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HOW TO AVOID MISSING HALF THE EVIDENCE: COMPARING THE USE OF GENERIC AND SPECIFIC ELECTRONIC SEARCH TERMS USED TO IDENTIFY HEALTH OUTCOMES FOR A SYSTEMATIC REVIEW.

Matt Egan, *senior investigator scientist*[1*], Alice MacLean, *investigator scientist*[1], Helen Sweeting, *senior investigator scientist*[1], Kate Hunt, *programme leader*[1].

[1] MRC / CSO Social and Public Health Sciences Unit (SPHSU), 4 Lilybank Gardens, Glasgow G12 8RZ.

[*]Correspondence to m.egan@sphsu.mrc.ac.uk

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Abstract

Objective

To compare the effectiveness of systematic review literature searches that use either generic or specific terms for health outcomes.

Design

Prospective comparative study of two electronic literature search strategies. The 'generic' search included general terms for health such as 'adolescent health', 'health status', 'morbidity', etc. The 'specific' search focused on terms for a range of specific illnesses, such as 'asthma', 'epilepsy', 'diabetes mellitus', etc.

Data sources

We searched Medline, Embase, the Cumulative Index to Nursing and Allied Health Literature (CINAHL), PsycINFO, and the Education Resources Information Center (ERIC) for studies published in English between 1992 and April 2010.

Main outcome measures

Number and proportion of studies included in the systematic review that were identified from each search.

Results

The two searches tended to identify different studies. Out of 41 studies included in the final review, only 3 (7%) were identified by both search strategies; 21 (51%) were identified by the generic search only; and 17 (41%) were identified by the specific search only. The two searches therefore identified a roughly equal share of the studies included in the review. While the generic search was particularly successful at

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identifying studies with multiple health outcomes, the specific search identified more single outcome studies.

Conclusions

Electronic literature searches (ELS) are a vital stage in conducting systematic reviews and therefore have an important role in the scientific community's attempts to inform and improve policy and practice with the best available evidence. Future systematic reviews that involve multiple health outcomes should include both generic and specific health terms in their literature search. Based on our findings, choosing only one or the other of these strategies could lead to systematic reviews that miss important evidence and consequently risk misinforming practitioners and other decision-makers.

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Introduction

Electronic literature searches (ELS) are an essential stage in most systematic reviews.[1-2] As such, they have a crucial role in the scientific community's attempts to inform and improve policy and practice with the best available evidence.[3-4] Designing ELS can be challenging and it is widely recognized that specialist skills and knowledge, such as those provided by an information scientist, are important for best practice in this field.[1-3] A key challenge when conducting ELS is the need to screen out irrelevant evidence (specificity), whilst successfully identifying the relevant evidence (sensitivity). Search strategies that are too specific risk encouraging potentially harmful decisions based on the findings of reviews that have failed to identify important evidence. Search strategies that are too sensitive risk pointlessly lengthy and resource-intensive searches which may delay the availability of evidence syntheses to inform decisions, and/or represent an ineffective allocation of scarce resources.[3-5] Hence, there is a pressing need to learn more about how best to negotiate the competing demands of specificity and sensitivity.

Previous research exploring how to improve the effectiveness and efficiency of search strategies has tended to focus on issues such as how to optimize search outputs from 'frontline' electronic databases (i.e. databases that are frequently searched for systematic reviews of medical interventions such as Medline and Embase), and how to identify randomised control trials (RCTs).[6-12] This research focus may in part reflect the influence of the Cochrane Collaboration, which has helped to stimulate considerable interest in systematic reviews of clinical trials.[1]

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However, not all systematic reviews (nor indeed all Cochrane Reviews[13]) focus on RCTs of clinical interventions. Interest in broader, non-clinical systematic reviews has steadily increased within the social and public health sciences and other disciplines.[3 5] As some of these non-clinical reviews tackle relatively under-researched topics, they often combine a scoping and hypothesis testing function by asking relatively broad research questions that, for example, cover a range of outcomes (e.g. *what are the health impacts of intervention x?; what health outcomes are associated with risk-factor y?*).[14-27] Evidence-informed guidance on how to conduct searches for this broader range of systematic reviews is therefore an emerging priority.

There are few examples of research that can help guide information scientists and reviewers to develop efficient but effective search strategies for these broader / non-clinical systematic reviews. The research that is available illustrates how searches for such reviews can become lengthy and complex.[28] For example Greenhaulgh et al recommended the development of iterative search strategies to search for complex evidence (e.g. multiple study designs). Ogilvie et al suggested that cross-disciplinary reviews may necessitate searching databases across a range of disciplines rather than focusing on frontline health databases.[4]

From our own experiences of conducting systematic reviews of non-clinical, public health research, the authors of this paper can identify additional challenges that have led to large and complex ELS. For example, search terms that involve commonly used words are likely to identify large numbers of irrelevant papers and non-clinical public health reviews often rely on commonly used terms to describe everyday settings, activities and outcomes (e.g. ‘walking’, ‘obesity’, ‘stress’, ‘workplace health’, ‘health

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promotion', and 'general health'). In comparison, an ELS for a clinical review will often involve very specific medical terminology that can help to focus the search on papers relevant to a particular field.[3]

Furthermore, the identification of studies for clinical reviews typically requires three lists of search terms (sometimes known as filters because they filter out unwanted studies): (i) terms that define the population who will receive the intervention; (ii) terms to describe the intervention; and (iii) terms to identify a particular study design (typically a filter for RCTs).[1] Systematic reviews that focus on a more general population sample, have no intervention, and/or are not limited to a single study design, lack one or more of these key filters, and so result in a less specific ELS.

All these challenges increase the chances of a search becoming lengthy and cost-ineffective. In such circumstances, reviewers may look for alternative means of increasing search specificity but there has been relatively little guidance on how this can be achieved without compromising sensitivity.

Filtering searches by health outcomes is one commonly used technique for increasing specificity in broader reviews.[14-24] However, if a review question is broad enough to include multiple health outcomes it is not obvious how a health outcome filter can best accommodate this breadth of scope. Some reviews have used generic health terms (e.g. 'health', 'illness', 'morbidity') to search for evidence that includes a range of health outcomes.[14-17] In other cases, reviewers have used more specific search terms to identify a number of diseases or symptoms considered to be of particular relevance to the review question.[18-21] Both approaches may be hypothesized to have

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risks. Generic search terms may either be too inclusive (virtually every study on Medline is about 'health') or may miss studies that only use more specialist vocabulary to describe a particular illness. Specific search terms are problematic if the reviewers want to avoid pre-specifying which health outcomes are relevant to the review (e.g. scoping reviews). Some reviews combine both generic and specific approaches[22-24], but the extent to which this either adds value to the search or merely adds to the workload is not known.

We know of no study that has compared the relative merits of ELS strategies that focus on either generic terms for health, or specific terms for particular health issues or illnesses. Nor do we know of any evidence to help reviewers determine whether these two approaches are likely to identify a similar or a different set of publications. When the authors of this paper recently conducted a systematic review that included multiple health outcomes, we felt that guidance on this issue would have been helpful. As there was an absence of evidence upon which to base such guidance, we ran two separate literature searches for our review: one that included generic health terms and one that used more specific health terms. Our aim was to see which approach was most effective in identifying studies that were included in the final review.

Hence, we examined whether the included studies tended to be identified from the generic search only, the specific search only, or both searches. We also explored efficiency by comparing the size of the searches (i.e. the number of references initially identified from the ELS – sometimes referred to as the number of 'hits') for each approach. Finally, we explored the extent to which the 'generic search' and the 'specific search' identified studies with different or similar types of health outcome.

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Methods

This paper focuses on one specific, but crucial, stage of a systematic review: the development of filters for the electronic literature search. We developed two contrasting strategies for searching electronic databases and compared their effectiveness in identifying studies for a specific systematic review. The systematic review itself is summarised in Panel 1, and described more fully in the publically available Protocol document (available as a supplemental document online), and the full report of the review which will be published separately to this methodological paper.

Panel 1. Summary of the systematic review used as the basis of this methodological study.

Title: How robust is the evidence of an emerging or increasing female excess in physical morbidity rates between childhood and adolescence? Results of a systematic literature review.

Hypothesis: That the incidence of physical morbidity amongst children tends to be higher amongst males in pre-adolescent childhood, but this male excess is replaced by an emergence of higher rates in females during the transition to adolescence.

Inclusion / Exclusion criteria: These criteria are summarised using the PICOS statement below. For full details of the inclusion and exclusion criteria, see the protocol: supplemental document).

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Included studies must have the following characteristics

Population: males and females between the ages of 4 and 17;

Intervention: none;

Comparator: sex and age (at least two age-groups);

Outcome: gender patterning, by age, in measures of physical morbidity;

Study design: longitudinal, cross-sectional and repeat cross-sectional studies (including analysis of study-specific data or routinely collected data).

Methods: The systematic review included methodological components suggested by the PRISMA guidelines (e.g. protocol, literature search, study selection, flow chart, data extraction, critical appraisal and synthesis), and was designed to meet the standards of that guidance. More details are provided in the protocol.

Data sources and search strategy

We searched five electronic databases (Medline, Embase, the Cumulative Index to Nursing and Allied Health Literature (CINAHL), PsycINFO, and the Education Resources Information Center (ERIC)) for studies published in English between 1992 and the date of search (April 2010). As it was our intention is to update a previous review conducted around twenty years previously,[29] we searched for studies published from 1992 to the present.

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We searched each database twice: once using 'generic' health subject headings and keywords and once using 'specific' subject headings and keywords relating to the health conditions we had selected for review. In this paper, we refer to these searches as the 'generic search' and the 'specific search'. The generic search included the terms: health status, attitude to health, health attitudes, health surveys, child health, adolescent health, health status indicators, symptoms, morbidity, health complaints, general health questionnaire, well being, self report, and wellness. The specific search included the terms: asthma, epilepsy, diabetes mellitus, primary headache, and migraine. The specific search terms related to health conditions that we judged to be relevant to the research question and for which we were likely to find evidence. We based this judgement on an initial scoping of the literature and an earlier review of this topic[29]. The precise search strategy differed between databases if different search facilities and search engines made it necessary to adapt our approach. Specific details of our searches are presented in the review protocol (see supplemental document).

Study Selection

One reviewer (AM) screened all the publications identified by both literature searches to exclude obviously irrelevant titles. The remaining (i.e. not excluded) publications were retrieved and, on reading, AM screened out those that were clearly not eligible for inclusion in the review (see Figure 1, 'First Sift'). Studies of uncertain eligibility were checked by two other reviewers (KH and HS) so that a decision to exclude or retrieve the full paper could be reached (see Figure 1, 'Second Sift'). Some retrieved papers were excluded at the initial reading ('Third Sift'), whilst others were excluded at the data extraction and appraisal stage (based on agreement from all the reviewers).

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At this final stage we also excluded studies that only explored asthma-related outcomes after finding a review that already applied our research question to this health outcome.

Comparing the two searches

We produced a series of Venn diagrams for each stage of the review process, showing the number of studies identified only by the specific literature search, the number identified only by the general literature search, and the number identified by both searches (see Figure 1). The purpose was to see if the two searches identified similar or different sets of documents. Studies that were included in the final review were then tabulated in more detail to help us assess whether there was any systematic variation in the types of health outcome identified by the different searches.

Results

Figure 1 shows for each stage of the review the number of studies identified exclusively by either the specific or the generic search, and (in each intersect) the number of studies identified by both searches.

The diagram makes two points apparent. Firstly, there was relatively little duplication between the two searches. For example, out of the 11509 total hits identified from both literature searches, only 413 (3.6%) were duplicates between the two searches. Throughout each stage of the study selection process, duplication between the two searches remained low, so that only three (7.3%) of the 41 studies selected for final

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inclusion in the review were identified by both search strategies (further details of the 41 included studies are available in a supplemental document).

Secondly, we note that the specific search led to less than half the number of initial hits, compared to the generic search (3299 vs. 8210, respectively), but both searches identified a similar number of studies included in the final review (17 vs. 21, and 3 duplicates).

Table 1: Studies included in the systematic review (n = 41) by summary health outcomes and by the search strategy used to identify each study.

Outcomes	Generic Search	Specific Search	Both Searches
Abdominal Pain		1	
Back pain	2		
Diabetes	1	6	
Epilepsy		3	
Headache	2	7	1
General physical health / wellbeing	5		
Multiple physical health outcomes*	11		2
Total (for each search)	21	17	3

* A range of health outcomes were included in these studies: usually involving measures of general health and bodily pain.

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We then examined the 41 studies included in the final review, categorizing them by the health outcomes each one investigated (see Table 1). The findings suggest some systematic differences in the health outcomes of studies identified using each of the two search strategies. The specific search tended to be more successful at identifying studies that focused on a single type of health outcome (i.e. those that related to the search terms), but less successful at identifying studies that explored general health or a mixture of different health outcomes. The opposite was found for the generic search strategy, which tended to be more successful at identifying studies with multiple health outcomes.

Discussion

We have compared two strategies for conducting an electronic literature search for a systematic review. One strategy used generic health terms, whilst the other used more specific health terms. The purpose was to explore whether literature searches with a relatively broad inclusion criteria (in terms of health outcomes) are better served by generic or specific health terms, or whether both are needed.

We found that both specific and generic health terms were necessary. The results were very striking: had we only used generic health terms in our search we would have missed around half the studies that we finally included in the review. Likewise, focusing exclusively on specific health terms in the literature search would have failed to identify around half the included papers. This represents a serious 'loss' of data (or, more correctly, a failure to find data) that would have compromised the credibility and accuracy of our review's findings.

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Furthermore we have found that the evidence identified by the two search strategies tended to be systematically different. The specific search tended to miss studies with general or multiple health outcomes, whilst the generic search tended to miss studies with single, specific health outcomes. This may appear intuitive, but we contend that the finding is actually surprising. It suggests, for example, that studies that look specifically at young people's diabetes, epilepsy and headache tend not to be identifiable by search terms such as "health status", "health surveys", "child health", "adolescent health", "health status indicators", "symptoms", "morbidity", "health complaints", etc. It also suggests that some studies that, for example, included headache as one of a number of different health outcomes may be identified by a search strategy that includes generic health terms, but could be missed by an ELS that specifically focuses on the term 'headache.'

This finding is at odds with what some authors of this paper initially expected. Prior to our exploring this issue, the authors assumed that the generic health search would identify the vast majority of included studies whilst the specific search would mainly identify a subset of those studies. If other systematic reviewers also make this assumption, then their reviews are at risk of being based on poor quality (highly insensitive) searches.

Strengths and limitations

We have conducted a prospective comparative study of two electronic literature search strategies that have been field tested whilst we conducted a systematic review. This kind of study is uncommon and hence novel. The prospective, comparative design is a key strength. The information scientist who advised on both search

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strategies, and the researcher who led the comparative study are experienced in conducting systematic reviews of broader public health topics and have a good understanding of the challenges involved with reviews of this kind.

The main limitation of this study is that it is based on a single review. There is some existing evidence that the effectiveness of different search strategies may vary depending on the subject of the review[5], so it is obviously worth testing our findings in the context of other reviews. The authors have also assumed that a health outcome filter was appropriate for their review, but we are aware that this assumption is open to challenge. Had our search strategy simply missed out the health outcome filter altogether there would have been no chance of any study being wrongly excluded due to a failure to electronically identify relevant health outcomes. This would have increased search sensitivity but, for reasons discussed in the introduction, it would also have created problems related to insufficient specificity: i.e. the search would have expanded greatly in size (and note that even with our health outcome filters, our initial search identified well in excess of 10,000 hits).

Implications and conclusions

Literature searching has a vital role to play in evidence-informed policy and practice, and it is plausible to theorise a direct pathway by which a poor search may lead to harmful decisions. Conducting research that may assist information scientists and reviewers to improve their search strategies should therefore be a priority. Such research can be nested within the processes of conducting systematic reviews: from our own experience this requires minimal additional resource to the cost of the overall review and can therefore be considered an inexpensive way of conducting useful

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research in an important field. We therefore hope that other reviewers will make use of similar opportunities to explore how best to optimise electronic searching.

In light of our findings, we recommend that future systematic reviews of topics that involve multiple health outcomes include both generic and specific health terms in their literature search (if a health outcome filter is considered necessary). Choosing only one or the other of these strategies could, based on our findings, lead to systematic reviews that miss half the available evidence.

Authors' interests

(1) All authors have completed the Unified Competing Interest form at www.icmje.org/coi_disclosure.pdf (available on request from the corresponding author) and declare that ME, AM, HS, KH have support from the Medical Research Council and Chief Scientist Office for the submitted work; (2) ME, AM, HS, KH have no relationships with any company that might have an interest in the submitted work in the previous 3 years; (3) their spouses, partners, or children have no financial relationships that may be relevant to the submitted work; and (4) ME, AM, HS, KH have no non-financial interests that may be relevant to the submitted work.

Ethics

As this study focused on searching for literature that was already in the public domain, it involved no patients, consents, nor other issues that required formal approval from an ethics committee.

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Data sharing

The review protocol (including search strategies) and a list of studies included in the final review are available in the supplemental documents submitted with this article. Further data related to the searches are available from the corresponding author at matt@sphsu.mrc.ac.uk.

Details of contributors

ME helped plan and conduct the study, analyse the findings, led on writing the manuscript and is guarantor for the study. AM, HS and KH helped plan the study and conduct the study, analyse the findings and provide content and comments on the manuscript. All authors, external and internal, had full access to all of the data (including statistical reports and tables) in the study and can take responsibility for the integrity of the data and the accuracy of the data analysis.

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What is already known on this subject

Literature searching has a vital role to play in systematic reviews that inform policy and practice. Evidence to help reviewers conduct effective literature searches tends to be based on reviews of randomised controlled trials of clinical interventions. There is relatively little evidence to help guide literature searches for other types of review, including more broadly focused reviews relevant to public health, epidemiology and health improvement.

What this study adds

Whilst conducting a systematic review that included a range of health outcomes, we compared two electronic literature search strategies – one that used generic terms for (ill)health and another than used terms for specific illnesses. Our findings suggest a need for combining generic and specific search strategies when conducting systemic reviews involving multiple health outcomes. Systematic review searches that use *either* only generic *or* only specific search terms for health outcomes risk missing out a large proportion of the relevant studies, which may lead to erroneous conclusions that misinform policy and practice.

