made”. While systematic reviews are considered to be the gold standard in knowledge synthesis, they are not without their limitations. For example, they usually require between 6 months and 2 years to complete and often focus on a narrow clinical question or set of questions. (2)

The key characteristics of a systematic review are (a) a clearly stated set of objectives with an explicit, reproducible methodology; (b) a systematic search that attempts to identify all studies that would meet the eligibility criteria; (c) an assessment of the validity of the findings of the included studies (e.g., assessment of risk of bias and confidence in cumulative estimates); and (d) systematic presentation, and synthesis, of the characteristics and findings of the included studies. (3) Systematic reviews should build on a protocol that describes the rationale, hypothesis, and planned methods of the review; few reviews report whether a protocol exists. Detailed, well-described protocols can facilitate the understanding and appraisal of the review methods, as well as the detection of modifications to methods and selective reporting in completed reviews (4).

Full systematic reviews (SR) and rapid reviews (RR) are two types of systematic reviews. They are often distinguished by the shorter time frame needed to produce a RR final report as well as by its less complex research topic (5): “Rapid reviews streamline traditional systematic review methods in order to synthesize evidence within a shortened timeframe” (6). As shown by Watt et al. RR did not have to be inherently inferior to SR despite “axiomatic differences” (7). He found that their conclusions did not differ much for similar research questions. However, SRs included methodologically more broader information, e.g. on economic evaluations or health service impact as well as more in-depth results, e.g. on clinical outcomes (8).

For SRs comprehensive guidance exists, e.g. based on the Cochrane Handbook (1). On the other hand, there is no standardized methodology for RRs and needs for at minimum increased transparency in reporting have been identified (5, 7).

R Rs were often considered to target specific policy questions or to be tailored to a mandate or to the intended end-users (7). Therefore, they are often performed to inform decision making or commissioned by health policy makers.

Description of format contents: PICO or according to PRISMA.

Description of stakeholder involvement

Stakeholders represent decision- and policy-makers and institutions responsible for the selection and implementation of health policies, HTAs, guidelines or end-users. Experts in the respective field are not considered as stakeholders except if they contribute in the policy-planning as described above. It is also known that stakeholders often represent more than one perspective. Concannon et al. described the 7Ps of stakeholders: patients and the public, providers, purchasers, payers, policy makers, product makers, principal investigators (9)

The U.S. EHC Program defined a “stakeholder” as a person or group with a vested interest in a particular clinical decision and the evidence that supports that decision, including:

- Patients, caregivers, and patient advocacy organizations
- Clinicians and their professional associations
- Institutional health care providers, such as hospital systems and medical clinics
- Government agencies
- Purchasers and payers, such as employers and public and private insurers
- Health care industry representatives
- Health care policymakers at the Federal, State and local levels
- Health care researchers and research institutions.

In general stakeholders might be engaged in evidence prioritization, e.g. proposing the title of a systematic review, setting research priorities, in evidence generation, such as performing trials and observational studies, in evidence synthesis, e.g. SR, RR, meta-analysis, in evidence integration, such as modeling, mapping gaps and evidence or cost effectiveness or in evidence dissemination and application, e.g. implementation formats or feedback assessment. Therefore, engagement of stakeholders may not only direct the targeted research topics but also influence the quality and dissemination of study results (9).

At what stages stakeholders can be involved in during the systematic review process? Cottrell et al. identified six categories where the systematic review might benefit: establishing credibility, anticipating controversy, ensuring transparency and accountability, improving relevance and enhancing quality as well as increasing dissemination and uptake of systematic review findings (10). Several challenges to involve stakeholders were determined. They include limited time and resources, skills of researcher to engage stakeholders, finding the right people, balancing multiple inputs and understanding the most appropriate time in the review process to engage different types of stakeholders (10). On the other hand, factors for successful stakeholder engagement included a-priori clarification of the expectations, the roles, types of activities, early engagement in the process, ongoing relationships to build trust and credibility, equality of different stakeholders, tentative inclusion of a trained facilitator and sensitivity to tight timelines (11).

Methods for identifying, selecting, contacting and engaging stakeholders might differ. Furthermore, characteristics of stakeholders themselves are important, e.g. their intensity and modes of engagement may vary according to their interests and expectations, their roles and

their level of power and influence (11). To date there have been few efforts to measure the benefits/tradeoffs of specific stakeholder engagement processes or differing approaches to selecting and engaging differing stakeholder types (10-13).

