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Overviews of Reviews: Concept and Development

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Abstract

Background: In the last years, overviews of systematic reviews, or umbrella reviews, have seen a dramatic increase in their use. An overview aims to provide a summary of the included reviews and will often examine research questions beyond those addressed in the systematic reviews being synthesised. The purpose of this article is to provide some recommendations on how overviews should be conducted and reported. Method: A literature review was performed to identify relevant papers on both methodological and applied overviews. Results: The current literature recommends carrying out overviews by following similar steps to those of systematic reviews: (a) Defining the overview research question; (b) inclusion and exclusion criteria; (c) literature search; (d) data extraction; (e) assessment of risk of bias and reporting quality; (f) overview results; and (g) reporting the overview. Of special interest is how to address dependencies between the systematic reviews. Conclusions: Overviews allow evidence to be efficiently combined from multiple systematic reviews. This offers the possibility of translating and summarizing large amounts of information. As in primary studies and systematic reviews, conducting and reporting of overviews must meet appropriate quality standards.

Keywords: Umbrella review, overview of reviews, meta-review, metameta-analysis, evidence synthesis.

Resumen

Meta-Revisiones: Concepto y Desarrollo. Antecedentes: en los últimos años, las meta-revisiones, o revisiones paraguas, han incrementado exponencialmente su uso. Una meta-revisión proporciona un resumen de las revisiones incluidas y, a menudo, aborda preguntas más allá de las planteadas en las revisiones sistemáticas sintetizadas. El propósito de este artículo fue proporcionar recomendaciones sobre cómo deben hacerse y reportarse las meta-revisiones. Método: se llevó a cabo una revisión de la literatura para identificar artículos relevantes sobre meta-revisiones, tanto metodológicos como aplicados. Resultados: a día de hoy la literatura sobre meta-revisiones recomienda seguir los mismos pasos que en las revisiones sistemáticas: (a) definición de la pregunta de investigación; (b) criterios de inclusión y exclusión; (c) búsqueda de la literatura; (d) extracción de datos; (e) valoración del riesgo de sesgo y de la calidad del reporte; (f) presentación de resultados; y (g) reporte de la meta-revisión. De especial interés es cómo abordar los problemas de dependencia entre las revisiones sistemáticas sintetizadas. Conclusiones: las meta-revisiones permiten combinar evidencia de múltiples revisiones sistemáticas de una forma eficiente. Esto permite transformar y resumir grandes cantidades de información. Al igual que en los estudios primarios y las revisiones sistemáticas, las meta-revisiones deben realizarse y reportarse cumpliendo con estándares mínimos de calidad.

Palabras clave: revisión paraguas, resumen de revisiones, meta-revisión, meta-meta-análisis, síntesis de la evidencia.

Over the past decades, systematic reviews and meta-analyses have reached and maintained the highest rank in the hierarchy of evidence (Cooper et al., 2019; Harbour & Miller, 2001; Higgins et al., 2019). Initially devised to facilitate the efficient accumulation of scientific knowledge, the proliferation of systematic reviews and meta-analyses published in recent years has brought along new challenges for practitioners and policy makers aiming to keep up to date in a specific field, as well as new opportunities for evidence synthesis (Bastian et al., 2010; Corral et al., 2021; Valero-Aguayo et al., 2021). This explains why evidence synthesis projects which entail summaries of existing systematic reviews are increasingly popular (cf. Pieper et al., 2012).

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Different terms have been proposed so far to label the synthesis of several systematic reviews. One of those is tertiary study, which results from considering individual studies with samples of participants as primary studies and systematic reviews of those as secondary studies, so that the synthesis of systematic reviews could be thought of as a third level (e.g., Biondi-Zoccai, 2016; Cruzes & Dybå, 2011). While this is an insightful label with regards to the underlying data structure, it has not been widely used to date. One explanation for this is that in the article where Gene Glass coined the term "meta-analysis" (one of the key references in the evidence synthesis literature), he advocated to use the label secondary research for "re-analysis of data for the purpose of answering the original research question with better statistical techniques, or answering new questions with old data" (Glass, 1976, p. 3).