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References

1. Higgins JPT, Green S. *Cochrane Handbook for Systematic Reviews of Interventions Version 5.0.2 [updated September 2009].* : The Cochrane Collaboration, 2009.
2. NHS Centre for Reviews and Dissemination. *Systematic Reviews: CRD's Guidance for Undertaking Reviews in Healthcare* York: CRD University of York, 2009.
3. Petticrew M, Roberts H. *Systematic Reviews in the Social Sciences: A practical guide.* Oxford: Blackwell Publishing, 2006.
4. Ogilvie D, Hamilton V, Egan M, Petticrew M. Systematic reviews of health effects of social interventions: 1. Finding the evidence: how far should you go? *J Epidemiol Community Health* 2005;59:804-08.
5. Bambra C. Real world reviews: a beginner's guide to undertaking systematic reviews of public health policy interventions. *J Epidemiol Community Health* 2011;65:14-19.
6. Glanville JM, Lefebvre C, Miles JN, Camosso-Stefinovic J. How to identify randomized controlled trials in MEDLINE. *J Med Libr Assoc* 2006;130-136:130-36.
7. Golder S, McIntosh HM, Duffy S, Glanville J, Centre for Reviews and Dissemination and UK Cochrane Centre Search Filters Design Group. Developing efficient search strategies to identify reports of adverse effects in MEDLINE and EMBASE. *Health Info Libr J* 2006;23:3-12.
8. Montori VM, Wilczynski NL, Morgan D, Haynes RB. Optimal search strategies for retrieving systematic reviews from Medline: analytical survey. *BMJ* 2005;330:68.
9. Wong SS, Wilczynski NL, Haynes RB. Developing optimal search strategies for detecting clinically sound treatment studies in EMBASE. *J Med Libr Assoc* 2006;94:41-47.
10. Dickersin K, Scherer R, Lefebvre C. Identifying relevant studies for systematic reviews. *BMJ* 1994;309:1286-91.
11. Eisinga A, Siegfried N, Clarke M. The sensitivity and precision of search terms in Phases I, II and III of the Cochrane Highly Sensitive Search Strategy for identifying reports of randomized trials in MEDLINE in a specific area of health care - HIV/AIDS prevention and treatment interventions. *Health Info Libr J* 2007; 24:103-09.
12. Royle P MR. Literature searching for randomized controlled trials used in Cochrane reviews: rapid versus exhaustive searches. *Int J Technol Assess Health Care* 2003;19:591-603.
13. Joyce K, Pabayo R, Critchley JA, Bambra C. Flexible working conditions and their effects on employee health and wellbeing. *Cochrane Database Syst Rev*, 2010.
14. Porta D, Milani S, Lazzarino A, Perucci C, Forastiere F. Systematic review of epidemiological studies on health effects associated with management of solid waste. *Environ Health* 2009;8(60):doi:10.1186/476-069X-8-60.
15. Uscher-Pines L. Health effects of relocation following disaster: a systematic review of the literature. *Disasters* 2009;33(1):1-22.
16. D'Onise K, Lynch J, Sawyer M, McDermott R. Can preschool improve child health outcomes? A systematic review. *Soc Sci Med* 2010;70(9):1423-40.
17. Egan M, Petticrew M, Ogilvie D, Hamilton V. New Roads and Human Health: A Systematic Review. *Am J Public Health* 2003;93(9):1463–71.

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18. Dangour A, Aikenhead A, Hayter A, Allen E, Lock K, Uauy R. Comparison of putative health effects of organically and conventionally produced foodstuffs: a systematic review: Report for the Food Standards Agency. London: Nutrition and Public Health Intervention Research Unit London School of Hygiene & Tropical Medicine, 2009.
19. Vickers AJ. Can acupuncture have specific effects on health? A systematic review of acupuncture antiemesis trials. *J R Soc Med* 1996;89:303-11.
20. Daykin N, Orme J, Evans D, Salmon D, McEachran M, Brain S. The Impact of Participation in Performing Arts on Adolescent Health and Behaviour: A Systematic Review of the Literature. *J Health Psychol* 2008;13:251-64.
21. Rogers G, Elston J, Garside R, Roome C, Taylor R, Younger P, et al. The harmful health effects of recreational ecstasy: a systematic review of observational evidence. *Health Technol Assess* 2009;13(6):DOI: 10.3310/hta13060.
22. Passchier W, Knottnerus A, Albering H, Walda I. Public health impact of large airports. *Rev Environ Health. 2000 Jan-Jun;15(1-2):* 2000;15(1-2):83-96.
23. Egan M, Petticrew M, Ogilvie D, Hamilton V, Drever F. "Profits before people"? A systematic review of the health and safety impacts of privatising public utilities and industries in developed countries. *J Epidemiol Community Health* 2007;61:862-70.
24. McCartney G, Thomas S, Thomson H, Scott J, Hamilton V, Hanlon P, et al. The health and socioeconomic impacts of major multi-sport events: systematic review (1978-2008). *BMJ* 2010;340:c2369.
25. Paradies Y. A systematic review of empirical research on self-reported racism and health. *Int J Epidemiol* 2006;35(4):888-901.
26. Vartanian LR, Schwartz MB, Brownell KD. Effects of soft drink consumption on nutrition and health: a systematic review and meta-analysis. *Am J Public Health* 2007;97(4):(4):667-75.
27. Bambra C, Whitehead M, Sowden A, Akers J, Petticrew M. "A hard day's night?" The effects of Compressed Working Week interventions on the health and work-life balance of shift workers: a systematic review. *J Epidemiol Community Health* 2008;62(9):764-77.
28. Greenhalgh T, Peacock R. Effectiveness and efficiency of search methods in systematic reviews of complex evidence: audit of primary sources. *BMJ* 2005;331:1064-65.
29. Sweeting H. Reversals of fortune? Sex differences in health in childhood and adolescence. *Soc Sci Med* 1995;40(1):77-90.

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Figure 1. Review study selection flow chart: studies identified by the 'generic' search only (purple circle); 'specific' search only (light blue circle); and by both searches (dark blue intersect).

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How robust is the evidence of an emerging or increasing female excess in morbidity rates between childhood and adolescence?

Review protocol

October 2011

Alice MacLean, Matt Egan, Helen Sweeting, Joy Adamson and Kate Hunt.

MRC Social & Public Health Sciences Unit

4 Lilybank Gardens

Glasgow

G12 8RZ

Tel: 0141 357 3949

e-mail: alice@sphsu.mrc.ac.uk

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BACKGROUND

This protocol is an updated version of the original, which was written in March 2010 and set out our plans for conducting the review. Any significant changes made to the review process, between the writing of the first protocol and this updated version, are highlighted using footnotes.

In 1995, Social Science & Medicine published a narrative review of research findings on sex differences in health among children and adolescents (Sweeting 1995). By examining and summarising the findings from a broad range of research on the physical health, psychological well-being and health service utilisation of children and adolescents, with a focus on the 7 to 15 age-range, Sweeting's review provides evidence of a 'gender reversal' in the distribution of ill-health across the transition from childhood to adolescence. Gender and age differences in rates of asthma are referred to in the review as one example of this reversal in physical health. It documents that in children less than 10 years old, rates of asthma are highest among boys but by adolescence boys' and girls' rates converge and after this time higher rates of asthma are often found among girls. A similar picture is presented in relation to psychological well-being; overall rates of psychiatric disorders are more prevalent amongst boys until early adolescence, however the referral rates for girls with psychiatric disorders have been found to rise after 12 years of age and exceed those of boys by age 15-16. As well as demonstrating an overall emergence of excess morbidity in females over early-mid adolescence, Sweeting's review highlighted a need for longitudinal studies to chart sex differences in physical and

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psychological health, as well as illness behaviours and beliefs, across the transition from childhood to adolescence.

Since the publication of Sweeting's (1995) narrative review, substantial research evidence has been found to suggest that higher rates of psychological morbidity found among males in childhood are replaced by an emergence of higher rates in females during the transition to adolescence (Petersen, Sarigiani et al. 1991; Cohen, Cohen et al. 1993; Schraedley, Gotlib et al. 1999; Ge, Conger et al. 2001; Marcotte, Fortin et al. 2002; Bennett, Ambrosini et al. 2005). This pattern has also been reported for asthma prevalence (Venn, Lewis et al. 1998; Nicolai, Pereszlenyiova-Bliznakova et al. 2003; Sears, Greene et al. 2003). Indeed, a number of reviews have synthesised and documented this evidence and have contributed to an established recognition of an emerging/increasing female excess in rates of psychological disorders (Nolen-Hoeksema and Girgus 1994; Hankin and Abramson 1999; Cyranowski, Frank et al. 2000; Shibley Hyde, Mezulis et al. 2008) and asthma[1] (Zannolli and Morgese 1997; Postma 2007; Almqvist, Worm et al. 2008).

However, there are no reviews, to our knowledge, which have been conducted with the aim of investigating the extent to which there is evidence of an emerging/increasing female excess in relation to other, or a range of, physical morbidity outcomes. This is surprising given that in recent decades several large-scale European and North American surveys of children and adolescents aged

[1] Originally we had planned to include asthma and psychological symptoms and conditions in this review. However, after identifying recent reviews which had explored the gender patterning of prevalence rates by age in relation to these health outcomes, we subsequently excluded studies which only presented data on asthma and psychological health outcomes.

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between 11 and 16 have reported comparable patterns of an overall emerging or increasing excess in girls' rates of reporting both psychological and physical symptoms (Eiser, Havermans et al. 1995; Eminson, Benjamin et al. 1996; Klepp, Aas et al. 1996; Haugland, Wold et al. 2001; Hetland, Torsheim et al. 2002; Sweeting and West 2003; Torsheim, Ravens-Sieberer et al. 2006). Indeed, the emergence of female excess morbidity during adolescence has been described as a central feature of adolescent health in 'a large proportion of the world's industrialised countries' (Torsheim, Ravens-Sieberer et al. 2006, p.823). Therefore, assessing the amount and quality of evidence suggesting that morbidity rates for a range of outcomes vary by gender according to age, may take us closer to explaining the emergence of higher reported morbidity in females.

REVIEW AIMS

This review aimed to investigate the extent to which research has found evidence of an emerging/increasing female excess in relation to physical morbidity rates across childhood and adolescence.

Our objectives, in terms of the PICOS statement, were as follows:

Population: males and females between the ages of 4 and 17

Intervention: none

Comparator: gender and age (at least two age-groups)

Outcome: gender patterning, by age, in measures of physical morbidity

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Study design: longitudinal, cross-sectional and repeat cross-sectional studies (including analysis of study-specific data or routinely collected data).

METHODS

Searching

The following bibliographic databases were searched: Medline; Embase; CINAHL (Cumulative Index to Nursing & Allied Health Literature); PsycINFO; and ERIC (Education from U.S. Department of Education, & Institute of Education Sciences). Academic research was targeted and no grey literature was included in the review. Searches were limited to articles published in English between 1992 and the date of search (April 2010). As it was our intention to update Sweeting's 1995 narrative review (written in 1994), we predicted that searching for articles published in the three years leading up to its publication would enable us to retrieve any relevant studies which may have been in the publication process at the same time as, and therefore not included in, the 1995 review.

The precise search strategy differed slightly between databases if different search facilities and search engines made it necessary to adapt our approach (see Appendix 1 and 2 for the full search strategies used in each database). Our searches included three groups of terms:

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1) Terms to identify the target group

Adolescent; adolescence; adolesc*; child; child, preschool; children; early adolescents; late adolescents; preschool child; minor*; pupil*; school child*; teenage*; young children; young pers*.

2) *Terms to identify the comparator*

Gender; gender differences; human sex difference; sex; sex distribution; sex factors.

3) Terms to identify health measures

We searched each database twice; once using *generic* health subject headings and keywords and once using *specific* subject headings and keywords relating to physical symptoms and conditions common in childhood[2].

a) Generic search terms: adolescent health; attitude to health; child health; general health questionnaire; health; health attitudes; health complaints; health status; health status indicators; health survey; morbidity; self-report; symptoms; well-being; wellness.

b) Specific search terms: diabetes mellitus; epilepsy; headache; headache disorders, primary; migraine; primary headache.

[2] Through the generic search we aimed to retrieve studies which explored general measures of physical health (e.g. self-rated health) or a mixture of physical morbidity outcomes (e.g. symptom prevalence rates). The specific search was intended to identify studies that reported on the prevalence of particular health conditions that are common in childhood. In conducting both searches, we hoped to achieve a wide coverage of the research conducted in relation to physical morbidity during childhood and adolescence.

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Data management

A 'search diary' was kept, which detailed the names of the databases searched, the search terms used and the search results (see Appendix 1 and 2). The results of each search were exported to an Endnote database, along with details of which database they were imported from and whether they were the results of the generic or specific search. Titles and abstracts were screened by one reviewer and inclusion/exclusion decisions recorded on the Endnote database. To check for consistency in screening, a random sample of abstracts was screened by two other reviewers and their decision to include or exclude was checked against the main reviewer's decision. Retrieved studies were filed according to inclusion/exclusion decisions.

Inclusion and exclusion criteria

The following inclusion and exclusion criteria were applied to all studies to determine their relevance to the review:

1) Age of participants

As the review focussed on child and adolescent health, studies which included participants between 4 and 17 years old were included. Studies of babies and toddlers (aged 0-3 years) were excluded on the grounds that they are not able to communicate their symptoms verbally in the same way as older children. Studies of those aged 18 years and over were classed as adult studies and therefore not included. As we aimed to look at change in prevalence rates according to age, studies were required to present prevalence data for at least two age-groups within

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the 4-17 range. Studies which only presented data for one age-group were excluded. Studies were included if they presented data in age-bands of no wider than five years (e.g. 11-14). Studies using age-bands wider than five years were excluded on the grounds that this would prevent us from looking at change in prevalence rates according to age. Studies which used age-bands that included some participants within the 4-17 age-range, such as 0-4 or 15-19, were included. However, if half or more of an individual age-band was not within our age-range, that age-band was excluded from our analyses. For example, 0-4 age-bands were excluded from analysis on the assumption that the majority of participants within that sample would be under four years of age. Often this did not result in the exclusion of studies as they presented prevalence data for at least a further two age-groups.

2) Sex of participants

The aim of the review was to assess the evidence for an emerging/increasing female excess in morbidity rates, so studies which presented data in relation to both males and females were included. Studies which presented data only in relation to either males or females were excluded.

3) Study design

Empirical studies which used quantitative data collection and analysis methods were included. Longitudinal, cross-sectional, repeat cross-sectional and studies which have analysed routine data (e.g. hospital records) were included. Studies which employed qualitative data collection and analysis methods were not included. Studies which presented only parent-report data were also excluded.

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4) Health measures

Studies which presented prevalence data on health measures (e.g. symptom and morbidity rates; health status; incidence of chronic illnesses in childhood etc.) were included in the review. Studies reporting only lifetime prevalence rates were not included because we were interested in current or recent (i.e. within last year) measures of physical morbidity. Studies about injuries or accidents were not included. Studies about health behaviours and symptoms resulting from health behaviours (e.g. impact of alcohol use on depressive symptoms) were also excluded. Studies focussing on dental health were excluded, as were studies which focussed on obesity rates and those which explored rates of symptoms which are the result of traumatic events (e.g. abuse).

5) Countries

Studies from current EU countries as well as the USA, Canada, Australia and New Zealand were included on the basis that their contextual similarity would aid comparison. Studies from all other countries were excluded.

The above criteria were applied to the titles and abstracts of the articles identified by the literature searches. Hard copies were obtained of all articles which met the inclusion criteria. In cases where inclusion or exclusion could not be determined from titles and abstracts, full papers were retrieved and checked. Each article was labelled in Endnote as to whether it was included or excluded and the number of articles included and excluded at the various stages of the review was recorded systematically.

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Data extraction

Data were extracted by one reviewer and three reviewers each independently extracted a sample of studies. Extraction forms were compared across reviewers. The following data were extracted:

1) **Publication details**: author; title; journal; date; primary focus; stated aims.
2) **Focus on emerging/increasing female excess:** mention or not of sex differences/similarities/'gender reversal' in introduction, results or discussion; explanations offered for changes in sex differences with age.
3) **Study details:** methods; sample (source, size, age range and age-groups; representativeness; response rate/completeness); primary outcomes; questions/instruments.
4) **Key data:** any figures for outcomes by sex and age (e.g. prevalence rate/incidence rate, both adjusted and unadjusted, figures extracted as reported in paper (means, OR, RR etc.) with as much detail as possible (95% confidence intervals, chi-square etc.)).

Assessment of methodological quality

Studies were critically appraised by one reviewer using the criteria below which were agreed by all reviewers. A quality index was developed for each criterion which ranged from 2 (lower potential for bias) to 0 (higher potential for bias). Studies were each given an indicative score for quality. Repeat cross-sectional/cross-sectional and routine data studies were scored out of a maximum of 12 and, due to the extra criterion for attrition rate, longitudinal studies were scored out of 14.

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Sample size

2 - Every age and gender sub-group is comprised of at least 100 participants.

1 - Every age and gender sub-group is comprised of at least 50 participants.

0 - <50 in any age and gender sub-group, or data not given.

Large/multi-site population

2 - International, national or statewide (e.g. as in a USA state) study, including multi-site studies in which the sites are spread across international, national or statewide areas.

1 - Local multi-site studies (e.g. same city, town/district, or villages within the same region).

0 – Single-site study (e.g. one school).

Age-ranges covered

2 - Three or more age points that include under 12 years of age and 12 years or older.

1 - Two age points that include under 12 years of age and 12 years or older.

0 - Age points do not compare those under 12 years of age with those aged 12 years or older.

Selection bias

2 - 80-100% response at baseline *or* routine data that covers at least 80% of population.

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1 - 60-79% response at baseline (or routine data coverage) and non-response confounding explored and found not to have a substantial gender or age bias *or* routine data covers 60-79% and no reason to assume age/gender bias in coverage.

0 - Response (or routine data coverage) less than 60%; or less than 80% with evidence of a substantial gender or age bias in attrition; or if non-response is not explored.

Outcome

2 - Physical examination by trained professional.

1 - Self-complete questionnaire using an established/validated questionnaire.

0 - Unvalidated questionnaire or questionnaire designed for study and there is no comment on validation.

Analysis and data reporting

2 - Use odds ratios/incidence rate ratio and 95% confidence intervals to determine whether there is a significant gender-by-age interaction associated with morbidity rates (or sufficient data to calculate ORs, IRRs and CIs).