Of all phases of the systematic review, the topic refinement and developing the key questions was repeatedly identified as the point where stakeholder engagement yielded the greatest benefit (10).

Definition of health services research

See Appendix 1.

Relevance of this study

Systematic reviews may benefit from involving stakeholders, but less is known about the practice, e.g. the proportion of involved stakeholders in recently published SRs and RRs and their type(s) and benefits of engagement in the process of a SR or RR. This information is considered to be relevant for the implementation and/or acceptance of SRs and RRs.

2. Objectives

Our aims are to assess, reflect and compare the current international practice of involving stakeholders in published full systematic reviews (SR) and in rapid reviews (RR) in the field of health services research.

3. Methods

Sample size considerations

Sample size considerations are guided by practical reasons and resources available. As the power of the two proportions is expected to be sufficiently high anyway, due to the big expected difference of the two proportions reporting stakeholder involvement. We consider a proportion of 0.70 for RRs and of 0.25 for SRs. This results in a minimal expected difference of 0.45.

Two groups with N=25 per group show an estimated power of 0.88 and an alpha of 0.023.

To account for potential subgroup analysis, we include a sample of N=30 systematic reviews per group, with 4 groups (SRs, RRs and Cochrane SRs, Cochrane additional database RRs). This allows us to include a maximum of six predictors in the analysis when we perform two-group comparisons.

We will proceed according to the STROBE guidelines as far as they are applicable.

Groups to be studied ("study population")

Full systematic reviews:

Two groups will be studied:

1. Full systematic reviews in general: self-declaration as SR within the title or Methods section of the article or registration as such in the databases searched.
2. Cochrane systematic reviews: SRs are performed according to the Cochrane handbook (1). They are registered, supervised and released within a Cochrane Review Group. They represent the gold standard.

These two groups are selected as they are expected to differ with respect to their transparency of conduct and quality assessment.

Rapid reviews:

Rapid reviews articles will be identified on the basis of the respective self-declaration within the title, Methods section of the abstract or registration as such in the databases searched.

As there is heterogeneity regarding methodology, different terms might be used, e.g. rapid review, rapid method, rapid evidence, rapid synthesis or evidence summary.

Inclusion criteria:

- titled as systematic review or rapid review or reported as such in the objective or method section of the abstract, respectively, or registered as such in the database searched
- studies including rapid review methodology as one part of the overall study
- published between January, 2011 and December, 2015 (it is most crucial to be sure to include sufficient RRs)
- targeting a health services research topic as included in the title or abstract of the article based on the definition provided in Appendix 1

Exclusion criteria:

- designed to assess exclusively economic or cost analysis as described in the title or abstract
- performing a review or overview of reviews
- describing a protocol of a systematic review
- efficacy studies, clinical trials phases I to III, epidemiological studies to assess determinants or risk factors, economic analyses, non-human studies, establishment of databases or registries
- unclear design or description of the intervention

The inclusion and exclusion criteria are presented in Appendix 2.

Targeted stakeholders

We will assess any kind of stakeholder involvement. In addition we will record the following groups of stakeholders:

- Institutional health care providers, such as hospitals and community services

- Government agencies
- Health care policymakers at the Federal, State and local levels

4. Outcomes

Primary outcomes:

- to assess any kind of stakeholder involvement: yes or no
- to assess type(s) of stakeholders involved, e.g. decision- or policy-maker, experts involved in policy-planning, such as institutional health care providers or community services
- to determine phase(s)-related contribution(s) of stakeholders, e.g. developing the research question, literature search, study relevance (quality assessment), formulating or judging recommendations etc.

in recently published SRs and RRs.

Secondary outcomes:

- to evaluate some potential predictors: type and topic of included studies, study population, setting (individual, population group, population, policy) and overall quality score
- to assess reported adverse effects or barriers of stakeholder involvement
- to verify stakeholder involvement
- to report publication issues, such as presence of a review protocol, institutional background of the authors, e.g. specialized in evidence synthesis, year of publication, stakeholder included as key word, dissemination information

5. Data collection

Databases to be searched:

- The following databases will be searched:
Biomedical/Health: Ovid Medline, Embase. Within the search strategy Cochrane SRs will be excluded.
- Cochrane library (for Cochrane SRs) and Cochrane additional databases (for RRs, e.g. DARE reviews, named as CD RR)

The search strategy will be presented in Appendix 1.