Another relevant term in this context is meta-epidemiological study. Since a landmark paper was published (Schulz et al., 1995), meta-epidemiology has become an established research area where associations between methodological characteristics of primary studies and the magnitude of reported outcomes are

investigated (Sterne et al., 2002; Trinquart et al., 2013). Although meta-epidemiology usually involves combining information from systematic reviews, this term is typically used only for evidence synthesis projects with a strong methodological focus which might include primary studies (especially randomised controlled trials) as well as systematic reviews (Biondi-Zoccai, 2016).

A third label, very commonly used, is *umbrella reviews*. One reason for this popularity is that this is a more flexible label, since umbrella reviews may address substantive and/or methodological research questions (Ioannidis, 2017). A second reason is that this term has been embraced by the Joanna Briggs Institute (JBI), an organisation promoting high-quality evidence synthesis projects in Healthcare (Aromataris et al., 2020; see also https://www.jbi.global).

Another very popular label is *overview of reviews*, or *overview of systematic reviews*. This term has been favoured by Cochrane, devoted to promoting high-quality reviews in Medicine and arguably the most influential organisation of its kind (Pollock et al., 2021; see also https://www.cochrane.org). Similarly, the Campbell Collaboration – a key institution promoting high-quality evidence synthesis projects in Education, Social Work, and Criminology– is also adopting the term 'overview' in this context (Littell, 2018; see also https://www.campbellcollaboration.org). Unlike other labels presented before, which may entail a combination of primary studies and reviews in the same synthesis, an overview of reviews only includes systematic reviews as analysis units (cf. Biondi-Zoccai, 2016).

Some other terms can still be found in the literature to label these studies, including meta-meta-analysis (e.g., Inthout et al., 2015), second order meta-analysis (Schmidt & Oh, 2013), meta-synthesis (e.g., Johnson et al., 2010), and meta-review (e.g., Lecomte et al., 2020). But regardless of the label used by different authors and in different disciplines, a look back to publication records reveals an exponential growth in the number of such syntheses over in the last 10-15 years. Figure 1 presents a graph describing the exponential growth of this kind of reviews in Psychology. The search was carried out in the PsycInfo data base in December 2021. The full strategy was as follows: TI "Umbrella review" OR TI "metasynthesis" OR TI "meta-review" OR TI "tertiary study" OR TI "meta-epidemiological study" OR TI "overview of reviews" OR TI "overview of systematic reviews" OR TI "meta-meta-analysis" OR TI "second order meta-analysis". The search was limited to journal articles without any limitation for date and language. The total number of results found was 760.

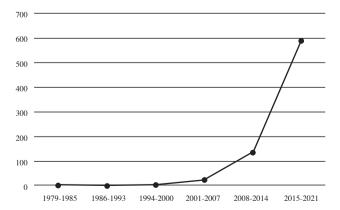


Figure 1. Number of articles including "overview of reviews" or a related term in the title over the past four decades

Throughout this paper, we will use the term 'overview', since we are focusing on combinations of systematic reviews only and this is a simple and widely accepted term for such studies. Based on the well-established framework of systematic reviews and meta-analyses, the purpose of this article was to present guidelines on how to conduct and report an overview of reviews, or an umbrella review.

Method

Defining the Overview Research Question

Conducting an overview can be motivated by several goals. First, researchers may want to provide a summary of the state of the art in a specific area. For instance, Andersson and colleagues (2019) examined the effectiveness of internet-delivered cognitive-behavioural therapy for adults with anxiety and mood disorders. Their overview included nine systematic reviews that showed evidence of effectiveness across different diagnostic categories.

An overview might also be conducted to address a controversial topic in the literature (e.g., a hypothesis that has been both supported and dismissed before). As an example, Catalán-Matamoros et al. (2016) were interested in determining whether exercise programmes are effective for older adults with depression. Their overview included three systematic reviews that overall suggested that exercise is safe and reduces depressive symptoms in older people.