1 - Use of alternative (to those above) methods of determining gender-by-age interactions associated with morbidity rates (e.g. continuous data or visual data without confidence intervals).

0 - Data on age, gender or morbidity compromised by unclear reporting or missing data.

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Attrition (longitudinal studies only)

2 - Final response is 80%-100% of baseline response.

1 - Final response is 60%-79% of baseline response and attrition confounding explored and found not to have a significant gender or age bias or a bias related to baseline health outcomes.

0 - Final response is <60% of baseline response.

Synthesis

As a meta-analysis was not possible, owing to the heterogeneity of studies, a narrative synthesis method was employed. The studies were grouped by symptoms and conditions as follows: self-assessed health; symptoms (abdominal pain; back pain; dizziness; headache; sleeping difficulties/tiredness); conditions (migraine; diabetes mellitus; epilepsy). Where data were available, odds ratios were calculated (with males serving as the reference group) and studies were tabulated to aid comparison.

DISSEMINATION

The findings from the review were written up and submitted for publication to an international public health journal. We have so far presented the findings at two national conferences.

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Appendix 1 – Specific search diary

Medline (Ovid interface)

06/05/2010

(child, preschool or child or adolescent).sh. and (sex factors or sex distribution).sh. and (asthma or epilepsy or headache disorders, primary or diabetes mellitus).sh.

limit to (english language and humans and yr="1992 -Current")

(.sh. = MeSH subject headings)

Results – 1426

Embase (Ovid interface)

06/05/2010

(child or school child or adolescent or preschool child).sh. and (sex difference or gender).sh. and (asthma or primary headache or migraine or diabetes mellitus or epilepsy).sh.

limit to (human and English language and yr="1992 -Current")

(.sh.= subject headings)

Results – 1526

CINAHL (Cumulative Index to Nursing and Allied Health) **(EBSCO Host interface)**

07/05/2010

(adolescence or child or child, preschool).sh. and (sex factors).sh. and (asthma or diabetes mellitus or headache or epilepsy).sh.

limit to (english language and yr="1992 -Current")

For peer review only

For peer review only - http://bmjopen.bmj.com/site/about/guidelines.xhtml

(.sh.= word in subject heading)

Results – 498

PsycINFO (EBSCO Host interface)

07/05/2010

(child* or adolesc* or young pers* or teenage* or pupil* or school child* or minor*).kw. and (human sex differences or sex).sh. and (asthma or diabetes mellitus or headache or epilepsy).sh.

limit to (english language and yr="1992 -Current")

(.kw. – keywords, .sh.= exact subjects)

Results – 38

ERIC (Education from US Department of Education, and Institute of Education Sciences) **(Ovid interface)**

07/05/2010

((children or young children or adolescents or early adolescents or late adolescents).sh. or (pupil* or school child* or minor*).ab.) and ((sex or gender differences).sh. or (sex or gender).ab.) and ((asthma or headache or migraine).ab. or (diabetes or epilepsy).sh.)

limit to (english language and yr="1992 -Current")

(.sh.= ERIC subject headings, .ab. = abstract)

Results – 22

Specific search total hits – 3510

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Unique hits - 2622

For peer review only

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Appendix 2 – Generic search diary

Medline (Ovid interface)

16/04/2010

(child, preschool or child or adolescent).sh. and (sex factors or sex distribution).sh. and (health status or attitude to health or health surveys or mental health[3]).sh.

limit to (english language and humans and yr="1992 -Current")

(.sh. = MeSH subject headings)

Results – 3587

Embase (Ovid interface)

19/04/2010

(child or school child or adolescent or preschool child).sh. and (sex difference or gender).sh. and (adolescent health or health survey or health status).sh.

limit to (human and English language and yr="1992 -Current")

(.sh.= subject headings)

Results – 2652

CINAHL (Cumulative Index to Nursing and Allied Health) **(EBSCO Host interface)**

19/04/2010

(adolescence or child or child, preschool).sh. and (sex factors).sh. and (health status or health status indicators or attitude to health or symptoms or morbidity or child health or adolescent health).sh.

limit to (english language and yr="1992 -Current")

[3] Note: following this initial search, the decision was made to focus the review on physical rather than mental health.

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(.sh.= word in subject heading)

Results – 1467

PsycINFO (EBSCO Host interface)

19/04/2010

(child* or adolesc* or young pers* or teenage* or pupil* or school child* or minor*).kw. and (human sex differences or sex).sh. and (health or health attitudes or health complaints or general health questionnaire or well being or self report or morbidity or symptoms).sh.

limit to (english language and yr="1992 -Current")

(.kw. – keywords, .sh.= subjects)

Results – 1136

ERIC (Education from US Department of Education, and Institute of Education Sciences) **(Ovid interface)**

(19/04/2010)

((children or young children or adolescents or early adolescents or late adolescents).sh. or (pupil* or school child* or minor*).ab.) and ((sex or gender differences).sh. or (sex or gender).ab.) and ((health or child health or adolescent health or well being or wellness).sh. or (morbidity or symptom*).ab.)

limit to (english language and yr="1992 -Current")

(.sh.= ERIC subject headings, .ab. = abstract)

Results – 593

For peer review only

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Generic search total hits – 9435

Unique hits - 8623

For peer review only

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References

Almqvist, C., M. Worm, et al. (2008). "Impact of gender on asthma in childhood and adolescence: a GA2LEN review." Allergy **63**(1): 47-57.

Bennett, D. S., P. J. Ambrosini, et al. (2005). "Gender differences in adolescent depression: Do symptoms differ for boys and girls?" Journal of Affective Disorders **89**: 35-44.

Cohen, P., J. Cohen, et al. (1993). "An epidemiological study of disorders in late childhood and adolescence: I. Age and gender-specific prevalence." Journal of Child Psychology and Psychiatry **34**(6): 851-867.

Cyranowski, J. M., E. Frank, et al. (2000). "Adolescent onset of the gender difference in lifetime rates of major depression." Archives of general psychiatry **57**: 21-27.

Eiser, C., T. Havermans, et al. (1995). "The emergence during adolescence of gender differences in symptom reporting." Journal of Adolescence **18**(3): 307-315.

Eminson, M., S. Benjamin, et al. (1996). "Physical symptoms and illness attitudes in adolescents: An epidemiological study." Journal of Child Psychology and Psychiatry **37**(5): 519-528.

Ge, X., R. D. Conger, et al. (2001). "Pubertal transition, stressful life events, and the emergence of gender differences in adolescent depressive symptoms." Developmental Psychology **37**(3): 404-417.

Hankin, B. L. and L. Y. Abramson (1999). "Development of gender differences in depression: description and possible explanations." Annals of Medicine **31**(6): 372-379.

Haugland, S., B. Wold, et al. (2001). "Subjective health complaints in adolescence: A cross-national comparison of prevalence and dimensionality." European Journal of Public Health **11**(1): 4-10.

Hetland, J., T. Torsheim, et al. (2002). "Subjective health complaints in adolescence: dimensional structure and variation across gender and age." Scandinavian Journal of Public Health **30**(3): 223-230.

Klepp, K. I., H. N. Aas, et al. (1996). "Self-reported health problems among school pupils." Tidsskr Nor Laegeforen **116**: 2032-2037.

Marcotte, D., L. Fortin, et al. (2002). "Gender differences in depressive symptoms during adolescence: role of gender-typed characteristics, self-esteem, body image, stressful life events, and pubertal status." Journal of Emotional and Behavioural Disorders **10**(1): 29-43.

Nicolai, T., L. Pereszlenyiova-Bliznakova, et al. (2003). "Longitudinal follow-up of the changing gender ratio in asthma from childhood to adulthood: role of delayed manifestation in girls." Pediatric Allergy & Immunology **14**(4): 280-283.

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Nolen-Hoeksema, S. and J. S. Girgus (1994). "The Emergence of Gender Differences in Depression During Adolescence." Psychological Bulletin **115**(2): 424-443.

Petersen, A. C., P. A. Sarigiani, et al. (1991). "Adolescent Depression: Why More Girls?" Journal of Youth and Adolescence **20**(2): 247-271.

Postma, D. S. (2007). "Gender differences in asthma development and progression." Gender Medicine **4 Suppl B**: S133-146.

Schraedley, P. K., I. H. Gotlib, et al. (1999). "Gender Differences in Correlates of Depressive Symptoms in Adolescence." Journal of Adolescent Health **25**: 98-108.

Sears, M. R., J. M. Greene, et al. (2003). "A longitudinal, population-based, cohort study of childhood asthma followed to adulthood." New England Journal of Medicine **349**(15): 1414-1422.

Shibley Hyde, J., A. H. Mezulis, et al. (2008). "The ABCs of Depression: Integrating affective, biological and cognitive models to explain the emergence of the gender difference in depression." Psychological Review **115**(2): 219-313.

Sweeting, H. (1995). "Reversals of fortune? Sex differences in health in childhood and adolescence." Social Science & Medicine **40**(1): 77-90.

Sweeting, H. and P. West (2003). "Sex differences in health at ages 11, 13 and 15." Social Science & Medicine **56**(1): 31-39.

Torsheim, T., U. Ravens-Sieberer, et al. (2006). "Cross-national variation of gender differences in adolescent subjective health in Europe and North America." Social Science & Medicine **62**(4): 815-827.

Venn, A., S. Lewis, et al. (1998). "Questionnaire study of effect of sex and age on the prevalence of wheeze and asthma in adolescence." BMJ **316**(7149): 1945-1946.

Zannolli, R. and G. Morgese (1997). "Does puberty interfere with asthma?" Medical Hypotheses **48**(1): 27-32.

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Supplemental document: Studies that were included in the final review, identified from the generic and specific literature searches

Table 2: Studies identified by both the generic and specific searches

Author, date	Title	Health outcome
Gordon et al, 2004[1]	Prevalence of reported migraine headaches in Canadian adolescents.	Migraine.
Petersen et al, 2003[2]	High prevalence of tiredness and pain in young school-children.	Backache; headache; stomach ache; tiredness.
Rhee et al, 2005[3]	Prevalence of recurrent physical symptoms in US adolescents.	Chest pain; cold sweat; dizziness; fatigue; feeling hot; frequent sore throat/cough; headache; stomach ache; musculoskeletal pain; painful/frequent urination.

Table 3: Studies identified by the generic health search only.

Author, date	Title	Health outcome
Bigal et al, 2007[4]	Migraine in adolescents: Association with socioeconomic status and family history.	Migraine.
Bisegger et al, 2005[5]	Health-related quality of life: gender differences in childhood and adolescence.	Health related quality of life.
Cavallo et al, 2006[6]	Girls growing through adolescence have a higher risk of poor health.	Backache; difficulties in sleeping; feeling dizzy; feeling low; feeling nervous; headache; irritability and bad temper; self-rated health;

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		stomach ache.
Gadin & Hammarstrom, 2000[7]	School-related health – A cross-sectional study among boys and girls.	Abdominal pain; backache; headache; nausea; self-worth; stress; tiredness.
Grimmer et al, 2006[8]	Longitudinal investigation of low back pain in Australian adolescents: a five-year study.	Low back pain.
Haugland et al, 2001[9]	Subjective health complaints in adolescence. A cross-national comparison of prevalence and dimensionality.	Abdominal pain; backache; dizziness; feeling low; feeling nervous; headache; irritability or bad mood; sleeping difficulties.
Holmberg, & Hellberg, 2007[10]	Age-related gender differences of relevance for health in Swedish adolescents.	Abdominal pain; feeling depressed; feeling healthy; headache; suicidal thoughts.
Jorngarden et al, 2006[11]	Measuring health-related quality of life in adolescents and young adults: Swedish normative data for the SF-36 and the HADS, and the influence of age, gender and method of administration.	Anxiety; depression; health related quality of life.
Kujala et al, 1999[12]	Leisure physical activity and various pain symptoms among adolescents.	Abdominal pain; headache; lower back pain; lower limb pain; neck and shoulder pain; upper back pain; upper limb pain.
Laaksonen et al, 2010[13]	The change in child self-assessed and parent-proxy assessed health related quality of life in early adolescence (age 10-12).	Health related quality of life.
Lundqvist et al, 2006[14]	Self-reported headache in	Headache.

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	schoolchildren: parents underestimate their children's headaches.	
Meland et al, 2007[15]	Body image and perceived health in adolescence.	Body image; perceived health.
Ostberg et al, 2006[16]	Living conditions and psychosomatic complaints in Swedish schoolchildren.	Difficulties falling asleep; headache; stomach ache.
Palacio-Vieira et al, 2008[17]	Changes in health-related quality of life in a population-based sample of children and adolescents after 3 years follow-up.	Health related quality of life.
Ravens-Sieberer et al, 2008[18]	Health-related quality of life in children and adolescents in Germany: results of the BELLA study.	Health related quality of life.
Skordis et al, 2002[19]	The incidence of type 1 diabetes mellitus in Greek-Cypriot children and adolescents in 1990-2000.	Type 1 diabetes mellitus.
Sleskova et al, 2005[20]	Health status among young people in Slovakia: comparisons on the basis of age, gender and education.	Health complaints (backache; bone/muscle ache; breathlessness; chest/heart pain; dizziness; full/bloated stomach; headache; listlessness; pins and needles; tiredness;upset stomach); mental health; long-standing illness; long-term wellbeing; self-rated health; vitality.
Sundblad et al, 2007[21]	Prevalence and co-occurrence of self-rated pain and perceived health	Abdominal pain; headache; loneliness; musculoskeletal

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	in school-children: age and gender differences.	pain; problems sleeping; sadness; tiredness.
Sweeting & West, 2003[22]	Sex differences in health at ages 11, 13 and 15.	Depression; general health; recent symptoms (aching back, legs or arms; asthma or wheeze; cold or flu; difficulty getting to sleep; dizzy or faint; headache; irritable or bad tempered; nervous, worried or anxious; sad, unhappy or low; spots, rashes or other skin problems; stomach ache or feeling sick).
Torsheim et al, 2006[23]	Cross-national variation of gender differences in adolescent subjective health in Europe and North America.	Health complaints (backache; depressed mood; dizziness; headache; irritable; nervousness; stomach ache; sleeping difficulties).
Wedderkopp et al, 2001[24]	Back pain reporting pattern in a Danish population-based sample of children and adolescents.	Back pain; neck pain.

Table 4: Studies identified by the specific health search only

Author, date	Title	Health outcome
Beilmann et al, 1999[25]	Incidence of childhood epilepsy in Estonia.	Epilepsy.
Carle et al, 2004[26]	Diabetes incidence in 0 to 14-year age group in Italy. A 10-year prospective study.	Type 1 diabetes mellitus.
Casu et al, 2004[27]	Type 1 diabetes among Sardinian	Type 1 diabetes mellitus.

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	Children is increasing. The Sardinian diabetes register for children aged 0-14 years (19889-1999).	
Christensen et al, 2007[28]	Incidence and prevalence of epilepsy in Denmark.	Epilepsy.
Cinek et al, 2000[29]	Type 1 diabetes mellitus in Czech children diagnosed in 1990-1997: a significant increase in incidence and male predominance in the age group 0-4 years.	Type 1 diabetes mellitus.
Cotellessa et al, 2003[30]	High incidence of type 1 diabetes in Liguria Italy, from 1989 to 1998.	Type 1 diabetes mellitus.
Freitag et al, 2001[31]	Incidence of epilepsies and epileptic syndromes in children and adolescents: a population-based prospective study in Germany.	Epilepsy.
Heinrich et al, 2009[32]	Self-report of headache in children and adolescents in Germany: possibilities and confines of questionnaire data for headache classification.	Headache; migraine.
Karvonen et al, 1999[33]	The onset age of type 1 diabetes in Finnish children has become younger.	Type 1 diabetes mellitus.
Larsson & Sund, 2005[34]	One-year incidence, course and outcome predictors of frequent headaches among early adolescents.	Headache.
Laurell et al, 2004[35]	Prevalence of headache in Swedish schoolchildren, with a focus on tension-type headache.	Headache; migraine.

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Leonardsson-Hellgren et al, 2001[36]	Headache and associations with lifestyle among pupils in senior level elementary school.	Headache.
Mavromichalis et al, 1999[37]	Prevalence of migraine in schoolchildren and some clinical comparisons between migraine with and without aura.	Migraine.
Michalkova et al, 1995[38]	Incidence and prevalence of childhood diabetes in Slovakia (1985-1992).	Type 1 diabetes mellitus.
Mortimer et al, 1992[39]	Epidemiology of headache and childhood migraine in an urban general practice using ad hoc, Vahlquist and IHS criteria.	Headache; migraine.
Mortimer et al, 1993[40]	Clinical epidemiology of childhood abdominal migraine in an urban general practice.	Abdominal migraine; headache; recurrent abdominal pain.
Santinello et al, 2008[41]	Primary headache in Italian early adolescents: the role of perceived teacher unfairness.	Headache.