Selection of studies

Following the searches in the targeted databases all results of the corresponding systematic review type (SR, RR, Cochrane SR, CD RR) will be pooled and duplicates excluded. These lists will be ordered randomly using the statistical program package R and will be exported to an excel file. The resulting lists will enter the study screening.

Screening of studies

In consecutive order, the randomly ordered references of each group of systematic reviews (SR, RR, Cochrane) will be checked for the general information and the inclusion and exclusion criteria (Appendix 2) until a total of 30 studies are included for each group.

One review authors (JF) will independently screen titles and abstracts for general information, inclusion and exclusion, cross-checked by MM. Inconsistencies or disagreement will be resolved through discussion and with consultation of another review author (MP) where necessary. The process of study selection will be carefully recorded in order to complete the PRISMA-flow diagram.

Data extraction and management

We will use a data collection form for study characteristics and outcome data which has been piloted on two reviews, one SR and one RR. One author, JF, performs full text extraction, supervised and spot-checked by a second researcher, MM. Within the full text article every step of the review will be checked for stakeholder involvement. We will resolve disagreements by consensus or by involving a third review author (MP).

Regarding Cochrane SRs, the protocol (and not the full text article) will be checked for stakeholder involvement as the protocol is relevant for performing the SR.

In addition to the screening form in Appendix 2 we will extract the following data:

- Institution performing the systematic review and contact details of the corresponding author
- Specific topic and intervention within the field of health services research
- Setting: population, population group, individual, policy; major characteristics of the population (overall N, age range or groups, sex)
- Funding for systematic review and notable conflicts of interest of authors
- Stakeholder involvement: any, number, type(s) and phase(s) of involvement, mentioned as key word
- Methodological quality assessment using the AMSTAR tool

All data will enter an Excel file.

To confirm the presented stakeholder involvement and to ask for reasons, whether benefits of stakeholder involvement were measured and about dissemination, the contact author of the included articles will be contacted by emails with pre-formulated questions. It will be indicated that this information will be used for a publication but only summarized and anonymously. Contact authors will be sent one reminder in case of missing responses.

If feasible, two experts in the field will be contacted to review and comment this protocol and to contribute to the final article in order to ensure generalizability and transparency.

We will conduct this study according to this protocol and report deviations from it in the 'Differences between protocol and review' section.

Potential for bias conducting this overview

- selection bias, e.g. regarding year of publication, no search for grey literature
- reporting bias, e.g. incomplete reporting, no study protocol

6. Data analysis

There will be a narrative description of the synthesis.

We will assess the overall proportion of stakeholder involvement as well as within subgroups and according to the outcomes described. We will compare the proportion of stakeholders involved between groups using the non-parametric Wilcoxon rank sumtest.

The results of the AMSTAR assessment will be entered as a score.

Differences between protocol and review

Will be listed if applicable.

Conflicts of interest

None.

Sources of support and funding

Involved authors: Jonas Feldmann (JF), Margot Mutsch (MM), Milo Alan Puhan (MP)

Internal sources

- Epidemiology, Biostatistics and Prevention Institute, University of Zurich, Zurich Switzerland

This is the host institution for the majority of the authors.

This work is part of the Master thesis of Jonas Feldmann.

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Appendix 1: Definition of health services research and search strategy (Ovid Medline)

Search strategy

Systematic reviews:

- Cochrane Database: “systematic review” (title or abstract)
- Ovid Medline and Embase: “systematic review” (ti, ab), excluding “Cochrane” in title or abstract.
- No language restriction
- Published between January, 2011 and December, 2015

Rapid reviews:

- Terms: “rapid systematic review”, “rapid review”, “rapid evidence assessment” or “rapid health technology assessment” in title or abstract
- No language restriction
- Published between January, 2011 and December, 2015

Definition of health services research

No longer available:

Fields of health services research as defined by the Swiss Academy of Medical Sciences (SAMW) (<http://www.samw.ch/de/Forschung/Versorgungsforschung/Kriterien.html>):

*Adaptiert nach (Zugriff 28.11.11)

www.bundesaerztekammer.de/page.asp?his=0.6.3289.3293.3295&all=true

Replaced by:

Lohr KN, Steinwachs DM. Health services research: an evolving definition of the field. *Health Serv Res.* 2002;37(1):7-9. PubMed PMID: 11949927:

“Health services research is the multidisciplinary field of scientific investigation that studies how social factors, financing systems, organizational structures and processes, health technologies, and personal behaviors affect access to health care, the quality and cost of health care, and ultimately, our health and well-being. Its research domains are individuals, families, organizations, institutions, communities, and populations.”