A third motivation is to explore a new research question that is better examined through the combination of systematic reviews. As an illustration, Sala and colleagues (2019) examined the near versus far transfer phenomena in cognitive training, that is, whether cognitive training programmes have any benefits beyond the trained (or a similar) task. The authors investigated whether the effects differed across different programmes and/or populations, and their overview of ten systematic reviews found consistent results across the different categories; in particular, once placebo effects and publication bias were controlled for, no far transfer effects were observed for any programme or population type.

Although the goals presented so far are substantive, overviews can also have a methodological focus. In this vein, meta-epidemiological studies investigate associations between methodological aspects of primary studies and/or systematic reviews and the magnitude and/or direction of the results. As an example, Inthout and colleagues (2015) were interested in testing empirically the association between sample size and effect size. Their overview of 3,263 meta-analyses found evidence that studies with smaller sample sizes reported more heterogeneous outcomes.

Last, another methodological area of interest for overviews is to examine how systematic reviews are conducted (e.g., Tsujimoto et al., 2022) and/or reported. For instance, López-Nicolás et al. (2021) conducted an overview of 100 meta-analyses examining the effectiveness of interventions in Clinical Psychology, and the results provided an overall picture of the reporting standards and motivated some recommendations for future reviews in this field.

Regardless of the goal of the overview, pre-registration is always a good practice. Platforms such as the International PROSPEctive Register Of Systematic Reviews (PROSPERO, https://www.crd. york.ac.uk/prospero/) and the Open Science Framework (https://www.osf.io) are popular choices for researchers aiming to comply with the requirements of the new, but widely supported, open

science movement (Lakens et al., 2016; Moreau & Gamble, 2020).

Inclusion and Exclusion Criteria

Once the research question has been defined, the next step in an overview involves setting the inclusion criteria. Same as in a systematic review, if the overview aims to compare two or more interventions then the PICO acronym is a good starting point (Aromataris et al., 2020; Smith et al., 2011), as this will help define the Population/s (for instance, children and adolescents, adults, or human participants of any age), Intervention/s, Comparator/s, and Outcome/s. The ultimate goal at this stage is to set a list of criteria sufficient to articulate an appropriate search strategy and to decide which reviews should be included when the search records are retrieved.

Exclusion criteria may also be considered. In this regard, organisations such as Cochrane, the Campbell Collaboration, and the Joanna Briggs Institute recommend that only high-quality systematic reviews be included in overviews. To align with these guidelines, overview authors might want to condition final inclusion on the results of the assessments of risk of bias and reporting quality (further details about those assessments are provided below).

A challenge specific to overviews at this stage is the overlap of primary studies, that is, the possibility that the same primary study might have been part of two or more of the systematic reviews included in the overview. This is somehow analogous to the phenomenon of multiplicity in systematic reviews, which happens when some primary studies included in a review report two or more relevant results based on the same (or partially the same) participants (López-López et al., 2018). Overlap of primary studies in overviews poses concerns such as overrepresentation of the overlapping studies and statistical dependency, and hence some authors consider that overlaps should be minimised, even excluding some eligible systematic reviews if necessary (e.g., Biondi-Zoccai, 2016). On the other hand, tools to appraise and report the extent of overlap have been proposed (Pieper et al., 2014; Pollock et al., 2021), and other authors have suggested that overlap might be enriching for overviews as long as statistical dependencies are appropriately handled (Hennessy & Johnson, 2020). Last, Pollock and colleagues (2021, Figure V.4.a) provide a flow chart to guide decision about inclusion of overlapping primary studies in overviews on intervention effectiveness.

Literature Search

Same as with any evidence synthesis project, overviews entail defining a search strategy and reporting it in a transparent way, including the databases searched, the search algorithms, and the temporal range (Hartling et al., 2012; Smith et al., 2011). Nowadays, electronic databases such as PsycInfo and MEDLINE/PubMed constitute a key resource for any review of published research, and a combination of several of them is highly recommended. In the case of overviews, the inclusion of databases dedicated to systematic reviews, such as the Cochrane Database of Systematic Reviews and the Database of Abstracts of Reviews of Effects (for a wider list, see Golder & Wright, 2016) becomes particularly relevant. Furthermore, contacting institutions devoted to evidence-based policy making – such as the National Institute for Clinical

Excellence (NICE) in the UK or the Food and Drug Agency (FDA) in the USA – might help retrieve reviews that were commissioned and undertaken but have not yet reached publication status (*grey literature*).