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References

1. Gordon KE, Dooley JM, Wood EP. Prevalence of reported migraine headaches in Canadian adolescents. *Canadian Journal of Neurological Sciences* 2004;31(3):324-7.
2. Petersen S, BergstrÃ¶m E, Brulin C. High prevalence of tiredness and pain in young schoolchildren. *Scandinavian Journal of Public Health* 2003;31(5):367-74.
3. Rhee H, Miles MS, Halpern CT, Holditch-Davis D. Prevalence of recurrent physical symptoms in U.S. adolescents. *Pediatric Nursing* 2005;31(4):314.
4. Bigal ME, Lipton RB, Winner P, Reed ML, Diamond S, Stewart WF, et al. Migraine in adolescents: association with socioeconomic status and family history. *Neurology* 2007;69(1):16-25.
5. Bisegger C, Cloetta B, von Rueden U, Abel T, Ravens-Sieberer U, European Kidscreen G. Health-related quality of life: gender differences in childhood and adolescence. *Sozial- und Praventivmedizin* 2005;50(5):281-91.
6. Cavallo F, Zambon A, Borraccino A, Raven-Sieberer U, Torsheim T, Lemma P, et al. Girls growing through adolescence have a higher risk of poor health. *Quality of Life Research* 2006;15(10):1577-85.
7. Gadin KG, Hammarstrom A. School-related health - A cross-sectional study among young boys and girls. *International Journal of Health Services* 2000;30(4):797-820.
8. Grimmer K, Nyland L, Milanese S. Longitudinal investigation of low back pain in Australian adolescents: a five-year study. *Physiotherapy Research International* 2006;11(3):161-72.
9. Haugland S, Wold B, Stevenson J, Aaroe LE, Woynarowska B. Subjective health complaints in adolescence: A cross-national comparison of prevalence and dimensionality. *European Journal of Public Health* 2001;11(1):4-10.
10. Holmberg LI, Hellberg D. Age-related gender differences of relevance for health in Swedish adolescents. *International Journal of Adolescent Medicine & Health* 2007;19(4):447-57.
11. Jorngarden A, Wettergen L, von Essen L, Jorngarden A, Wettergen L, von Essen L. Measuring health-related quality of life in adolescents and young adults: Swedish normative data for the SF-36 and the HADS, and the influence of age, gender, and method of administration. *Health & Quality of Life Outcomes* 2006;4:91.
12. Kujala UM, Taimela S, Viljanen T. Leisure physical activity and various pain symptoms among adolescents. *British Journal of Sports Medicine* 1999;33(5):325-8.
13. Laaksonen CB, Aromaa ME, Asanti RE, Heinonen OJ, Koivusilta LK, Koski PJ, et al. The change in child self-assessed and parent proxy-assessed Health Related Quality of Life (HRQL) in early adolescence (age 10-12). *Scandinavian Journal of Public Health* 2010;38(1):9-16.
14. Lundqvist C, Clench-Aas J, Hofoss D, Bartonova A, Lundqvist C, Clench-Aas J, et al. Self-reported headache in schoolchildren: parents underestimate their children's headaches. *Acta Paediatrica* 2006;95(8):940-6.
15. Meland E, Haugland S, Breidablik HJ, Meland E, Haugland S, Breidablik H-J. Body image and perceived health in adolescence. *Health Education Research* 2007;22(3):342-50.
16. Ostberg V, Alfven G, Hjern A, Ostberg V, Alfven G, Hjern A. Living conditions and psychosomatic complaints in Swedish schoolchildren. *Acta Paediatrica* 2006;95(8):929-34.

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17. Palacio-Vieira JA, Villalonga-Olives E, Valderas JM, Espallargues M, Herdman M, Berra S, et al. Changes in health-related quality of life (HRQoL) in a population-based sample of children and adolescents after 3 years of follow-up. *Quality of Life Research* 2008;17(10):1207-15.
18. Ravens-Sieberer U, Erhart M, Wille N, Bullinger M, group Bs, Ravens-Sieberer U, et al. Health-related quality of life in children and adolescents in Germany: results of the BELLA study. *European Child & Adolescent Psychiatry* 2008;17 Suppl 1:148-56.
19. Skordis N, Theodorou S, Apsiotou T, Stavrou S, Herakleous E, Savva SC. The incidence of type 1 diabetes mellitus in Greek-Cypriot children and adolescents in 1990-2000. *Pediatric Diabetes* 2002;3(4):200-04.
20. Sleskova M, Salonna F, Madarasova Geckova A, van Dijk JP, Groothoff JW. Health status among young people in Slovakia: comparisons on the basis of age, gender and education. *Social Science & Medicine* 2005;61(12):2521-7.
21. Sundblad GMB, Saartok T, Engstrom LMT. Prevalence and co-occurrence of self-rated pain and perceived health in school-children: Age and gender differences. *European Journal of Pain* 2007;11(2):171-80.
22. Sweeting H, West P. Sex differences in health at ages 11, 13 and 15. *Social Science & Medicine* 2003;56(1):31-9.
23. Torsheim T, Ravens-Sieberer U, Hetland J, Valimaa R, Danielson M, Overpeck M, et al. Cross-national variation of gender differences in adolescent subjective health in Europe and North America. *Social Science & Medicine* 2006;62(4):815-27.
24. Wedderkopp N, Leboeuf-Yde C, Andersen LB, Froberg K, Hansen HS. Back pain reporting pattern in a Danish population-based sample of children and adolescents. *Spine* 2001;26(17):1879-83.
25. Beilmann A, Napa A, Hamarik M, Soot A, Talvik I, Talvik T. Incidence of childhood epilepsy in Estonia. *Brain & Development* 1999;21(3):166-74.
26. Carle F, Gesuita R, Bruno G, Coppa GV, Falorni A, Lorini R, et al. Diabetes incidence in 0- to 14-year age-group in Italy: a 10-year prospective study. *Diabetes Care* 2004;27(12):2790-96.
27. Casu A, Pascutto C, Bernardinelli L, Songini M. Type 1 diabetes among Sardinian children is increasing: The Sardinian diabetes register for children aged 0-14 years (1989-1999). *Diabetes Care* 2004;27(7):1623-29.
28. Christensen J, Vestergaard M, Pedersen MG, Pedersen CB, Olsen J, Sidenius P, et al. Incidence and prevalence of epilepsy in Denmark. *Epilepsy Research* 2007;76(1):60-5.
29. Cinek O, Lanska V, Kolouskova S, Sumnik Z, Snajderova M, Ronningen KS, et al. Type I diabetes mellitus in Czech children diagnosed in 1990-1997: A significant increase in incidence and male predominance in the age group 0-4 years. *Diabetic Medicine* 2000;17(1):64-69.
30. Cotellessa M, Barbieri P, Mazzella M, Bonassi S, Minicucci L, Lorini R. High incidence of childhood type 1 diabetes in Liguria, Italy, from 1989 to 1998. *Diabetes Care* 2003;26(6):1786-89.
31. Freitag CM, May TW, Pfafflin M, Konig S, Rating D. Incidence of epilepsies and epileptic syndromes in children and adolescents: a population-based prospective study in Germany. *Epilepsia* 2001;42(8):979-85.

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32. Heinrich M, Morris L, Kroner-Herwig B. Self-report of headache in children and adolescents in Germany: Possibilities and confines of questionnaire data for headache classification. *Cephalalgia* 2009;29(8):864-72.
33. Karvonen M, Pitkaniemi J, Tuomilehto J. The onset age of type 1 diabetes in Finnish children has become younger. *Diabetes Care* 1999;22(7):1066-70.
34. Larsson B, Sund AM. One-year incidence, course, and outcome predictors of frequent headaches among early adolescents. *Headache: The Journal of Head & Face Pain* 2005;45(6):684-91.
35. Laurell K, Larsson B, Eeg-Olofsson O. Prevalence of headache in Swedish schoolchildren, with a focus on tension-type headache. *Cephalalgia* 2004;24(5):380-88.
36. Leonardsson-Hellgren M, Gustavsson U, Lindblad U. Headache and associations with lifestyle among pupils in senior level elementary school. *Scandinavian Journal of Primary Health Care* 2001;19(2):107-11.
37. Mavromichalis I, Anagnostopoulos D, Metaxas N, Papanastassiou E. Prevalence of migraine in schoolchildren and some clinical comparisons between migraine with and without aura. *Headache* 1999;39(10):728-36.
38. Michalkova DM, Cernay J, Dankova A, Rusnak M, Fandakova K. Incidence and prevalence of childhood diabetes in Slovakia (1985-1992). *Diabetes Care* 1995;18(3):315-20.
39. Mortimer MJ, Kay J, Jaron A. Epidemiology of headache and childhood migraine in an urban general practice using Ad Hoc, Vahlquist and IHS criteria. *Developmental Medicine and Child Neurology* 1992;34(12):1095-101.
40. Mortimer MJ, Kay J, Jaron A. Clinical epidemiology of childhood abdominal migraine in an urban general practice. *Developmental Medicine and Child Neurology* 1993;35(3):243-48.
41. Santinello M, Vieno A, De Vogli R, Santinello M, Vieno A, De Vogli R. Primary headache in Italian early adolescents: the role of perceived teacher unfairness. *Headache* 2009;49(3):366-74.

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STROBE Statement—checklist of items that should be included in reports of observational studies, followed by a table showing how the current study conforms to the STROBE statement.

	Item No	Recommendation
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found
Introduction		
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported
Objectives	3	State specific objectives, including any prespecified hypotheses
Methods		
Study design	4	Present key elements of study design early in the paper
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection
Participants	6	(*a*) *Cohort study*—Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up *Case-control study*—Give the eligibility criteria, and the sources and methods of case ascertainment and control selection. Give the rationale for the choice of cases and controls *Cross-sectional study*—Give the eligibility criteria, and the sources and methods of selection of participants
		(*b*) *Cohort study*—For matched studies, give matching criteria and number of exposed and unexposed *Case-control study*—For matched studies, give matching criteria and the number of controls per case
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group
Bias	9	Describe any efforts to address potential sources of bias
Study size	10	Explain how the study size was arrived at
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding
		(*b*) Describe any methods used to examine subgroups and interactions
		(*c*) Explain how missing data were addressed
		(*d*) *Cohort study*—If applicable, explain how loss to follow-up was addressed *Case-control study*—If applicable, explain how matching of cases and controls was addressed *Cross-sectional study*—If applicable, describe analytical methods taking account of sampling strategy
		(*e*) Describe any sensitivity analyses

Continued on next page

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Results		
Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed
		(b) Give reasons for non-participation at each stage
		(c) Consider use of a flow diagram
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders
		(b) Indicate number of participants with missing data for each variable of interest
		(c) *Cohort study*—Summarise follow-up time (eg, average and total amount)
Outcome data	15*	*Cohort study*—Report numbers of outcome events or summary measures over time
		Case-control study—Report numbers in each exposure category, or summary measures of exposure
		Cross-sectional study—Report numbers of outcome events or summary measures
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included
		(*b*) Report category boundaries when continuous variables were categorized
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses
Discussion		
Key results	18	Summarise key results with reference to study objectives
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence
Generalisability	21	Discuss the generalisability (external validity) of the study results
Other information		
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.

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STROBE Checklist: How to avoid missing half the evidence: comparing the use of generic and specific electronic search terms used to identify health outcomes for a systematic review.

STROBE Item No.	Authors comment
1 (a)	Done – see abstract, main article
1 (b)	Done – see abstract, main article
2	Done – main article (pages 4-6)
3	Done – main article (page 6)
4	Done – main article (page 8)
5	Done – a literature search is internet based rather than set in a specific location so we gave details of the databases searched and the review that the searches were conducted for. Main article (pages 8-9).
6 (a)	Done – main article (pages 8-10).
6 (b)	Not applicable – the study includes no matching of the kind described in STROBE.
7	Done – main article (pages 11).
8	Done – main article (pages 11).
9	Done – use of multiple reviewers during search and selection process (page 10).
10	Not applicable - the study did not require a power calculation as it includes no participants (in the conventional use of the terms). The text does state that the two searches were field tested during an actual systematic review – main article (pages 8-10) and protocol.
11	Done – main article (page 11).
12 a to e	Done – main article (pages 11).
13	Done – figure 1 and text in main article (pages 11-12)
14 a	Done – table 1, main article (page12)
14 b and c	Not applicable
15	Not applicable
16	Done in so far as applicable (the study does not involve estimates, statistical adjustment or missing data as described by STROBE). Main article (page 11-13).
17	Done – main article (page 11-13).
18	Done – main article (page 13)
19	Done – main article (page 14-15)
20	Done - main article (page 15-16)
21	Done - main article (page 15-16)
22	Done - main article (page 16-17)

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Comparing the effectiveness of using generic and specific electronic search terms to identify health outcomes for a systematic review: a prospective comparative study of literature search methods.

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COMPARING THE EFFECTIVENESS OF USING GENERIC AND SPECIFIC ELECTRONIC SEARCH TERMS TO IDENTIFY HEALTH OUTCOMES FOR A SYSTEMATIC REVIEW: A PROSPECTIVE COMPARATIVE STUDY OF LITERATURE SEARCH METHODS.

Matt Egan, *senior investigator scientist*[1*], Alice MacLean, *investigator scientist*[1], Helen Sweeting, *senior investigator scientist*[1], Kate Hunt, *programme leader*[1].

[1] MRC / CSO Social and Public Health Sciences Unit (SPHSU), 4 Lilybank Gardens, Glasgow G12 8RZ.

[*]Correspondence to m.egan@sphsu.mrc.ac.uk

Running title: 'How to avoid missing half the evidence'

Key words: Systematic review, literature search, evidence informed policy.

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Abstract

Objective

To compare the effectiveness of systematic review literature searches that use either generic or specific terms for health outcomes.

Design

Prospective comparative study of two electronic literature search strategies. The 'generic' search included general terms for health such as 'adolescent health', 'health status', 'morbidity', etc. The 'specific' search focused on terms for a range of specific illnesses, such as 'headache', 'epilepsy', 'diabetes mellitus', etc.

Data sources

We searched Medline, Embase, the Cumulative Index to Nursing and Allied Health Literature (CINAHL), PsycINFO, and the Education Resources Information Center (ERIC) for studies published in English between 1992 and April 2010.

Main outcome measures

Number and proportion of studies included in the systematic review that were identified from each search.

Results

The two searches tended to identify different studies. Out of 41 studies included in the final review, only 3 (7%) were identified by both search strategies; 21 (51%) were identified by the generic search only; and 17 (41%) were identified by the specific search only. Five of the 41 studies were also identified through handsearching methods. Studies identified by the two ELS differed in terms of reported health

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outcomes, whilst each ELS uniquely identified some of the review's higher quality studies.

Conclusions

Electronic literature searches (ELS) are a vital stage in conducting systematic reviews and therefore have an important role in the scientific community's attempts to inform and improve policy and practice with the best available evidence. Future systematic reviews that involve multiple health outcomes should include both generic and specific health terms in their literature search. Based on our findings, choosing only one or the other of these strategies could lead to systematic reviews that miss important evidence and consequently risk misinforming practitioners and other decision-makers. Future research should test the generalisability of these findings.

Abstract word count: 295

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Introduction

Electronic literature searches (ELS) are an essential stage in most systematic reviews.[1-2] As such, they have a crucial role in the scientific community's attempts to inform and improve policy and practice with the best available evidence.[3-4] Designing ELS can be challenging and it is widely recognized that specialist skills and knowledge, such as those provided by an information scientist, are important for best practice in this field.[1-3] A key challenge when conducting ELS is the need to screen out irrelevant evidence (specificity), whilst successfully identifying the relevant evidence (sensitivity). Search strategies that are insufficiently sensitive risk encouraging potentially harmful decisions based on the findings of reviews that have failed to identify important evidence. Search strategies that aim to comprehensively identify all the relevant evidence can present challenges in situations where reviewers have limited time or other resources (e.g. as a result of research funding requirements, or because findings are considered to be needed urgently), or when extending a search fails to identify relevant evidence and might therefore represent an ineffective allocation of scarce resources.[3-5] Some systematic reviews are based on comprehensive searches which aim to have high recall and retrieve references to all relevant papers, whereas others are based on more restricted searches which may limit the number of relevant papers identified.[5] Either way, there is a pressing need to learn more about how best to negotiate the competing demands of specificity and sensitivity.

Previous research exploring how to improve the effectiveness and efficiency of search strategies has tended to focus on issues such as how to optimize search outputs from

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'frontline' electronic databases (i.e. databases that are frequently searched for systematic reviews of medical interventions such as Medline and Embase), and how to identify randomised control trials (RCTs).[6-12] This research focus may in part reflect the influence of the Cochrane Collaboration, which has helped to stimulate considerable interest in systematic reviews of clinical trials.[1]

However, not all systematic reviews (nor indeed all Cochrane Reviews[13]) focus on RCTs of clinical interventions. Interest in broader, non-clinical systematic reviews has steadily increased within the social and public health sciences and other disciplines.[3 5] As some of these non-clinical reviews tackle relatively under-researched topics, they often combine a scoping and hypothesis testing function by asking relatively broad research questions that, for example, cover a range of outcomes (e.g. *what are the health impacts of intervention x?; what health outcomes are associated with risk-factor y?*).[14-27] Evidence-informed guidance on how to conduct searches for this broader range of systematic reviews is therefore an emerging priority.

There are few examples of research that can help guide information scientists and reviewers to develop efficient but effective search strategies for these broader / non-clinical systematic reviews. The research that is available illustrates how searches for such reviews can become lengthy and complex.[28] For example Greenhaulgh et al recommended the development of iterative search strategies to search for complex evidence (e.g. multiple study designs). Ogilvie et al suggested that cross-disciplinary reviews may necessitate searching databases across a range of disciplines rather than focusing on frontline health databases.[4]

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From our own experiences of conducting systematic reviews of non-clinical, public health research, the authors of this paper can identify additional challenges that have led to large and complex ELS. For example, search terms that involve commonly used words are likely to identify large numbers of irrelevant papers and non-clinical public health reviews often rely on commonly used terms to describe everyday settings, activities and outcomes (e.g. 'walking', 'obesity', 'stress', 'workplace health', 'health promotion', and 'general health'). In comparison, an ELS for a clinical review will often involve very specific medical terminology that can help to focus the search on papers relevant to a particular field.[3]

Furthermore, the identification of studies for clinical reviews typically requires three lists of search terms: (i) terms that define the population who will receive the intervention; (ii) terms to describe the intervention; and (iii) terms to identify a particular study design (typically a filter for RCTs).[1] Systematic reviews that focus on a more general population sample, have no intervention, and/or are not limited to a single study design, lack one or more of these three search components, and so result in a less specific ELS.

All these challenges increase the chances of a search becoming lengthy and cost-ineffective. In such circumstances, reviewers may look for alternative means of increasing search specificity but there has been relatively little guidance on how this can be achieved without compromising sensitivity.

Including search terms that relate to health outcomes is one commonly used technique for increasing precision in broader reviews.[14-24] However, if a review question is

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broad enough to include multiple health outcomes it is not obvious how an ELS that includes health outcomes can best accommodate this breadth of scope. Some reviews have used generic health terms (e.g. 'health', 'illness', 'morbidity') to search for evidence that includes a range of health outcomes.[14-17] In other cases, reviewers have used more specific search terms to identify a number of diseases or symptoms considered to be of particular relevance to the review question.[18-21] Both approaches may be hypothesized to have risks. Generic search terms may either be too inclusive (virtually every study on Medline is about 'health') or may miss studies that only use more specialist vocabulary to describe a particular illness. Specific search terms are problematic if the reviewers want to avoid pre-specifying which health outcomes are relevant to the review (e.g. scoping reviews). Some reviews combine both generic and specific approaches[22-24], but the extent to which this either adds value to the search or merely adds to the workload is not known.

We know of no study that has compared the relative merits of ELS strategies that focus on either generic terms for health, or specific terms for particular health issues or illnesses. Nor do we know of any evidence to help reviewers determine whether these two approaches are likely to identify a similar or a different set of publications. When the authors of this paper recently conducted a systematic review that included multiple health outcomes, we felt that guidance on this issue would have been helpful. As there was an absence of evidence upon which to base such guidance, we ran two separate literature searches for our review: one that included generic health terms and one that used more specific health terms. Our aim was to see which approach was most effective in identifying studies that were included in the final review.