Health Technology Assessments (HTAs) targeting effectiveness as well as meta-analyses and systematic reviews for effectiveness and utility, pharmaceutical trials under everyday conditions, basic research in care-related fields, quality research, methodological developments in the field of health services research, the development and application of new technologies (e-health) and the implementation of knowledge into the clinical practice were included. We excluded studies that focused exclusively on economic or cost analyses, performed a narrative review, an overview of reviews or a protocol of a systematic review. Efficacy studies without assessing the quality of life, clinical efficacy trials phases I to III, epidemiological studies to assess determinants or risk factors, non-human studies, establishment of databases or registries, studies with unclear design or description of the intervention were also excluded.

Appendix 2: Screening of general information and check for inclusion and exclusion

General information

Study ID:	Report ID:	Data form completed: Version number:
First author:	Year of study:	Data extractor:
Citation:		
Publication type (specify):		

Inclusion and exclusion criteria

A) For full systematic reviews

Inclusion criteria:	Criteria fulfilled? Please tick.	
	Yes	No
Is the article titled as systematic review or designed as such in the method section of the abstract or registered as such within the database?		
Is the article published (at least online) between January, 2010 and August, 2015?		
Does the article investigate a health services research topic as defined by the SAMW and presented in the title or abstract?		
Does the article assess an intervention to reduce a condition as presented in the title or abstract?		

To be included, all questions in the above table have to be answered with Yes.

Exclusion criteria:	Criteria fulfilled?	
	Yes	No
Does the article investigate efficacy or describes clinical trials phases I to III, epidemiological studies to assess determinants or risk factors, non-human studies or the establishment of databases or registries as described in the title or the objective or methods section of the abstract?		
Does the article exclusively describe an economic assessment or cost analysis in the title or abstract?		
Does the article describe an overview or review of reviews within the title or abstract?		
Does the article describe a protocol of a systematic review?		

Is the design or the intervention of the article unclear?		
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To be included, all questions in the above table have to be answered with No.

B) For rapid reviews

Inclusion criteria:	Criteria fulfilled? Please tick.	
	Yes	No
Is the article titled as rapid review, rapid method or rapid synthesis or designed as such in the method section of the abstract or registered as such within the database?		
Is the article published (at least online) between January, 2010 and August, 2015?		
Does the article investigate a health services research topic as defined by the SAMW and presented in the title or abstract?		
Does the article assess an intervention to reduce a condition as presented in the title or abstract?		

To be included, all questions in the above table have to be answered with Yes.

The exclusion criteria are the same as described in A).

C) For Cochrane systematic reviews

Inclusion criteria:	Criteria fulfilled? Please tick.	
	Yes	No
Is the article registered and published as Cochrane systematic review within the Cochrane database?		
Is the article published (at least online) between January, 2010 and August, 2015?		
Does the article investigate a health services research topic as defined by the SAMW and presented in the title or abstract?		
Does the article assess an intervention to reduce a condition as presented in the title or abstract?		

To be included, all questions in the above table have to be answered with Yes.

The exclusion criteria are the same as described in A).

Appendix 3: Data extraction for full text assessment

Variables to be assessed (excel file):

- Study type (Cochrane SR, SR, RR, CD RR), Study ID, Report ID
- First study author, contact details of corresponding author, country, mail contact
- Reference citation
- Year of study (publication), version (update or first review)
- Initials of data extractor, initials of people who cross-checked extraction
- Study protocol mentioned, PRISMA checklist used
- Specific review topic, thematic focus, type of intervention, setting
- Study population: size, type, age range of participants included, sex
- Funding source, Conflict of interest statement
- Stakeholder involvement (SI): SI mentioned, SI as key word, number of stakeholders involved, types, stages involved, reasons for SI mentioned, effects of SI mentioned,
- Amstar rating, comparison of rating (Health evidence/health system evidence)

AMSTAR – a measurement tool to assess the methodological quality of systematic reviews.

1. Was an 'a priori' design provided?

The research question and inclusion criteria should be established before the conduct of the review.

Note: Need to refer to a protocol, ethics approval, or pre-determined/a priori published research objectives to score a "yes."

- Yes
- No
- Can't answer
- Not applicable

2. Was there duplicate study selection and data extraction?

There should be at least two independent data extractors and a consensus procedure for disagreements should be in place.

Note: 2 people do study selection, 2 people do data extraction, consensus process or one person checks the other's work.