In contrast to systematic reviews, exhaustivity is not necessarily required for all overviews. An exhaustive literature search will be normally expected for content-focused overviews, such as those intended to summarise the state of the art in a research area or aiming to solve discrepancies (unless the conflicting reviews are a relatively small number and can be easily identified). On the other hand, overviews with a methodological focus often select a subset of the reviews that can be reasonably considered as a representative sample from the target population of reviews (e.g., Inthout et al., 2015; Rubio-Aparicio et al., 2018).

Data Extraction

Like in a systematic review, study selection and data extraction in an overview should involve two independent reviewers to avoid biases (Hartling et al., 2012). The nature and volume of extracted data will largely depend on the scope and specific objectives of the overview (for overviews focused on intervention effectiveness, see Pollock et al., 2021 for a list of suggested variables). Sometimes the information required to examine on overview hypotheses might not be reported in the retrieved publications, but data can be augmented through requests to review authors (Papageorgiou & Biondi-Zoccai, 2016). At this stage, the elaboration (and possible updating) of a codebook file detailing the information to be extracted and guiding the decisions that will need to be made can be very helpful (e.g., López-Nicolás et al., 2021).

Assessment of Risk of Bias and Reporting Quality

The methodological rigour of published reviews varies considerably (Smith et al., 2011). A further challenge at this stage is that, while there is abundance of instruments to appraise the risk of bias of primary studies, the toolkit for systematic reviews is more limited. However, some checklists have been developed. One of such tools is ROBIS (Whiting et al., 2016), which is intended for use in reviews of effectiveness, aetiology, diagnosis and prognosis. Another option is AMSTAR 2, which was developed to appraise the methodological quality of systematic reviews including randomised and/or non-randomised studies in healthcare (Shea et al., 2017). Although advisable in any overview, risk of bias assessment is particularly important in overviews with a substantive goal such as providing a summary of the state of the art or solving discrepancies between existing reviews.

Another domain to be considered at this stage is reporting quality. In the last years, different guidelines and checklists on how to adequately report systematic reviews have proliferated. The most widely used instrument is PRISMA 2020 (Preferred Reporting Items for Systematic Reviews and Meta-Analyses; Page et al., 2021), although alternative instruments tailored to specific contexts are also available, including MOOSE (Meta-analysis Of Observational Studies in Epidemiology; Stroup et al., 2000) and REGEMA (REliability GEneralization MEta-analysis, Sánchez-Meca et al., 2021). Adequate reporting of systematic reviews is receiving more attention nowadays, as it is considered crucial in the pursuit of a more transparent and reproducible science (Lakens et al., 2016; Moreau & Gamble, 2020).

An important aspect when examining the methodological and reporting quality of systematic reviews is publication bias. Every review including a meta-analysis should report analyses of the potential extent of publication bias, ideally combining several approaches (Vevea et al., 2019), and the absence of such analyses does not discard publication bias as a threat to the validity of the review results. A methodological overview of 116 systematic reviews found that publication bias analyses were not reported in 31% of them; among these, *post-hoc* sensitivity analyses conducted by the overview authors yielded evidence of publication bias in one in every five of them (Onishi & Furukawa, 2014).

Results

Once the previous stages have been accomplished, it is time to obtain and present the results of an overview. This section outlines different approaches to address this task, and it also includes some recommendations with regards to the reporting of an overview.

Overview Results

Same as for systematic reviews, the first goal in an overview is to provide a descriptive analysis. Overviews with a substantive focus might include more information at the study level (e.g., description of the samples and interventions/programmes examined), with the aim to characterise the primary studies comprising each review. On the other hand, overviews with a methodological focus might pay more attention to the reporting practices among reviews or to some parameter estimates from the meta-analyses reported therein. Regardless of the focus of the overview, some key aspects that should be examined at this point include number of studies synthesized in each review and range of research designs and sample sizes of the primary studies. The main characteristics of the systematic reviews and meta-analyses included in an overview of reviews should always be reported in a comprehensive table. A description of these characteristics will offer a picture of the systematic reviews synthesized in the overview.