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Hence, we examined whether the included studies tended to be identified from the generic search only, the specific search only, or both searches. We also explored efficiency by comparing the size of the searches (i.e. the number of references initially identified from the ELS – sometimes referred to as the number of 'hits') for each approach. Finally, we explored the extent to which the 'generic search' and the 'specific search' identified studies with different or similar types of health outcome.

Our review was conducted within a limited time frame (originally planned as nine months and then extended to 18 months), and we believe the implications of this study are of particular relevance to reviews of broader public health topics and reviews with time or other resource limitations.

Methods

This paper focuses on one specific, but crucial, stage of a systematic review: the literature search. We developed two contrasting strategies for searching electronic databases and compared their effectiveness in identifying studies for a specific systematic review. The systematic review itself is summarised in Panel 1, and described more fully in the publically available Protocol document (available as a supplemental document online), and the full report of the review which will be published separately to this methodological paper.

Panel 1. Summary of the systematic review used as the basis of this methodological study.

Title: How robust is the evidence of an emerging or increasing female excess in physical morbidity rates between childhood and adolescence? Results of a systematic

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literature review.

Hypothesis: That the incidence of physical morbidity amongst children tends to be higher amongst males in pre-adolescent childhood, but this male excess is replaced by an emergence of higher rates in females during the transition to adolescence.

Inclusion / Exclusion criteria: These criteria are summarised using the PICOS statement below. For full details of the inclusion and exclusion criteria, see the protocol: supplemental document).

Included studies must have the following characteristics

Population: males and females between the ages of 4 and 17;

Intervention: none;

Comparator: sex and age (at least two age-groups);

Outcome: gender patterning, by age, in measures of physical morbidity;

Study design: longitudinal, cross-sectional and repeat cross-sectional studies (including analysis of study-specific data or routinely collected data).

Methods: The systematic review included methodological components suggested by the PRISMA guidelines (e.g. protocol, literature search, study selection, flow chart, data extraction, critical appraisal and synthesis), and was designed to meet the standards of that guidance. More details are provided in the protocol.

Data sources and search strategy

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We searched five electronic databases (Medline, Embase, the Cumulative Index to Nursing and Allied Health Literature (CINAHL), PsycINFO, and the Education Resources Information Center (ERIC)) for studies published in English between 1992 and the date of search (April 2010). As it was our intention is to update a previous review conducted around twenty years previously,[29] we searched for studies published from 1992 to the present. Supplemental document 1 describes the review methods and search strategy in more detail. Following test-searches using pre-identified papers, an information scientist advised on database selection and search terms. As the review's timeframe was limited, the information scientist advised on a search strategy that limited the number of records retrieved by the searches so that they could be processed within the time frame. Prior to the electronic search we manually searched private collections (three of the reviewers have worked in the field of gender and adolescent health for several years or, in two cases, approximately two decades); conducted a relatively unstructured internet search and also identified papers that had cited the earlier review.[29] At the end of our study selection process we manually checked the bibliographies of included studies.

We searched each database twice: once using 'generic' health subject headings and keywords and once using 'specific' subject headings and keywords relating to the health conditions we had selected for review. In this paper, we refer to these searches as the 'generic search' and the 'specific search'. The generic search included the terms: health status, attitude to health, health attitudes, health surveys, child health, adolescent health, health status indicators, symptoms, morbidity, health complaints, general health questionnaire, well being, self report, and wellness. The specific search included the terms: asthma, epilepsy, diabetes mellitus, primary headache, and migraine. The specific search terms related to health conditions that we judged to be

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relevant to the research question and for which we were likely to find evidence. We based this judgement on an initial scoping of the literature and an earlier review of this topic[29]. The precise search strategy differed between databases if different search facilities and search engines made it necessary to adapt our approach. Specific details of our searches are presented in the review protocol (see supplemental document 1).

Study Selection

One reviewer (AM) screened all the publications identified by both literature searches to exclude obviously irrelevant titles. The remaining (i.e. not excluded) publications were retrieved and, on reading, AM screened out those that were clearly not eligible for inclusion in the review (see Figure 1, 'First Sift'). Studies of uncertain eligibility were checked by two other reviewers (KH and HS) so that a decision to exclude or retrieve the full paper could be reached (see Figure 1, 'Second Sift'). Some retrieved papers were excluded at the initial reading ('Third Sift'), whilst others were excluded at the data extraction and appraisal stage (based on agreement from all the reviewers). At this final stage we also excluded studies that only explored asthma-related outcomes after finding a review that already applied our research question to this health outcome.

Outcomes

Our main outcomes measures for this analysis were the number and proportion of studies included in the systematic review that were identified from each ELS. We also collected data on (i) the number of studies identified by each ELS at all stages of the

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reviews' search and selection process; (ii) the types of health outcomes identified by each ELS; and (iii) the number of studies identified by handsearches.

Comparing the two searches

We produced a series of Venn diagrams for each stage of the review process, showing the number of studies identified only by the specific literature search, the number identified only by the general literature search, and the number identified by both searches (see Figure 1). The purpose was to see if the two searches identified similar or different sets of documents. Studies that were included in the final review were then tabulated in more detail to help us assess whether there was any systematic variation in the types of health outcome identified by the different searches. Comparisons involved the calculation of frequencies and percentages.

Results

Figure 1 shows for each stage of the review the number of studies identified exclusively by either the specific or the generic search, and (in each intersect) the number of studies identified by both searches.

The diagram makes two points apparent. Firstly, there was relatively little duplication between the two searches. For example, out of the 11509 total hits identified from both literature searches, only 413 (3.6%) were duplicates between the two searches. Throughout each stage of the study selection process, duplication between the two searches remained low, so that only three (7.3%) of the 41 studies selected for final

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inclusion in the review were identified by both search strategies (further details of the 41 included studies are available in a supplemental document).

Secondly, we note that the specific search led to less than half the number of initial hits, compared to the generic search (3299 vs. 8210, respectively), but both searches identified a similar number of studies included in the final review (17 vs. 21, and 3 duplicates).

Four final inclusion studies were identified from our initial handsearch but the generic ELS also identified each of these four studies. Further bibliographic checking revealed that one of the studies identified using the generic ELS could also have been found by checking the bibliographies of included studies identified from the specific search. One study identified from the specific ELS was also identified from a bibliography check of the generic ELS studies and the initial handsearch. This means that the generic ELS in combination with the handsearch and bibliography check would have identified 24 of the 41 included studies. The specific ELS in combination with the handsearch and bibliography check would have identified 25 of the 41 included studies.

We then examined the 41 studies included in the final review, categorising them by the health outcomes each one investigated (see Table 1). The findings suggest some systematic differences in the health outcomes of studies identified using each of the two search strategies. The specific search tended to be the more successful at identifying studies that focused on a single type of health outcome (i.e. those that related to the search terms). The opposite was found for the generic search strategy,

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which tended to be more successful at identifying studies with multiple health outcomes.

Table 1: Studies included in the systematic review (n = 41) by summary health outcomes and by the search strategy used to identify each study.

Outcomes	Generic Search	Specific Search	Both Searches
Abdominal Pain		1	
Back pain	2		
Diabetes	1	6	
Epilepsy		3	
Headache	2	7	1
General physical health / wellbeing	5		
Multiple physical health outcomes*	11		2
Total (for each search)	21	17	3

* A range of health outcomes were included in these studies: usually involving measures of general health and bodily pain. See tables in supplemental document 2.

Most notably, we found that the specific ELS alone (i.e. not the generic ELS or handsearch) identified all three included studies of epilepsy and all but one of the seven studies on diabetes. Therefore, failure to run the specific search would have meant that our review would have missed most of the evidence relating to these two outcomes. Within the context of our review's findings, this omission would have been

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important because whilst the evidence for the other health outcomes presented in Table 1 tended to support our review's main hypothesis, findings for diabetes and epilepsy uniquely suggested a counter-hypothesis. Failing to identify evidence to support the counter-hypothesis would have directly affected our review's conclusions.

The tables in supplemental document 2 describe the studies identified by the different ELS by summarizing information on health outcome, journal, study design, appraisal score and country. Three longitudinal studies and six studies classed as higher scoring following the study appraisal were amongst those identified by the generic ELS (although three of these were also identified using the handsearch). Five higher scoring studies (but no longitudinal studies) were amongst those only identified by the specific ELS. Both searches identified evidence from a similar (but not identical) range of European countries but only the generic search identified any North American studies. All the studies identified were published in medical/health journals.

Discussion

We have compared two strategies for conducting an electronic literature search for a systematic review. One strategy used generic health terms, whilst the other used more specific health terms. The purpose was to explore whether literature searches with relatively broad inclusion criteria (in terms of health outcomes) are better served by generic or specific health terms, or whether both are needed.

We found that both specific and generic health terms were necessary. They each uniquely identified some of the review's more robust studies. They also identified different types of health outcome. Failure to identify some of those outcomes would

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have directly affected our review's conclusions. Had we only used generic health terms in our search we would have missed around half the studies that we finally included in the review. Likewise, focusing exclusively on specific health terms in the literature search would have failed to identify around half the included papers. A small proportion of these studies would have been identified by our handsearch and bibliography check but failing to conduct either of the ELS approaches would still have led to a serious 'loss' of data (or, more correctly, a failure to find data) that would have compromised the credibility and accuracy of our review's findings.

We found that the specific search tended to miss studies with general or multiple health outcomes, whilst the generic search tended to miss studies with single, specific health outcomes. This may appear intuitive, but we contend that the finding is actually surprising. It suggests, for example, that studies that look specifically at young people's diabetes, epilepsy and headache tend not to be identifiable by search terms such as "health status", "health surveys", "child health", "adolescent health", "health status indicators", "symptoms", "morbidity", "health complaints", etc. It also suggests that some studies that, for example, included headache as one of a number of different health outcomes may be identified by a search strategy that includes generic health terms, but could be missed by an ELS that specifically focuses on the term 'headache.'

This finding is at odds with what some authors of this paper initially expected. Prior to our exploring this issue, the authors assumed that the generic health search would identify the vast majority of included studies whilst the specific search would mainly identify a subset of those studies. If other systematic reviewers also make this

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assumption, then their reviews are at risk of being based on poor quality (highly insensitive) searches.

Strengths and limitations

We have conducted a prospective comparative study of two electronic literature search strategies that have been field tested whilst we conducted a systematic review. This kind of study is uncommon and hence novel, whilst the prospective and comparative design is a key strength.

The review that we based the study on does not focus on the effectiveness of an intervention, which means that specificity cannot be easily increased by including simple study design search terms, and the outcomes are also very complex, which probably increases the difficulty of sensitive and specific searching. These may be regarded as unusual features affecting the generalisablity of our findings but we have argued in our introduction that 'unusual' (i.e. not clinical intervention) reviews are becoming more common and hence are an emerging priority in terms of review methods. The same may be said about time-limited reviews. Ours took eighteen months to complete – not an unusual timeframe in our experience but we are aware that some systematic reviews (e.g. many Cochrane and Campbell reviews) take longer and involve more comprehensive searches. The information scientist who advised on both search strategies, and the researcher who led the comparative study have a good understanding of the challenges involved with reviews of this kind.

The main limitation of this study is that it is based on a single review. There is some existing evidence that the effectiveness of different search strategies may vary

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depending on the subject of the review.[5] It may also be hypothesised that conducting a more extensive ELS and handsearch could have led to a greater number of, and possibly more overlap between, studies identified by each component of our search strategy. Ways to achieve a more extensive search could have included using more electronic databases and other relevant data sources; identifying a wider number of synonyms for both the health outcomes and other concepts included in the review; using both subject headings and words in the title and abstract to search for every concept in the search strategy; and minimising reliance on the accuracy of database indexers. Therefore, it is worth testing our findings in the context of other reviews and different types of literature search, including more sensitive searches. Missing out health outcomes altogether is an alternative means of increasing search sensitivity but we note that our initial search identified well in excess of 10,000 hits. Given the broad review question, attempts to vastly expand the search risked increasing the number of hits to unmanageable levels.

Implications and conclusions

Literature searching has a vital role to play in evidence-informed policy and practice, and it is plausible to theorise a direct pathway by which a poor search may lead to harmful decisions. Conducting research that may assist information scientists and reviewers to improve their search strategies should therefore be a priority. Such research can be nested within the processes of conducting systematic reviews: from our own experience this requires minimal additional resources to the cost of the overall review and can therefore be considered an inexpensive way of conducting useful research in an important field. We therefore hope that other reviewers will

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make use of similar opportunities to explore how best to optimise electronic searching.

In light of our findings, we recommend that future systematic reviews of topics that involve multiple health outcomes include both generic and specific health terms in their literature search (if a health outcome search is considered necessary), along with handsearching. Choosing only one of these search components could, based on our findings, increase the risk of reviewers missing robust evidence and making misleading conclusions.

Authors' interests

(1) All authors have completed the Unified Competing Interest form at www.icmje.org/coi_disclosure.pdf (available on request from the corresponding author) and declare that ME, AM, HS, KH have support from the Medical Research Council and Chief Scientist Office for the submitted work; (2) ME, AM, HS, KH have no relationships with any company that might have an interest in the submitted work in the previous 3 years; (3) their spouses, partners, or children have no financial relationships that may be relevant to the submitted work; and (4) ME, AM, HS, KH have no non-financial interests that may be relevant to the submitted work.

Ethics

As this study focused on searching for literature that was already in the public domain, it involved no patients, consents, nor other issues that required formal approval from an ethics committee.

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Data sharing

The review protocol (including search strategies) and a list of studies included in the final review are available in the supplemental documents submitted with this article. Further data related to the searches are available from the corresponding author at matt@sphsu.mrc.ac.uk.

Details of contributors

ME helped plan and conduct the study, analyse the findings, led on writing the manuscript and is guarantor for the study. AM, HS and KH helped plan the study and conduct the study, analyse the findings and provide content and comments on the manuscript. All authors, external and internal, had full access to all of the data (including statistical reports and tables) in the study and can take responsibility for the integrity of the data and the accuracy of the data analysis.

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What is already known on this subject

Literature searching has a vital role to play in systematic reviews that inform policy and practice. Evidence to help reviewers conduct effective literature searches tends to be based on reviews of randomised controlled trials. There is relatively little evidence to help guide literature searches for other types of systematic review.

What this study adds

Whilst conducting a systematic review that included a range of health outcomes, we compared two electronic literature search strategies – one that used generic terms for (ill)health and another than used terms for specific illnesses. Our findings suggest that systematic review searches that use *only* generic *or* specific search terms (rather than a combination of the two) for health outcomes risk missing a large proportion of the relevant evidence, potentially leading to erroneous conclusions that may, in turn, misinform policy and practice.

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References

1. Higgins JPT, Green S. *Cochrane Handbook for Systematic Reviews of Interventions Version 5.0.2 [updated September 2009]*. : The Cochrane Collaboration, 2009.
2. NHS Centre for Reviews and Dissemination. *Systematic Reviews: CRD's Guidance for Undertaking Reviews in Healthcare* York: CRD University of York, 2009.
3. Petticrew M, Roberts H. *Systematic Reviews in the Social Sciences: A practical guide.* Oxford: Blackwell Publishing, 2006.
4. Ogilvie D, Hamilton V, Egan M, Petticrew M. Systematic reviews of health effects of social interventions: 1. Finding the evidence: how far should you go? *J Epidemiol Community Health* 2005;59:804-08.
5. Bambra C. Real world reviews: a beginner's guide to undertaking systematic reviews of public health policy interventions. *J Epidemiol Community Health* 2011;65:14-19.
6. Glanville JM, Lefebvre C, Miles JN, Camosso-Stefinovic J. How to identify randomized controlled trials in MEDLINE. *J Med Libr Assoc* 2006;130-136:130-36.
7. Golder S, McIntosh HM, Duffy S, Glanville J, Centre for Reviews and Dissemination and UK Cochrane Centre Search Filters Design Group. Developing efficient search strategies to identify reports of adverse effects in MEDLINE and EMBASE. *Health Info Libr J* 2006;23:3-12.
8. Montori VM, Wilczynski NL, Morgan D, Haynes RB. Optimal search strategies for retrieving systematic reviews from Medline: analytical survey. *BMJ* 2005;330:68.
9. Wong SS, Wilczynski NL, Haynes RB. Developing optimal search strategies for detecting clinically sound treatment studies in EMBASE. *J Med Libr Assoc* 2006;94:41-47.
10. Dickersin K, Scherer R, Lefebvre C. Identifying relevant studies for systematic reviews. *BMJ* 1994;309:1286-91.
11. Eisinga A, Siegfried N, Clarke M. The sensitivity and precision of search terms in Phases I, II and III of the Cochrane Highly Sensitive Search Strategy for identifying reports of randomized trials in MEDLINE in a specific area of health care - HIV/AIDS prevention and treatment interventions. *Health Info Libr J* 2007; 24:103-09.
12. Royle P MR. Literature searching for randomized controlled trials used in Cochrane reviews: rapid versus exhaustive searches. *Int J Technol Assess Health Care* 2003;19:591-603.
13. Joyce K, Pabayo R, Critchley JA, Bambra C. Flexible working conditions and their effects on employee health and wellbeing. *Cochrane Database Syst Rev*, 2010.
14. Porta D, Milani S, Lazzarino A, Perucci C, Forastiere F. Systematic review of epidemiological studies on health effects associated with management of solid waste. *Environ Health* 2009;8(60):doi:10.1186/476-069X-8-60.
15. Uscher-Pines L. Health effects of relocation following disaster: a systematic review of the literature. *Disasters* 2009;33(1):1-22.
16. D'Onise K, Lynch J, Sawyer M, McDermott R. Can preschool improve child health outcomes? A systematic review. *Soc Sci Med* 2010;70(9):1423-40.
17. Egan M, Petticrew M, Ogilvie D, Hamilton V. New Roads and Human Health: A Systematic Review. *Am J Public Health* 2003;93(9):1463–71.

