A systematic mapping and bibliometric analysis of systematic reviews of health economic evaluations: A protocol

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Abstract

Background: Systematic reviews of health economic evaluations (SR-HEs) provide a critical tool for synthesizing published literature to guide decision makers towards implementing evidence-based policy and healthcare practice. However, the quality of the methodology and reporting of SRs is often flawed due to insufficiencies in their design, conduct and reporting. Meta-research has led to methodological improvements in many research fields but is not widely established in health economic evidence synthesis. To enable future meta-research on SR-HEs, we will create a database of SR-HEs and provide a bibliometric analysis of this literature.

Methods: We will perform a systematic search in MEDLINE for systematic reviews with/without meta-analyses of health economic evaluations. We will include studies that performed a systematic review of cost-effectiveness, cost-minimization, cost-utility, cost-benefit, and/or cost-consequence analyses. We will automatically extract data from Ovid MEDLINE and Web of Science to conduct a bibliometric analysis by examining publication, citation, and collaboration patterns.

Discussion: Our study will provide a map of SR-HEs accompanied by a thorough bibliographic analysis of this research field. Our publicly accessible database can be used to assist future research and meta-research efforts in the field of health economics. Our bibliometric analysis will provide insights into the evolution of this research field over the years in terms of publications, citations, and scientific collaborations.

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Background

With rising pressure on health budgets (1), pricing and reimbursement decisions are gaining increasing importance. Health economic evaluation, which is the comparative analysis of alternative health interventions to understand their use of resources and costs relative to their effectiveness, responds to this need as it aims to contribute to cost minimization and the maximization of clinical benefits for patients and healthcare systems (2, 3).

Systematic reviews (SRs) are a tool to comprehensively summarize the evidence on a specific research question. They also offer the opportunity to quantitively synthesize the available evidence and to identify potential knowledge gaps (4). With a greatly expanding body of literature on health economic evaluations, SRs can integrate information from multiple sources and provide a critical tool for synthesizing published literature to guide decision makers towards implementing evidence-based policy and healthcare practice (5, 6). There are a few meta-research efforts that assessed the quality of SRs of health economic evaluations (SR-HEs), indicating flaws in the methodology, design and reporting of these SRs (7–10). In addition, synthesis of health economic evidence presents challenges due to the heterogeneity of populations, cost estimates, data on effectiveness and choice of comparators, as well as

methodological differences between modelling studies that might influence cost-effectiveness results (5, 11).

Creation of a comprehensive database of SR-HEs may facilitate further in-depth assessments of the patterns, strengths and weaknesses of this literature. Basic bibliometric analysis can also offer additional insights into this research field and its evolution. The investigation of publications, citation and collaboration patterns in SR-HEs may help understand and map the cumulative scientific knowledge and evolutionary nuances of SRs in the field of health economics (14). To our knowledge, there is no comprehensive published database, and bibliometric overview and analysis of the SR-HEs literature. Our study aims to fill this gap by creating a database of SR-HEs. Then, we will use this database to investigate the publication, citation, and collaboration patterns and trends.

Methods

We use the PRISMA-P (Preferred Reporting Items for Systematic review and Meta-Analysis Protocols) checklist to develop and report the methods for this protocol, where appropriate (15). Also, our systematic review will follow the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) statement, where appropriate (16). However, in this paper, we will focus on creating a database and a comprehensive bibliometric analysis of SR-HEs. We will make no effort to perform an evidence synthesis or risk of bias assessment in the present protocol, since the literature is likely to be vast and heterogeneous and any evidence synthesis and risk of bias assessment efforts may be decided in subsequent work. We will document any deviations from, or amendments to this protocol including details of the date, changes made, and the rationale for changes.

Eligibility criteria

We will include SRs with/without meta-analyses of full economic evaluations that included a search in at least one bibliographic database. By full economic evaluations we include cost-effectiveness, cost-minimization, cost-utility, cost-benefit, and cost-consequence analyses. We will not use any exclusion criterion based on the type of assessed interventions or medical conditions. We will include articles published in the English language. We will exclude (a) SRs of partial economic studies (e.g., cost-of-illness studies or program cost studies), (b) SRs of health economic assessments not focused on humans. We will also exclude clinical practice guidelines and secondary reports of health technology assessments, because our study will focus on scientific literature instead of documents stemming from regulatory and clinical guidance processes. We will also exclude conference abstracts, protocols, narrative reviews, commentaries, and editorials. We will exclude overviews of SRs with/without meta-analyses of health economic evaluations. Table 1 shows a detailed description of our eligibility criteria in terms of population, intervention, comparison, outcome, timing, and setting.

Table 1
PICO characteristics

Population	**Patients with any medical condition (for health economic evaluations of treatment interventions) or healthy individuals (for health economic evaluations of preventive interventions)**
Intervention	Any interventions, such as pharmacological, psychological/behavioral, or surgical interventions, and vaccinations
Comparator	Any
Outcome	Average or incremental cost-effectiveness and net benefit outcomes.