Effect sizes provide the best summary of the results from primary studies (e.g., Cooper et al., 2019). If at least some of the systematic reviews report effect sizes (or an average of those using meta-analytic techniques), these data will often be the basis for the core part of the results in an overview. Many overviews summarise information on effect sizes using tables and/or figures but without performing a statistical synthesis, and this is in line with guidelines from influential entities such as the JBI (cf. Aromataris et al., 2020). A table with the main statistical results reported in each meta-analysis of the overview should be included in this section. Statistical information such as the number of studies, average effect size, 95% confidence interval, 95% prediction interval, heterogeneity statistics (e.g., Q statistic, I^2 index, between-study standard deviation, between-study variance estimator) and how publication bias and small-study effects were assessed (e.g., failsafe N, funnel plots, trim-and-fill method for imputing missing effect sizes, Egger test, p-uniform, p-curve, etc.) should be included in a table for each meta-analysis. In meta-analyses that report results for different outcomes, the table should report separately all of these statistical results.

Some analysis opportunities might also arise if at least part of the systematic reviews carried out a meta-analysis. For instance, Inthout and colleagues (2015) were interested in testing empirically whether small studies are more heterogeneous than large ones. They examined 3,263 meta-analyses and concluded that heterogeneity among small studies was greater than among larger studies. Since they were interested on a meta-analytic parameter – the heterogeneity variance (\square) – it is clear how taking an overview perspective provides an advantage in this context (for advanced methods to examine this question at the level of a single meta-analysis, see Viechtbauer & López-López, 2021).

When examining whether the magnitude and/or variability among effect sizes is associated with some characteristics of the primary studies or the systematic reviews, a simple strategy is to define two categories and calculate the ratio between the average effect size (or estimate of \square) from each category (e.g., Inthout et al., 2015; Schulz et al., 1995). Other possibilities include metaregression at a higher order (e.g., Sterne et al., 2002) and multilevel modelling to account for the nested structure of the database (e.g., Rhodes et al., 2015). In most overviews involving use of statistical models at this stage, analysis strategies to appropriately model (or account for) dependency will become more relevant as the degree of overlap of primary studies increases (López-López et al., 2018).

If the systematic reviews being combined involve multiple treatments that can be connected across reviews, the overview authors might be tempted to draw inferences about the comparative effectiveness of each pair of interventions (e.g. by performing network meta-analysis directly from the effect estimates reported in the reviews). However, recent guidelines from Cochrane discourage this practice (Pollock et al., 2021), since the transitivity assumption – the key assumption underlying indirect comparisons – can rarely be assessed from the information typically reported in systematic reviews. If researchers are primarily interested in multiple treatment comparisons, an alternative is to design and conduct a systematic review and network meta-analysis including a search strategy that feeds from existing reviews where only pairwise meta-analyses were carried out (e.g., López-López et al., 2019).

Reporting an Overview

The report of an overview, typically in the form of a scientific paper, needs to communicate the process and results of the overview in a systematic and transparent manner. Research replicability is one of the most important tenets of the scientific method. Therefore, there must be enough detail to make the overview replicable should an independent research team want to do so.

Several guidelines are available for researchers reporting an overview. A key resource is the Preferred Reporting Items for Overviews of Reviews (PRIOR, Pollock et al., 2019), which was developed as an extension of reporting guidelines such as PRISMA 2020 in the context of overviews. Moreover, both Cochrane (Pollock et al., 2021) and the JBI (Aromataris et al., 2020) provide recommendations to support authors in the process of reporting an overview.

Table 1 summarizes the recommendations provided in this paper.