- Yes
- No
- Can't answer
- Not applicable

3. Was a comprehensive literature search performed?

At least two electronic sources should be searched. The report must include years and databases used (e.g., Central, EMBASE, and MEDLINE). Key words and/or MESH terms must be stated and where feasible the search strategy should be provided. All searches should be supplemented by consulting current contents, reviews, textbooks, specialized registers, or experts in the particular field of study, and by reviewing the references in the studies found.

Note: If at least 2 sources + one supplementary strategy used, select "yes" (Cochrane register/Central counts as 2 sources; a grey literature search counts as supplementary).

- Yes
- No
- Can't answer
- Not applicable

4. Was the status of publication (i.e. grey literature) used as an inclusion criterion?

The authors should state that they searched for reports regardless of their publication type. The authors should state whether or not they excluded any reports (from the systematic review), based on their publication status, language etc.

Note: If review indicates that there was a search for "grey literature" or "unpublished literature," indicate "yes." SIGLE database, dissertations, conference proceedings, and

trial registries are all considered grey for this purpose. If searching a source that contains both grey and non-grey, must specify that they were searching for grey/unpublished lit.

- Yes
- No
- Can't answer
- Not applicable

5. Was a list of studies (included and excluded) provided?

A list of included and excluded studies should be provided.

Note: Acceptable if the excluded studies are referenced. If there is an electronic link to the list but the link is dead, select "no."

- Yes
- No
- Can't answer
- Not applicable

6. Were the characteristics of the included studies provided?

In an aggregated form such as a table, data from the original studies should be provided on the participants, interventions and outcomes. The ranges of characteristics in all the studies analyzed e.g., age, race, sex, relevant socioeconomic data, disease status, duration, severity, or other diseases should be reported.

Note: Acceptable if not in table format as long as they are described as above.

- Yes
- No
- Can't answer
- Not applicable

7. Was the scientific quality of the included studies assessed and documented?

'A priori' methods of assessment should be provided (e.g., for effectiveness studies if the author(s) chose to include only randomized, double-blind, placebo controlled studies, or allocation concealment as inclusion criteria); for other types of studies alternative items will be relevant.

Note: Can include use of a quality scoring tool or checklist, e.g., Jadad scale, risk of bias, sensitivity analysis, etc., or a description of quality items, with some kind of result for EACH study ("low" or "high" is fine, as long as it is clear which studies scored "low" and which scored "high"; a summary score/range for all studies is not acceptable).

- Yes
- No
- Can't answer
- Not applicable

8. Was the scientific quality of the included studies used appropriately in formulating conclusions?

The results of the methodological rigor and scientific quality should be considered in the analysis and the conclusions of the review, and explicitly stated in formulating recommendations.

Note: Might say something such as "the results should be interpreted with caution due to poor quality of included studies." Cannot score "yes" for this question if scored "no" for question 7.

- Yes
- No
- Can't answer
- Not applicable

9. Were the methods used to combine the findings of studies appropriate?

For the pooled results, a test should be done to ensure the studies were combinable, to assess their homogeneity (i.e., Chi-squared test for homogeneity, I_2). If heterogeneity exists a random effects model should be used and/or the clinical appropriateness of combining should be taken into consideration (i.e., is it sensible to combine?).

Note: Indicate "yes" if they mention or describe heterogeneity, i.e., if they explain that they cannot pool because of heterogeneity/variability between interventions.

- Yes
- No
- Can't answer
- Not applicable

10. Was the likelihood of publication bias assessed?

An assessment of publication bias should include a combination of graphical aids (e.g., funnel plot, other available tests) and/or statistical tests (e.g., Egger regression test, Hedges-Olken).

Note: If no test values or funnel plot included, score "no". Score "yes" if mentions that

publication bias could not be assessed because there were fewer than 10 included studies.

- Yes
- No
- Can't answer
- Not applicable

11. Was the conflict of interest included?

Potential sources of support should be clearly acknowledged in both the systematic review and the included studies.

Note: To get a "yes," must indicate source of funding or support for the systematic review AND for each of the included studies.

- Yes
- No
- Can't answer
- Not applicable

Shea *et al.* *BMC Medical Research Methodology* 2007 **7**:10 doi:10.1186/1471-2288-7-10

Additional notes (in italics) made by Michelle Weir, Julia Worswick, and Carolyn Wayne based on conversations with

Bev Shea and/or Jeremy Grimshaw in June and October 2008 and July and September 2010.