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18. Dangour A, Aikenhead A, Hayter A, Allen E, Lock K, Uauy R. Comparison of putative health effects of organically and conventionally produced foodstuffs: a systematic review: Report for the Food Standards Agency. London: Nutrition and Public Health Intervention Research Unit London School of Hygiene & Tropical Medicine, 2009.
19. Vickers AJ. Can acupuncture have specific effects on health? A systematic review of acupuncture antiemesis trials. *J R Soc Med* 1996;89:303-11.
20. Daykin N, Orme J, Evans D, Salmon D, McEachran M, Brain S. The Impact of Participation in Performing Arts on Adolescent Health and Behaviour: A Systematic Review of the Literature. *J Health Psychol* 2008;13:251-64.
21. Rogers G, Elston J, Garside R, Roome C, Taylor R, Younger P, et al. The harmful health effects of recreational ecstasy: a systematic review of observational evidence. *Health Technol Assess* 2009;13(6):DOI: 10.3310/hta13060.
22. Passchier W, Knottnerus A, Albering H, Walda I. Public health impact of large airports. *Rev Environ Health. 2000 Jan-Jun;15(1-2):* 2000;15(1-2):83-96.
23. Egan M, Petticrew M, Ogilvie D, Hamilton V, Drever F. "Profits before people"? A systematic review of the health and safety impacts of privatising public utilities and industries in developed countries. *J Epidemiol Community Health* 2007;61:862-70.
24. McCartney G, Thomas S, Thomson H, Scott J, Hamilton V, Hanlon P, et al. The health and socioeconomic impacts of major multi-sport events: systematic review (1978-2008). *BMJ* 2010;340:c2369.
25. Paradies Y. A systematic review of empirical research on self-reported racism and health. *Int J Epidemiol* 2006;35(4):888-901.
26. Vartanian LR, Schwartz MB, Brownell KD. Effects of soft drink consumption on nutrition and health: a systematic review and meta-analysis. *Am J Public Health* 2007;97(4):(4):667-75.
27. Bambra C, Whitehead M, Sowden A, Akers J, Petticrew M. "A hard day's night?" The effects of Compressed Working Week interventions on the health and work-life balance of shift workers: a systematic review. *J Epidemiol Community Health* 2008;62(9):764-77.
28. Greenhalgh T, Peacock R. Effectiveness and efficiency of search methods in systematic reviews of complex evidence: audit of primary sources. *BMJ* 2005;331:1064-65.
29. Sweeting H. Reversals of fortune? Sex differences in health in childhood and adolescence. *Soc Sci Med* 1995;40(1):77-90.

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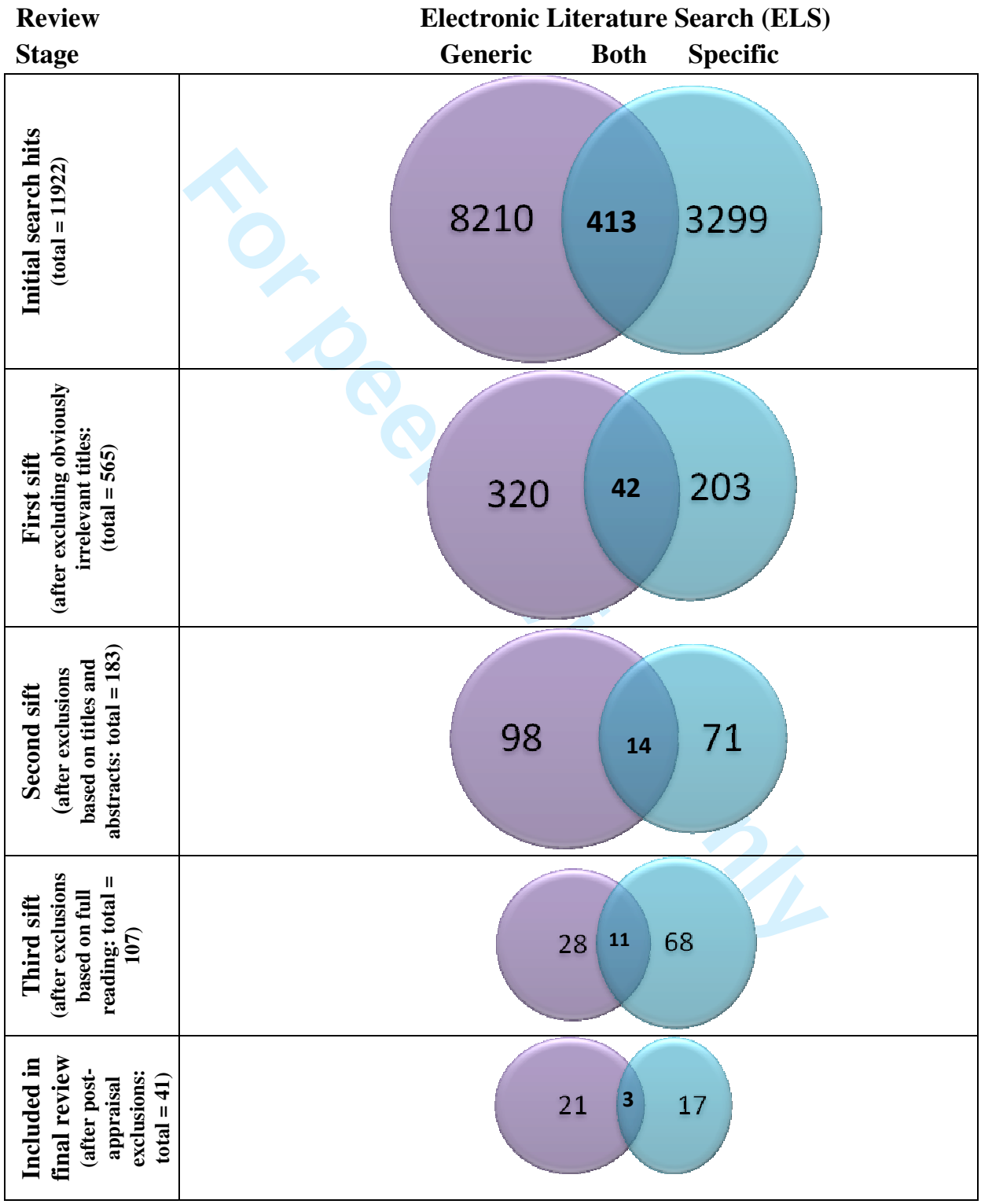
Figure 1. Review study selection flow chart: studies identified by the 'generic' search only (purple circle); 'specific' search only (light blue circle); and by both searches (dark blue intersect).



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Note: from the above figures the following can be calculated.

Generic ELS: sensitivity = 58.5%; precision = 0.3%; number needed to read = 112 (1.3%).

Specific ELS: sensitivity = 48.8%; precision = 0.5%; number needed to read = 85 (2.3%).

An initial manual search identified ten articles for full reading, of which 4 were included in the final review. These studies were also identified from the generic ELS, and are included as such in the figure above.

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Supplemental document 1

How robust is the evidence of an emerging or increasing female excess in morbidity rates between childhood and adolescence?

Review protocol

October 2011

Alice MacLean, Matt Egan, Helen Sweeting, Joy Adamson and Kate Hunt.

MRC Social & Public Health Sciences Unit
4 Lilybank Gardens
Glasgow
G12 8RZ

Tel: 0141 357 3949
e-mail: alice@sphsu.mrc.ac.uk

BACKGROUND

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Supplemental document 1

This protocol is an updated version of the original, which was written in March 2010 and set out our plans for conducting the review. Any significant changes made to the review process, between the writing of the first protocol and this updated version, are highlighted using footnotes.

In 1995, Social Science & Medicine published a narrative review of research findings on sex differences in health among children and adolescents (Sweeting 1995). By examining and summarising the findings from a broad range of research on the physical health, psychological well-being and health service utilisation of children and adolescents, with a focus on the 7 to 15 age-range, Sweeting's review provides evidence of a 'gender reversal' in the distribution of ill-health across the transition from childhood to adolescence. Gender and age differences in rates of asthma are referred to in the review as one example of this reversal in physical health. It documents that in children less than 10 years old, rates of asthma are highest among boys but by adolescence boys' and girls' rates converge and after this time higher rates of asthma are often found among girls. A similar picture is presented in relation to psychological well-being; overall rates of psychiatric disorders are more prevalent amongst boys until early adolescence, however the referral rates for girls with psychiatric disorders have been found to rise after 12 years of age and exceed those of boys by age 15-16. As well as demonstrating an overall emergence of excess morbidity in females over early-mid adolescence, Sweeting's review highlighted a need for longitudinal studies to chart sex differences in physical and psychological health, as well as illness behaviours and beliefs, across the transition from childhood to adolescence.

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Since the publication of Sweeting's (1995) narrative review, substantial research evidence has been found to suggest that higher rates of psychological morbidity found among males in childhood are replaced by an emergence of higher rates in females during the transition to adolescence (Petersen, Sarigiani et al. 1991; Cohen, Cohen et al. 1993; Schraedley, Gotlib et al. 1999; Ge, Conger et al. 2001; Marcotte, Fortin et al. 2002; Bennett, Ambrosini et al. 2005). This pattern has also been reported for asthma prevalence (Venn, Lewis et al. 1998; Nicolai, Pereszlenyiova-Bliznakova et al. 2003; Sears, Greene et al. 2003). Indeed, a number of reviews have synthesised and documented this evidence and have contributed to an established recognition of an emerging/increasing female excess in rates of psychological disorders (Nolen-Hoeksema and Girgus 1994; Hankin and Abramson 1999; Cyranowski, Frank et al. 2000; Shibley Hyde, Mezulis et al. 2008) and asthma[1] (Zannolli and Morgese 1997; Postma 2007; Almqvist, Worm et al. 2008).

However, there are no reviews, to our knowledge, which have been conducted with the aim of investigating the extent to which there is evidence of an emerging/increasing female excess in relation to other, or a range of, physical morbidity outcomes. This is surprising given that in recent decades several large-scale European and North American surveys of children and adolescents aged between 11 and 16 have reported comparable patterns of an overall emerging or increasing excess in girls' rates of reporting both psychological and physical symptoms (Eiser, Havermans et al. 1995; Eminson, Benjamin et al. 1996; Klepp, Aas et al. 1996; Haugland, Wold et al. 2001; Hetland, Torsheim et al. 2002; Sweeting and

[1] Originally we had planned to include asthma and psychological symptoms and conditions in this review. However, after identifying recent reviews which had explored the gender patterning of prevalence rates by age in relation to these health outcomes, we subsequently excluded studies which only presented data on asthma and psychological health outcomes.

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West 2003; Torsheim, Ravens-Sieberer et al. 2006). Indeed, the emergence of female excess morbidity during adolescence has been described as a central feature of adolescent health in 'a large proportion of the world's industrialised countries' (Torsheim, Ravens-Sieberer et al. 2006, p.823). Therefore, assessing the amount and quality of evidence suggesting that morbidity rates for a range of outcomes vary by gender according to age, may take us closer to explaining the emergence of higher reported morbidity in females.

REVIEW AIMS

This review aimed to investigate the extent to which research has found evidence of an emerging/increasing female excess in relation to physical morbidity rates across childhood and adolescence.

Our objectives, in terms of the PICOS statement, were as follows:

Population: males and females between the ages of 4 and 17

Intervention: none

Comparator: gender and age (at least two age-groups)

Outcome: gender patterning, by age, in measures of physical morbidity

Study design: longitudinal, cross-sectional and repeat cross-sectional studies (including analysis of study-specific data or routinely collected data).

METHODS

Searching

The following bibliographic databases were searched: Medline; Embase; CINAHL (Cumulative Index to Nursing & Allied Health Literature); PsycINFO; and ERIC

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(Education from U.S. Department of Education, & Institute of Education Sciences). Academic research was targeted and no grey literature was included in the review. Searches were limited to articles published in English between 1992 and the date of search (April 2010). As it was our intention to update Sweeting's 1995 narrative review (written in 1994), we predicted that searching for articles published in the three years leading up to its publication would enable us to retrieve any relevant studies which may have been in the publication process at the same time as, and therefore not included in, the 1995 review. The search strategy was developed in collaboration with an information scientist. Prior to the electronic search we manually searched private collections; a relatively unstructured internet search and we identified papers that had cited Sweeting' earlier review

The precise search strategy differed slightly between databases if different search facilities and search engines made it necessary to adapt our approach (see Appendix 1 and 2 for the full search strategies used in each database). Our searches included three groups of terms:

1) Terms to identify the target group

Adolescent; adolescence; adolesc*; child; child, preschool; children; early adolescents; late adolescents; preschool child; minor*; pupil*; school child*; teenage*; young children; young pers*.

2) *Terms to identify the comparator*

Gender; gender differences; human sex difference; sex; sex distribution; sex factors.

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3) Terms to identify health measures

We searched each database twice; once using *generic* health subject headings and keywords and once using *specific* subject headings and keywords relating to physical symptoms and conditions common in childhood[2].

a) Generic search terms: adolescent health; attitude to health; child health; general health questionnaire; health; health attitudes; health complaints; health status; health status indicators; health survey; morbidity; self-report; symptoms; well-being; wellness.

b) Specific search terms: diabetes mellitus; epilepsy; headache; headache disorders, primary; migraine; primary headache.

Data management

A 'search diary' was kept, which detailed the names of the databases searched, the search terms used and the search results (see Appendix 1 and 2). The results of each search were exported to an Endnote database, along with details of which database they were imported from and whether they were the results of the generic or specific search. Titles and abstracts were screened by one reviewer and inclusion/exclusion decisions recorded on the Endnote database. To check for consistency in screening, a random sample of abstracts was screened by two other reviewers and their decision to include or exclude was checked against the main reviewer's decision. Retrieved studies were filed according to inclusion/exclusion decisions.

[2] Through the generic search we aimed to retrieve studies which explored general measures of physical health (e.g. self-rated health) or a mixture of physical morbidity outcomes (e.g. symptom prevalence rates). The specific search was intended to identify studies that reported on the prevalence of particular health conditions that are common in childhood. In conducting both searches, we hoped to achieve a wide coverage of the research conducted in relation to physical morbidity during childhood and adolescence.

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Inclusion and exclusion criteria

The following inclusion and exclusion criteria were applied to all studies to determine their relevance to the review:

1) Age of participants

As the review focussed on child and adolescent health, studies which included participants between 4 and 17 years old were included. Studies of babies and toddlers (aged 0-3 years) were excluded on the grounds that they are not able to communicate their symptoms verbally in the same way as older children. Studies of those aged 18 years and over were classed as adult studies and therefore not included. As we aimed to look at change in prevalence rates according to age, studies were required to present prevalence data for at least two age-groups within the 4-17 range. Studies which only presented data for one age-group were excluded. Studies were included if they presented data in age-bands of no wider than five years (e.g. 11-14). Studies using age-bands wider than five years were excluded on the grounds that this would prevent us from looking at change in prevalence rates according to age. Studies which used age-bands that included some participants within the 4-17 age-range, such as 0-4 or 15-19, were included. However, if half or more of an individual age-band was not within our age-range, that age-band was excluded from our analyses. For example, 0-4 age-bands were excluded from analysis on the assumption that the majority of participants within that sample would be under four years of age. Often this did not result in the exclusion of studies as they presented prevalence data for at least a further two age-groups.

2) Sex of participants

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The aim of the review was to assess the evidence for an emerging/increasing female excess in morbidity rates, so studies which presented data in relation to both males and females were included. Studies which presented data only in relation to either males or females were excluded.

3) Study design

Empirical studies which used quantitative data collection and analysis methods were included. Longitudinal, cross-sectional, repeat cross-sectional and studies which have analysed routine data (e.g. hospital records) were included. Studies which employed qualitative data collection and analysis methods were not included. Studies which presented only parent-report data were also excluded.

4) Health measures

Studies which presented prevalence data on health measures (e.g. symptom and morbidity rates; health status; incidence of chronic illnesses in childhood etc.) were included in the review. Studies reporting only lifetime prevalence rates were not included because we were interested in current or recent (i.e. within last year) measures of physical morbidity. Studies about injuries or accidents were not included. Studies about health behaviours and symptoms resulting from health behaviours (e.g. impact of alcohol use on depressive symptoms) were also excluded. Studies focussing on dental health were excluded, as were studies which focussed on obesity rates and those which explored rates of symptoms which are the result of traumatic events (e.g. abuse).

5) Countries

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Studies from current EU countries as well as the USA, Canada, Australia and New Zealand were included on the basis that their contextual similarity would aid comparison. Studies from all other countries were excluded.

The above criteria were applied to the titles and abstracts of the articles identified by the literature searches. Hard copies were obtained of all articles which met the inclusion criteria. In cases where inclusion or exclusion could not be determined from titles and abstracts, full papers were retrieved and checked. Each article was labelled in Endnote as to whether it was included or excluded and the number of articles included and excluded at the various stages of the review was recorded systematically.

Data extraction

Data were extracted by one reviewer and three reviewers each independently extracted a sample of studies. Extraction forms were compared across reviewers. The following data were extracted:

1) **Publication details**: author; title; journal; date; primary focus; stated aims.
2) **Focus on emerging/increasing female excess:** mention or not of sex differences/similarities/'gender reversal' in introduction, results or discussion; explanations offered for changes in sex differences with age.
3) **Study details:** methods; sample (source, size, age range and age-groups; representativeness; response rate/completeness); primary outcomes; questions/instruments.
4) **Key data:** any figures for outcomes by sex and age (e.g. prevalence rate/incidence rate, both adjusted and unadjusted, figures extracted as reported in paper (means, OR, RR etc.) with as much detail as possible (95% confidence intervals, chi-square etc.)).

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Assessment of methodological quality

Studies were critically appraised by one reviewer using the criteria below which were agreed by all reviewers. A quality index was developed for each criterion which ranged from 2 (lower potential for bias) to 0 (higher potential for bias). Studies were each given an indicative score for quality. Repeat cross-sectional/cross-sectional and routine data studies were scored out of a maximum of 12 and, due to the extra criterion for attrition rate, longitudinal studies were scored out of 14.[3]

Sample size

2 - Every age and gender sub-group is comprised of at least 100 participants.

1 - Every age and gender sub-group is comprised of at least 50 participants.

0 - <50 in any age and gender sub-group, or data not given.

Large/multi-site population

2 - International, national or statewide (e.g. as in a USA state) study, including multi-site studies in which the sites are spread across international, national or statewide areas.

1 - Local multi-site studies (e.g. same city, town/district, or villages within the same region).

0 – Single-site study (e.g. one school).