Literature search

We will systematically search Ovid MEDLINE for articles published from inception to April 19, 2022, using the following search algorithm: ((economic$.ti. or cost$.ti. or cost benefit analysis/ or (treatment outcome/ and ec.fs.)) not ((animals/ not humans/) or letter.pt.)) and (MEDLINE.tw. or systematic review.tw. or meta-analysis.pt. or intervention$.ti.). We created this search algorithm by combining a validated search filter for economic evaluations with a validated filter for the retrieval of systematic reviews (17, 18). The use of validated algorithms provides a good balance between sensitivity and precision.

Two raters (among MH, LMG, TM, LB) will independently assess the eligibility of each record identified in the literature search based on the above criteria. We will resolve conflicts through discussion and consensus between the reviewers. We will make the records of the search and selection process available at the time of publication of the review and document the study selection process in a PRISMA flowchart.

Data extraction

Due to the expected large number of eligible articles (expected to be several thousand eligible articles based on preliminary searches), we will focus our data extraction in this protocol on items we can retrieve and process automatically. We will collect all available publication metadata from Ovid MEDLINE. We will extract information about the name of scientific journal, year of publication, number of authors, and the affiliation and country of the first author and senior author. We will also examine whether the keyword “systematic review” or “meta-analysis” is reported in the title of the eligible articles, and whether a pre-registration number is reported in the abstract. Moreover, we will access Web of Science to enrich our dataset with journal subject areas, retrieve journal impact metrics, and to extract the number of citations received by the eligible articles.

Statistical analysis

We will present descriptive statistics (median and interquartile range for continuous variables, and frequencies for binary or categorical variables) for the extracted variables. We will also present these descriptive statistics by publication year, and by journal subject area.

Techniques for bibliometric analyses consist of performance analysis and science mapping. Performance analysis focuses on the contributions of research constituents to a research field (14). Specifically, we will identify the top 10 of (a) scientific journals, (b) countries, (c) academic or research institutions, and (d) first and senior authors publishing the highest number of SRs-HEs. We will examine whether there is an increasing trend in the number of SRs-HEs across the years, unadjusted and after adjusting for total number of MEDLINE items per year. Moreover, we will examine whether there is a change in the ranking of scientific journals, countries, and academic or research institutions publishing the highest number of SRs-HEs across the years (i.e., before 2000, 2000 to 2010, 2010 to 2020, and after 2020).

Science mapping refers to the study of the relationships between research constituents (14). Specifically, we will perform a citation analysis to identify the most highly cited SR-HEs. Also, we will inspect the collaborations among scholars and among different academic or research institutions, and we will identify the most productive collaborations.

We will also examine whether there are differences in publication patterns based on journal subject area, and geographic region. For this purpose, we will classify journals into three categories: general medical journals, medical subspecialty journals, and health economics journals. We will categorize countries according to World Bank country classifications (low, lower-middle, upper-middle, and high-income). We will use parametric tests (or exact tests, when necessary) to examine differences in publication patterns between the categories of scientific journals and geographic regions. We will use two-sided statistical tests and will present point estimates with 95% confidence intervals.

Software

We will use Abstrackr (19) for Title/Abstract screening, Zotero for literature management, and R statistical software for data management and statistical analysis.

Discussion

As the number of SR-HEs increases, it is important to track and study their publication patterns. Until now, there has been no systematic and comprehensive mapping of this literature. Our research article will have two main research outputs. First, it will provide a publicly available database of SR-HEs. Our database can be used by health economists to rapidly identify relevant SR-HEs for their research field. Also, it can be used by meta-researchers that aim to improve the quality of health economic evaluations and SRs thereof. Currently, there are some meta-research efforts for SR-HEs (7–10), but researchers always systematically review the literature from scratch. By making our database publicly available, we will facilitate and enable further meta-research in the field of SR-HEs. Second, our research article will take advantage of a large volume of publications to present a thorough bibliometric analysis of SR-HEs. This analysis will result in conclusions about the cumulative scientific knowledge and evolutionary nuances of this research field.

Abbreviations

SR-HE	Systematic review of health economic evaluations
SR	Systematic review

Declarations

Ethics approval and consent to participate: Ethics approval is not necessary as we use only previously published data.

Consent for publication: Not applicable.

Availability of data and materials: The datasets that will be generated and analyzed during the current study, and the code for the statistical analysis will be made publicly available upon the publication of the research article. Both datasets and code will be made publicly available under a license that allows for reuse after publication.

Competing interests: The authors declare that they have no direct competing interests. TM is a protocol editor of Systematic Reviews.

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Author contributions: MH and LB had the original idea, designed, and wrote the first draft for the protocol of this study. JPAI and TM critically commented on the protocol. MH, LMG, TM and LB will perform the systematic literature search and select the eligible studies. MH and LB will perform the statistical analysis. All authors will interpret the analyses and will write the final paper.

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