Discussion

This paper provides guidelines to identify overviews of systematic reviews, as well as recommendations to conduct and

Table 1 Summary of recommendations when conducting an overview of reviews	
Stage	Recommendations
Defining the research question	Some examples of goals include: - To provide a summary of the state of the art - To address a controversial topic - To explore a new research question - To investigate associations between methods and results - To examine the conducting and/or reporting of systematic reviews Pre-registration is advisable (e.g., using PROSPERO, OSF)
Inclusion/exclusion criteria	PICO may help for overviews of interventions Consider inclusion based on methodological quality standards Consider the issue of overlapping primary studies
Literature search	A combination of sources is highly recommended Consider the inclusion of databases dedicated to systematic reviews (e.g, CDSR, DARE) Grey literature may be retrieved by contacting institutions devoted to evidence-based policy making (such as NICE and the FDA) Exhaustivity will be key or accessory depending on the research question
Data extraction	By two independent reviewers Nature and volume will depend on the research question Requests to review authors might help retrieve missing information A codebook file is a great way to guide and document this stage
Assessment of risk of bias	Consider using ROBIS and/or AMSTAR 2
Assessment of reporting quality	PRISMA 2020 is the most popular tool Alternatives designed for specific types of reviews are available (e.g., MOOSE, REGEMA)
Overview results	Describe the main characteristics of each systematic review using tables If some of the reviews reported meta-analyses, use tables to report their results Analyses of publication bias conducted in the reviews should also be presented Analysis opportunities (sometimes using the review as the analysis unit) can be considered Network meta-analysis directly from the effect estimates reported in the reviews should not be conducted
Reporting of the overview	Keep replicability in mind Consider using PRIOR at this stage Extensive guidance is also available from Cochrane and JBI

report them. Importantly, the goal in an overview is not to replicate the tasks completed in the included systematic reviews such as search, coding, and risk of bias assessment of the individual studies (Pollock et al., 2021); instead, an overview aims to provide a summary of the included reviews and will often examine research questions beyond those addressed in the systematic reviews being synthesised.

Overviews allow combining evidence from multiple systematic reviews in a smooth and efficient fashion. This offers the possibility to translate and summarise large amounts of information. By taking a higher ground, typically using broader research questions than those articulated in systematic reviews (Pollock et al., 2021), overviews also have the potential to provide novel findings.

Overviews also have the potential to enhance the methodological quality of evidence synthesis as a field (Ioannidis, 2017), as they allow a higher-level synthesis of the evidence and a better recognition of the uncertainties, biases and knowledge gaps. For instance, Polanin and colleagues (2016) reviewed 383 meta-analyses and found strong evidence that published studies yielded larger effect sizes than unpublished studies.

It is even possible to conceive an evidence synthesis project in which overviews constitute the analysis unit, and indeed several such studies have already been conducted (e.g., Hartling et al., 2012; Page et al., 2016; Pieper et al., 2012, 2014). So far, their focus has been primarily methodological.

As a novel area of research, the field of overviews is not one without challenges. An example of this is the inconsistency among the terminology used to label such studies, with some articles failing to include a descriptor of the study design in the title and many others choosing one among different options (e.g., overview of reviews, umbrella review, meta-meta-analysis...). Such inconsistencies hamper the ability for electronic searches to locate overviews, which in turns limits their impact. Some recommendations are available in this regard, both to researchers interested in identifying overviews and to those reporting them (Lunny et al., 2016).

Beyond terminology, inconsistencies have been found among the methodological rigour followed by published overviews (cf. Pieper et al., 2012), which suggests that early approaches to overviews of reviews were more opportunistic than systematic (Hartling et al., 2012). To this respect, the development in recent years of guidelines from entities such as Cochrane (Pollock et al., 2021) and the JBI (Aromataris et al., 2020) is aimed at improving the consistency and the quality of the methods adopted in future overviews.

In conclusion, overviews of reviews constitute a novel and increasingly popular approach to evidence synthesis. Although

the methodological inconsistencies noted by several authors cast doubts regarding their current place in the hierarchy of evidence, overviews already offer a powerful tool to appraise and improve the quality of research synthesis, as well as to examine research questions that benefit from the use of systematic reviews as analysis units.

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