[3] In the final review, the median critical appraisal score was 10 out of a possible 14 for longitudinal studies (range 6-12); 8 (out of a possible 12) for (repeat) cross-sectional surveys (range 4-11); and 11 (out of 12) for studies presenting analyses of routinely collected data (range 9-12). These three median scores are used as benchmarks to compare the relative quality of included studies within each type. Thus we describe longitudinal studies, cross-sectional surveys and routine data analysis studies scoring >10, >8 and >11 respectively as 'higher scoring'

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Age-ranges covered

2 - Three or more age points that include under 12 years of age and 12 years or older.

1 - Two age points that include under 12 years of age and 12 years or older.

0 - Age points do not compare those under 12 years of age with those aged 12 years or older.

Selection bias

2 - 80-100% response at baseline *or* routine data that covers at least 80% of population.

1 - 60-79% response at baseline (or routine data coverage) and non-response confounding explored and found not to have a substantial gender or age bias *or* routine data covers 60-79% and no reason to assume age/gender bias in coverage.

0 - Response (or routine data coverage) less than 60%; or less than 80% with evidence of a substantial gender or age bias in attrition; or if non-response is not explored.

Outcome

2 - Physical examination by trained professional.

1 - Self-complete questionnaire using an established/validated questionnaire.

0 - Unvalidated questionnaire or questionnaire designed for study and there is no comment on validation.

Analysis and data reporting

2 - Use odds ratios/incidence rate ratio and 95% confidence intervals to determine whether there is a significant gender-by-age interaction associated with morbidity rates (or sufficient data to calculate ORs, IRRs and CIs).

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1 - Use of alternative (to those above) methods of determining gender-by-age interactions associated with morbidity rates (e.g. continuous data or visual data without confidence intervals).

0 - Data on age, gender or morbidity compromised by unclear reporting or missing data.

Attrition (longitudinal studies only)

2 - Final response is 80%-100% of baseline response.

1 - Final response is 60%-79% of baseline response and attrition confounding explored and found not to have a significant gender or age bias or a bias related to baseline health outcomes.

0 - Final response is <60% of baseline response.

Synthesis

As a meta-analysis was not possible, owing to the heterogeneity of studies, a narrative synthesis method was employed. The studies were grouped by symptoms and conditions as follows: self-assessed health; symptoms (abdominal pain; back pain; dizziness; headache; sleeping difficulties/tiredness); conditions (migraine; diabetes mellitus; epilepsy). Where data were available, odds ratios were calculated (with males serving as the reference group) and studies were tabulated to aid comparison.

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The findings from the review were written up and submitted for publication to an international public health journal. We have so far presented the findings at two national conferences.

Appendix 1 – Specific search diary

Medline (Ovid interface)

06/05/2010

(child, preschool or child or adolescent).sh. and (sex factors or sex distribution).sh. and (asthma or epilepsy or headache disorders, primary or diabetes mellitus).sh.

limit to (english language and humans and yr="1992 -Current")

(.sh. = MeSH subject headings)

Results – 1426

Embase (Ovid interface)

06/05/2010

(child or school child or adolescent or preschool child).sh. and (sex difference or gender).sh. and (asthma or primary headache or migraine or diabetes mellitus or epilepsy).sh.

limit to (human and English language and yr="1992 -Current")

(.sh.= subject headings)

Results – 1526

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CINAHL (Cumulative Index to Nursing and Allied Health) **(EBSCO Host interface)**

07/05/2010

(adolescence or child or child, preschool).sh. and (sex factors).sh. and (asthma or diabetes mellitus or headache or epilepsy).sh.

limit to (english language and yr="1992 -Current")

(.sh.= word in subject heading)

Results – 498

PsycINFO (EBSCO Host interface)

07/05/2010

(child* or adolesc* or young pers* or teenage* or pupil* or school child* or minor*).kw. and (human sex differences or sex).sh. and (asthma or diabetes mellitus or headache or epilepsy).sh.

limit to (english language and yr="1992 -Current")

(.kw. – keywords, .sh.= exact subjects)

Results – 38

ERIC (Education from US Department of Education, and Institute of Education Sciences) **(Ovid interface)**

07/05/2010

((children or young children or adolescents or early adolescents or late adolescents).sh. or (pupil* or school child* or minor*).ab.) and ((sex or gender differences).sh. or (sex or gender).ab.) and ((asthma or headache or migraine).ab. or (diabetes or epilepsy).sh.)

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limit to (english language and yr="1992 -Current")

(.sh.= ERIC subject headings, .ab. = abstract)

Results – 22

Specific search total hits – 3510

Unique hits - 2622

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Appendix 2 – Generic search diary

Medline (Ovid interface)

16/04/2010

(child, preschool or child or adolescent).sh. and (sex factors or sex distribution).sh. and (health status or attitude to health or health surveys or mental health[4]).sh.

limit to (english language and humans and yr="1992 -Current")

(.sh. = MeSH subject headings)

Results – 3587

Embase (Ovid interface)

19/04/2010

(child or school child or adolescent or preschool child).sh. and (sex difference or gender).sh. and (adolescent health or health survey or health status).sh.

limit to (human and English language and yr="1992 -Current")

(.sh.= subject headings)

Results – 2652

CINAHL (Cumulative Index to Nursing and Allied Health) **(EBSCO Host interface)**

19/04/2010

(adolescence or child or child, preschool).sh. and (sex factors).sh. and (health status or health status indicators or attitude to health or symptoms or morbidity or child health or adolescent health).sh.

limit to (english language and yr="1992 -Current")

[4] Note: following this initial search, the decision was made to focus the review on physical rather than mental health.

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Supplemental document 1

(.sh.= word in subject heading)

Results – 1467

PsycINFO (EBSCO Host interface)

19/04/2010

(child* or adolesc* or young pers* or teenage* or pupil* or school child* or minor*).kw. and (human sex differences or sex).sh. and (health or health attitudes or health complaints or general health questionnaire or well being or self report or morbidity or symptoms).sh.

limit to (english language and yr="1992 -Current")

(.kw. – keywords, .sh.= subjects)

Results – 1136

ERIC (Education from US Department of Education, and Institute of Education Sciences) **(Ovid interface)**

(19/04/2010)

((children or young children or adolescents or early adolescents or late adolescents).sh. or (pupil* or school child* or minor*).ab.) and ((sex or gender differences).sh. or (sex or gender).ab.) and ((health or child health or adolescent health or well being or wellness).sh. or (morbidity or symptom*).ab.)

limit to (english language and yr="1992 -Current")

(.sh.= ERIC subject headings, .ab. = abstract)

Results – 593

Generic search total hits – 9435

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Unique hits - 8623

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References

Almqvist, C., M. Worm, et al. (2008). "Impact of gender on asthma in childhood and adolescence: a GA2LEN review." Allergy **63**(1): 47-57.

Bennett, D. S., P. J. Ambrosini, et al. (2005). "Gender differences in adolescent depression: Do symptoms differ for boys and girls?" Journal of Affective Disorders **89**: 35-44.

Cohen, P., J. Cohen, et al. (1993). "An epidemiological study of disorders in late childhood and adolescence: I. Age and gender-specific prevalence." Journal of Child Psychology and Psychiatry **34**(6): 851-867.

Cyranowski, J. M., E. Frank, et al. (2000). "Adolescent onset of the gender difference in lifetime rates of major depression." Archives of general psychiatry **57**: 21-27.

Eiser, C., T. Havermans, et al. (1995). "The emergence during adolescence of gender differences in symptom reporting." Journal of Adolescence **18**(3): 307-315.

Eminson, M., S. Benjamin, et al. (1996). "Physical symptoms and illness attitudes in adolescents: An epidemiological study." Journal of Child Psychology and Psychiatry **37**(5): 519-528.

Ge, X., R. D. Conger, et al. (2001). "Pubertal transition, stressful life events, and the emergence of gender differences in adolescent depressive symptoms." Developmental Psychology **37**(3): 404-417.

Hankin, B. L. and L. Y. Abramson (1999). "Development of gender differences in depression: description and possible explanations." Annals of Medicine **31**(6): 372-379.

Haugland, S., B. Wold, et al. (2001). "Subjective health complaints in adolescence: A cross-national comparison of prevalence and dimensionality." European Journal of Public Health **11**(1): 4-10.

Hetland, J., T. Torsheim, et al. (2002). "Subjective health complaints in adolescence: dimensional structure and variation across gender and age." Scandinavian Journal of Public Health **30**(3): 223-230.

Klepp, K. I., H. N. Aas, et al. (1996). "Self-reported health problems among school pupils." Tidsskr Nor Laegeforen **116**: 2032-2037.

Marcotte, D., L. Fortin, et al. (2002). "Gender differences in depressive symptoms during adolescence: role of gender-typed characteristics, self-esteem, body image, stressful life events, and pubertal status." Journal of Emotional and Behavioural Disorders **10**(1): 29-43.

Nicolai, T., L. Pereszlenyiova-Bliznakova, et al. (2003). "Longitudinal follow-up of the changing gender ratio in asthma from childhood to adulthood: role of delayed manifestation in girls." Pediatric Allergy & Immunology **14**(4): 280-283.

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Nolen-Hoeksema, S. and J. S. Girgus (1994). "The Emergence of Gender Differences in Depression During Adolescence." Psychological Bulletin **115**(2): 424-443.

Petersen, A. C., P. A. Sarigiani, et al. (1991). "Adolescent Depression: Why More Girls?" Journal of Youth and Adolescence **20**(2): 247-271.

Postma, D. S. (2007). "Gender differences in asthma development and progression." Gender Medicine **4 Suppl B**: S133-146.

Schraedley, P. K., I. H. Gotlib, et al. (1999). "Gender Differences in Correlates of Depressive Symptoms in Adolescence." Journal of Adolescent Health **25**: 98-108.

Sears, M. R., J. M. Greene, et al. (2003). "A longitudinal, population-based, cohort study of childhood asthma followed to adulthood." New England Journal of Medicine **349**(15): 1414-1422.

Shibley Hyde, J., A. H. Mezulis, et al. (2008). "The ABCs of Depression: Integrating affective, biological and cognitive models to explain the emergence of the gender difference in depression." Psychological Review **115**(2): 219-313.

Sweeting, H. (1995). "Reversals of fortune? Sex differences in health in childhood and adolescence." Social Science & Medicine **40**(1): 77-90.

Sweeting, H. and P. West (2003). "Sex differences in health at ages 11, 13 and 15." Social Science & Medicine **56**(1): 31-39.

Torsheim, T., U. Ravens-Sieberer, et al. (2006). "Cross-national variation of gender differences in adolescent subjective health in Europe and North America." Social Science & Medicine **62**(4): 815-827.

Venn, A., S. Lewis, et al. (1998). "Questionnaire study of effect of sex and age on the prevalence of wheeze and asthma in adolescence." BMJ **316**(7149): 1945-1946.

Zannolli, R. and G. Morgese (1997). "Does puberty interfere with asthma?" Medical Hypotheses **48**(1): 27-32.

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Supplemental document 2

Supplemental document: Studies that were included in the final review, identified from the generic and specific literature searches

Table 2: Studies identified by both the generic and specific searches

Author, date	Title	Journal	Health outcome	Design	Appraisal score[1] (+ = higher scoring)	Country
Gordon et al, 2004[1]	Prevalence of reported migraine headaches in Canadian adolescents.	Canadian Journal of Neurological Sciences	Migraine.	Cross-sectional	7	Canada
Petersen et al, 2003[2]	High prevalence of tiredness and pain in young school-children.	Scandinavian Journal of Public Health	Backache; headache; stomach ache; tiredness.	Cross-sectional	9 (+)	Sweden
Rhee et al, 2005[3]	Prevalence of recurrent physical symptoms in US adolescents.	Pediatric Nursing	Chest pain; cold sweat; dizziness; fatigue; feeling hot; frequent sore throat/cough; headache; stomach ache; musculoskeletal pain; painful/frequent urination.	Cross-sectional	8 (+)	USA

[1] Note: appraisal score criteria and range varied by study design – see protocol.

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Table 3: Studies identified by the generic health search only.

Author, date	Title	Journal	Health Outcome	Design	Appraisal score[1] (+ = higher scoring)	Country
Bigal et al, 2007[4]	Migraine in adolescents: Association with socioeconomic status and family history.	Neurology	Migraine.	Cross-sectional	6	USA
Bisegger et al, 2005[5]	Health-related quality of life: gender differences in childhood and adolescence.	Sozial- und Praventivmedizin	Health related quality of life.	Cross-sectional	7	Austria, France, Germany, Spain, Switzerland, UK ; Netherlands
Cavallo et al, 2006[6]	Girls growing through adolescence have a higher risk of poor health.	Quality of Life Research	Backache; difficulties in sleeping; feeling dizzy; feeling low; feeling nervous; headache; irritability and bad temper; self-rated health; stomach ache.	Cross-sectional	9 (+)	Europe and North America
*Gadin & Hammarstrom, 2000[7]	School-related health – A cross-sectional study among boys and girls.	International Journal of Health	Abdominal pain; backache; headache; nausea; self-worth;	Cross-sectional	8	Sweden

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		Services	stress; tiredness.			
Grimmer et al, 2006[8]	Longitudinal investigation of low back pain in Australian adolescents: a five-year study.	Physiotherapy Research International	Low back pain.	Longitudinal	6	Australia
*Haugland et al, 2001[9]	Subjective health complaints in adolescence. A cross-national comparison of prevalence and dimensionality.	European Journal of Public Health	Abdominal pain; backache; dizziness; feeling low; feeling nervous; headache; irritability or bad mood; sleeping difficulties.	Cross-sectional	11 (+)	Finland, Norway, Poland and Scotland
Holmberg, & Hellberg, 2007[10]	Age-related gender differences of relevance for health in Swedish adolescents.	International Journal of Adolescent Medicine & Health	Abdominal pain; feeling depressed; feeling healthy; headache; suicidal thoughts.	Cross-sectional	8	Sweden
Jorngarden et al, 2006[11]	Measuring health-related quality of life in adolescents and young adults: Swedish normative data for the SF-36 and the HADS, and the influence of age, gender and method of administration.	Health & Quality of Life Outcomes	Anxiety; depression; health related quality of life.	Cross-sectional	5	Sweden
Kujala et al, 1999[12]	Leisure physical activity and various pain symptoms among adolescents.	British Journal of Sports Medicine	Abdominal pain; headache; lower back pain; lower limb pain; neck and shoulder pain; upper back pain; upper	Cross-sectional	5	Sweden

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			limb pain.			
Laaksonen et al, 2010[13]	The change in child self-assessed and parent-proxy assessed health related quality of life in early adolescence (age 10-12).	Scandinavian Journal of Public Health	Health related quality of life.	Longitudinal	10	Finland
Lundqvist et al, 2006[14]	Self-reported headache in schoolchildren: parents underestimate their children's headaches.	Acta Paediatrica	Headache.	Cross-sectional	8	Norway
Meland et al, 2007[15]	Body image and perceived health in adolescence.	Health Education Research	Body image; perceived health.	Cross-sectional	8	Norway
Ostberg et al, 2006[16]	Living conditions and psychosomatic complaints in Swedish schoolchildren.	Acta Paediatrica	Difficulties falling asleep; headache; stomach ache.	Cross-sectional	9 (+)	Sweden
Palacio-Vieira et al, 2008[17]	Changes in health-related quality of life in a population-based sample of children and adolescents after 3 years follow-up.	Quality of Life Research	Health related quality of life.	Longitudinal	7	Spain
Ravens-Sieberer et al, 2008[18]	Health-related quality of life in children and adolescents in Germany: results of the BELLA study.	European Child & Adolescent Psychiatry	Health related quality of life.	Cross-sectional	8	Germany
Skordis et al, 2002[19]	The incidence of type 1 diabetes mellitus in Greek-Cypriot children and adolescents in 1990-	Pediatric Diabetes	Type 1 diabetes mellitus.	Routine data	11	Cyprus

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	2000.					
Sleskova et al, 2005[20]	Health status among young people in Slovakia: comparisons on the basis of age, gender and education.	Social Science & Medicine	Health complaints (backache; bone/muscle ache; breathlessness; chest/heart pain; dizziness; full/bloated stomach; headache; listlessness; pins and needles; tiredness;upset stomach); mental health; long-standing illness; long-term wellbeing; self-rated health; vitality.	Cross-sectional	7	Slovakia
Sundblad et al, 2007[21]	Prevalence and co-occurrence of self-rated pain and perceived health in school-children: age and gender differences.	European Journal of Pain	Abdominal pain; headache; loneliness; musculoskeletal pain; problems sleeping; sadness; tiredness.	Cross-sectional	11 (+)	Sweden
*Sweeting & West, 2003[22]	Sex differences in health at ages 11, 13 and 15.	Social Science & Medicine	Depression; general health; recent symptoms (aching back, legs or arms; asthma or wheeze; cold or flu; difficulty getting to sleep; dizzy or faint; headache; irritable or bad tempered;	Longitudinal	12 (+)	Scotland

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			nervous, worried or anxious; sad, unhappy or low; spots, rashes or other skin problems; stomach ache or feeling sick).			
*Torsheim et al, 2006[23]	Cross-national variation of gender differences in adolescent subjective health in Europe and North America.	Social Science & Medicine	Health complaints (backache; depressed mood; dizziness; headache; irritable; nervousness; stomach ache; sleeping difficulties).	Cross-sectional	9 (+)	Europe and North America (29 countries)
Wedderkopp et al, 2001[24]	Back pain reporting pattern in a Danish population-based sample of children and adolescents.	Spine	Back pain; neck pain.	Cross-sectional	8	Denmark

*Studies that were also identified through manual searching

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Table 4: Studies identified by the specific health search only

Author, date	Title	Journal	Health outcome	Design	Appraisal score[1] (+ = higher scoring)	Country
Beilmann et al, 1999[25]	Incidence of childhood epilepsy in Estonia.	Brain & Development	Epilepsy.	Routine data	12 (+)	Estonia
Carle et al, 2004[26]	Diabetes incidence in 0 to 14-year age group in Italy. A 10-year prospective study.	Diabetes Care	Type 1 diabetes mellitus.	Routine data	12 (+)	Italy
Casu et al, 2004[27]	Type 1 diabetes among Sardinian Children is increasing. The Sardinian diabetes register for children aged 0-14 years (19889-1999).	Diabetes Care	Type 1 diabetes mellitus.	Routine data	10	Sardinia
Christensen et al, 2007[28]	Incidence and prevalence of epilepsy in Denmark.	Epilepsy Research	Epilepsy.	Routine data	9	Denmark
Cinek et al, 2000[29]	Type 1 diabetes mellitus in Czech children diagnosed in 1990-1997: a significant increase in incidence and male predominance in the age group 0-4 years.	Diabetic Medicine	Type 1 diabetes mellitus.	Routine data	11	Czech Republic

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Cotellessa et al, 2003[30]	High incidence of type 1 diabetes in Liguria Italy, from 1989 to 1998.	Diabetes Care	Type 1 diabetes mellitus.	Routine data	11	Italy
Freitag et al, 2001[31]	Incidence of epilepsies and epileptic syndromes in children and adolescents: a population-based prospective study in Germany.	Epilepsia	Epilepsy.	Routine data	10	Germany
Heinrich et al, 2009[32]	Self-report of headache in children and adolescents in Germany: possibilities and confines of questionnaire data for headache classification.	Cephalalgia	Headache; migraine.	Cross-sectional	9 (+)	Germany
Karvonen et al, 1999[33]	The onset age of type 1 diabetes in Finnish children has become younger.	Diabetes Care	Type 1 diabetes mellitus.	Routine data	11	Finland
Larsson & Sund, 2005[34]	One-year incidence, course and outcome predictors of frequent headaches among early adolescents.	Headache	Headache.	Longitudinal	10	Norway
Laurell et al, 2004[35]	Prevalence of headache in Swedish schoolchildren, with a focus on tension-type headache.	Cephalalgia	Headache; migraine.	Cross-sectional	8	Sweden
Leonardsson-Hellgren et al, 2001[36]	Headache and associations with lifestyle among pupils in senior level elementary school.	Scandinavian Journal of Primary Health Care	Headache.	Cross-sectional	5	Sweden

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Mavromichalis et al, 1999[37]	Prevalence of migraine in schoolchildren and some clinical comparisons between migraine with and without aura.	Headache	Migraine.	Cross-sectional	9 (+)	Greece
Michalkova et al, 1995[38]	Incidence and prevalence of childhood diabetes in Slovakia (1985-1992).	Diabetes Care	Type 1 diabetes mellitus.	Routine data	11	Slovakia
*Mortimer et al, 1992[39]	Epidemiology of headache and childhood migraine in an urban general practice using ad hoc, Vahlquist and IHS criteria.	Developmental Medicine and Child Neurology	Headache; migraine.	Cross-sectional	6	UK
Mortimer et al, 1993[40]	Clinical epidemiology of childhood abdominal migraine in an urban general practice.	Developmental Medicine and Child Neurology	Abdominal migraine; headache; recurrent abdominal pain.	Cross-sectional	5	UK
Santinello et al, 2008[41]	Primary headache in Italian early adolescents: the role of perceived teacher unfairness.	Headache	Headache.	Cross-sectional	9 (+)	Italy

* This study was also referenced in a different publication identified via the generic ELS.

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References

1. Gordon KE, Dooley JM, Wood EP. Prevalence of reported migraine headaches in Canadian adolescents. *Canadian Journal of Neurological Sciences* 2004;31(3):324-7.
2. Petersen S, BergstrÃ¶m E, Brulin C. High prevalence of tiredness and pain in young schoolchildren. *Scandinavian Journal of Public Health* 2003;31(5):367-74.
3. Rhee H, Miles MS, Halpern CT, Holditch-Davis D. Prevalence of recurrent physical symptoms in U.S. adolescents. *Pediatric Nursing* 2005;31(4):314.
4. Bigal ME, Lipton RB, Winner P, Reed ML, Diamond S, Stewart WF, et al. Migraine in adolescents: association with socioeconomic status and family history. *Neurology* 2007;69(1):16-25.
5. Bisegger C, Cloetta B, von Rueden U, Abel T, Ravens-Sieberer U, European Kidscreen G. Health-related quality of life: gender differences in childhood and adolescence. *Sozial- und Praventivmedizin* 2005;50(5):281-91.
6. Cavallo F, Zambon A, Borraccino A, Raven-Sieberer U, Torsheim T, Lemma P, et al. Girls growing through adolescence have a higher risk of poor health. *Quality of Life Research* 2006;15(10):1577-85.
7. Gadin KG, Hammarstrom A. School-related health - A cross-sectional study among young boys and girls. *International Journal of Health Services* 2000;30(4):797-820.
8. Grimmer K, Nyland L, Milanese S. Longitudinal investigation of low back pain in Australian adolescents: a five-year study. *Physiotherapy Research International* 2006;11(3):161-72.
9. Haugland S, Wold B, Stevenson J, Aaroe LE, Woynarowska B. Subjective health complaints in adolescence: A cross-national comparison of prevalence and dimensionality. *European Journal of Public Health* 2001;11(1):4-10.
10. Holmberg LI, Hellberg D. Age-related gender differences of relevance for health in Swedish adolescents. *International Journal of Adolescent Medicine & Health* 2007;19(4):447-57.
11. Jorngarden A, Wettergen L, von Essen L, Jorngarden A, Wettergen L, von Essen L. Measuring health-related quality of life in adolescents and young adults: Swedish normative data for the SF-36 and the HADS, and the influence of age, gender, and method of administration. *Health & Quality of Life Outcomes* 2006;4:91.
12. Kujala UM, Taimela S, Viljanen T. Leisure physical activity and various pain symptoms among adolescents. *British Journal of Sports Medicine* 1999;33(5):325-8.
13. Laaksonen CB, Aromaa ME, Asanti RE, Heinonen OJ, Koivusilta LK, Koski PJ, et al. The change in child self-assessed and parent proxy-assessed Health Related Quality of Life (HRQL) in early adolescence (age 10-12). *Scandinavian Journal of Public Health* 2010;38(1):9-16.
14. Lundqvist C, Clench-Aas J, Hofoss D, Bartonova A, Lundqvist C, Clench-Aas J, et al. Self-reported headache in schoolchildren: parents underestimate their children's headaches. *Acta Paediatrica* 2006;95(8):940-6.
15. Meland E, Haugland S, Breidablik HJ, Meland E, Haugland S, Breidablik H-J. Body image and perceived health in adolescence. *Health Education Research* 2007;22(3):342-50.

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Supplemental document 2

16. Ostberg V, Alfven G, Hjern A, Ostberg V, Alfven G, Hjern A. Living conditions and psychosomatic complaints in Swedish schoolchildren. *Acta Paediatrica* 2006;95(8):929-34.
17. Palacio-Vieira JA, Villalonga-Olives E, Valderas JM, Espallargues M, Herdman M, Berra S, et al. Changes in health-related quality of life (HRQoL) in a population-based sample of children and adolescents after 3 years of follow-up. *Quality of Life Research* 2008;17(10):1207-15.
18. Ravens-Sieberer U, Erhart M, Wille N, Bullinger M, group Bs, Ravens-Sieberer U, et al. Health-related quality of life in children and adolescents in Germany: results of the BELLA study. *European Child & Adolescent Psychiatry* 2008;17 Suppl 1:148-56.
19. Skordis N, Theodorou S, Apsiotou T, Stavrou S, Herakleous E, Savva SC. The incidence of type 1 diabetes mellitus in Greek-Cypriot children and adolescents in 1990-2000. *Pediatric Diabetes* 2002;3(4):200-04.
20. Sleskova M, Salonna F, Madarasova Geckova A, van Dijk JP, Groothoff JW. Health status among young people in Slovakia: comparisons on the basis of age, gender and education. *Social Science & Medicine* 2005;61(12):2521-7.
21. Sundblad GMB, Saartok T, Engstrom LMT. Prevalence and co-occurrence of self-rated pain and perceived health in school-children: Age and gender differences. *European Journal of Pain* 2007;11(2):171-80.
22. Sweeting H, West P. Sex differences in health at ages 11, 13 and 15. *Social Science & Medicine* 2003;56(1):31-9.
23. Torsheim T, Ravens-Sieberer U, Hetland J, Valimaa R, Danielson M, Overpeck M, et al. Cross-national variation of gender differences in adolescent subjective health in Europe and North America. *Social Science & Medicine* 2006;62(4):815-27.
24. Wedderkopp N, Leboeuf-Yde C, Andersen LB, Froberg K, Hansen HS. Back pain reporting pattern in a Danish population-based sample of children and adolescents. *Spine* 2001;26(17):1879-83.
25. Beilmann A, Napa A, Hamarik M, Soot A, Talvik I, Talvik T. Incidence of childhood epilepsy in Estonia. *Brain & Development* 1999;21(3):166-74.
26. Carle F, Gesuita R, Bruno G, Coppa GV, Falorni A, Lorini R, et al. Diabetes incidence in 0-to 14-year age-group in Italy: a 10-year prospective study. *Diabetes Care* 2004;27(12):2790-96.
27. Casu A, Pascutto C, Bernardinelli L, Songini M. Type 1 diabetes among Sardinian children is increasing: The Sardinian diabetes register for children aged 0-14 years (1989-1999). *Diabetes Care* 2004;27(7):1623-29.
28. Christensen J, Vestergaard M, Pedersen MG, Pedersen CB, Olsen J, Sidenius P, et al. Incidence and prevalence of epilepsy in Denmark. *Epilepsy Research* 2007;76(1):60-5.
29. Cinek O, Lanska V, Kolouskova S, Sumnik Z, Snajderova M, Ronningen KS, et al. Type I diabetes mellitus in Czech children diagnosed in 1990-1997: A significant increase in incidence and male predominance in the age group 0-4 years. *Diabetic Medicine* 2000;17(1):64-69.
30. Cotellessa M, Barbieri P, Mazzella M, Bonassi S, Minicucci L, Lorini R. High incidence of childhood type 1 diabetes in Liguria, Italy, from 1989 to 1998. *Diabetes Care* 2003;26(6):1786-89.
31. Freitag CM, May TW, Pfafflin M, Konig S, Rating D. Incidence of epilepsies and epileptic syndromes in children and adolescents: a population-based prospective study in Germany. *Epilepsia* 2001;42(8):979-85.
32. Heinrich M, Morris L, Kroner-Herwig B. Self-report of headache in children and adolescents in Germany: Possibilities and confines of questionnaire data for headache classification. *Cephalalgia* 2009;29(8):864-72.

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Supplemental document 2

33. Karvonen M, Pitkaniemi J, Tuomilehto J. The onset age of type 1 diabetes in Finnish children has become younger. *Diabetes Care* 1999;22(7):1066-70.
34. Larsson B, Sund AM. One-year incidence, course, and outcome predictors of frequent headaches among early adolescents. *Headache: The Journal of Head & Face Pain* 2005;45(6):684-91.
35. Laurell K, Larsson B, Eeg-Olofsson O. Prevalence of headache in Swedish schoolchildren, with a focus on tension-type headache. *Cephalalgia* 2004;24(5):380-88.
36. Leonardsson-Hellgren M, Gustavsson U, Lindblad U. Headache and associations with lifestyle among pupils in senior level elementary school. *Scandinavian Journal of Primary Health Care* 2001;19(2):107-11.
37. Mavromichalis I, Anagnostopoulos D, Metaxas N, Papanastassiou E. Prevalence of migraine in schoolchildren and some clinical comparisons between migraine with and without aura. *Headache* 1999;39(10):728-36.
38. Michalkova DM, Cernay J, Dankova A, Rusnak M, Fandakova K. Incidence and prevalence of childhood diabetes in Slovakia (1985-1992). *Diabetes Care* 1995;18(3):315-20.
39. Mortimer MJ, Kay J, Jaron A. Epidemiology of headache and childhood migraine in an urban general practice using Ad Hoc, Vahlquist and IHS criteria. *Developmental Medicine and Child Neurology* 1992;34(12):1095-101.
40. Mortimer MJ, Kay J, Jaron A. Clinical epidemiology of childhood abdominal migraine in an urban general practice. *Developmental Medicine and Child Neurology* 1993;35(3):243-48.
41. Santinello M, Vieno A, De Vogli R, Santinello M, Vieno A, De Vogli R. Primary headache in Italian early adolescents: the role of perceived teacher unfairness. *Headache* 2009;49(3):366-74.

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STROBE Statement—checklist of items that should be included in reports of observational studies, followed by a table showing how the current study conforms to the STROBE statement.

	Item No	Recommendation
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found
Introduction		
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported
Objectives	3	State specific objectives, including any prespecified hypotheses
Methods		
Study design	4	Present key elements of study design early in the paper
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection
Participants	6	(*a*) *Cohort study*—Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up *Case-control study*—Give the eligibility criteria, and the sources and methods of case ascertainment and control selection. Give the rationale for the choice of cases and controls *Cross-sectional study*—Give the eligibility criteria, and the sources and methods of selection of participants
		(*b*) *Cohort study*—For matched studies, give matching criteria and number of exposed and unexposed *Case-control study*—For matched studies, give matching criteria and the number of controls per case
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group
Bias	9	Describe any efforts to address potential sources of bias
Study size	10	Explain how the study size was arrived at
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding
		(*b*) Describe any methods used to examine subgroups and interactions
		(*c*) Explain how missing data were addressed
		(*d*) *Cohort study*—If applicable, explain how loss to follow-up was addressed *Case-control study*—If applicable, explain how matching of cases and controls was addressed *Cross-sectional study*—If applicable, describe analytical methods taking account of sampling strategy
		(*e*) Describe any sensitivity analyses

Continued on next page

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Results		
Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed
		(b) Give reasons for non-participation at each stage
		(c) Consider use of a flow diagram
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders
		(b) Indicate number of participants with missing data for each variable of interest
		(c) *Cohort study*—Summarise follow-up time (eg, average and total amount)
Outcome data	15*	*Cohort study*—Report numbers of outcome events or summary measures over time
		Case-control study—Report numbers in each exposure category, or summary measures of exposure
		Cross-sectional study—Report numbers of outcome events or summary measures
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included
		(*b*) Report category boundaries when continuous variables were categorized
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses
Discussion		
Key results	18	Summarise key results with reference to study objectives
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence
Generalisability	21	Discuss the generalisability (external validity) of the study results
Other information		
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.

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STROBE Checklist: How to avoid missing half the evidence: comparing the use of generic and specific electronic search terms used to identify health outcomes for a systematic review.

STROBE Item No.	Authors comment
1 (a)	Done – see abstract, main article
1 (b)	Done – see abstract, main article
2	Done – main article (pages 4-6)
3	Done – main article (page 6)
4	Done – main article (page 8)
5	Done – a literature search is internet based rather than set in a specific location so we gave details of the databases searched and the review that the searches were conducted for. Main article (pages 8-9).
6 (a)	Done – main article (pages 8-10).
6 (b)	Not applicable – the study includes no matching of the kind described in STROBE.
7	Done – main article (pages 11).
8	Done – main article (pages 11).
9	Done – use of multiple reviewers during search and selection process (page 10).
10	Not applicable - the study did not require a power calculation as it includes no participants (in the conventional use of the terms). The text does state that the two searches were field tested during an actual systematic review – main article (pages 8-10) and protocol.
11	Done – main article (page 11).
12 a to e	Done – main article (pages 11).
13	Done – figure 1 and text in main article (pages 11-12)
14 a	Done – table 1, main article (page12)
14 b and c	Not applicable
15	Not applicable
16	Done in so far as applicable (the study does not involve estimates, statistical adjustment or missing data as described by STROBE). Main article (page 11-13).
17	Done – main article (page 11-13).
18	Done – main article (page 13)
19	Done – main article (page 14-15)
20	Done - main article (page 15-16)
21	Done - main article (page 15-16)
22	Done - main article (page 16-17)

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COMPARING THE EFFECTIVENESS OF USING GENERIC AND SPECIFIC SEARCH TERMS IN ELECTRONIC DATABASES TO IDENTIFY HEALTH OUTCOMES FOR A SYSTEMATIC REVIEW: A PROSPECTIVE COMPARATIVE STUDY OF LITERATURE SEARCH METHODS.

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COMPARING THE EFFECTIVENESS OF USING GENERIC AND SPECIFIC SEARCH TERMS IN ELECTRONIC DATABASES TO IDENTIFY HEALTH OUTCOMES FOR A SYSTEMATIC REVIEW: A PROSPECTIVE COMPARATIVE STUDY OF LITERATURE SEARCH METHODS.

Matt Egan, *senior investigator scientist*[1*], Alice MacLean, *investigator scientist*[1], Helen Sweeting, *senior investigator scientist*[1], Kate Hunt, *programme leader*[1].

[1] MRC / CSO Social and Public Health Sciences Unit (SPHSU), 4 Lilybank Gardens, Glasgow G12 8RZ.

[*]Correspondence to m.egan@sphsu.mrc.ac.uk

Running title: 'How to avoid missing half the evidence'

Key words: Systematic review, literature search, evidence informed policy.

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Abstract

Objective

To compare the effectiveness of systematic review literature searches that use either generic or specific terms for health outcomes.

Design

Prospective comparative study of two electronic literature search strategies. The 'generic' search included general terms for health such as 'adolescent health', 'health status', 'morbidity', etc. The 'specific' search focused on terms for a range of specific illnesses, such as 'headache', 'epilepsy', 'diabetes mellitus', etc.

Data sources

We searched Medline, Embase, the Cumulative Index to Nursing and Allied Health Literature (CINAHL), PsycINFO, and the Education Resources Information Center (ERIC) for studies published in English between 1992 and April 2010.

Main outcome measures

Number and proportion of studies included in the systematic review that were identified from each search.

Results

The two searches tended to identify different studies. Out of 41 studies included in the final review, only 3 (7%) were identified by both search strategies; 21 (51%) were identified by the generic search only; and 17 (41%) were identified by the specific search only. Five of the 41 studies were also identified through manual searching methods. Studies identified by the two ELS differed in terms of reported health

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outcomes, whilst each ELS uniquely identified some of the review's higher quality studies.

Conclusions

Electronic literature searches (ELS) are a vital stage in conducting systematic reviews and therefore have an important role in attempts to inform and improve policy and practice with the best available evidence. Whilst the use of both generic and specific health terms is conventional for many reviewers and information scientists, there are also reviews that rely solely on either generic or specific terms. Based on our findings, reliance on only the generic or specific approach could increase the risk of systematic reviews missing important evidence and, consequently, misinforming decision-makers. However, future research should test the generalisability of these findings.

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Introduction

Electronic literature searches (ELS) are an essential stage in most systematic reviews.[1-2] As such, they have a crucial role in the scientific community's attempts to inform and improve policy and practice with the best available evidence.[3-4] Designing ELS can be challenging and it is widely recognized that specialist skills and knowledge, such as those provided by an information scientist, are important for best practice in this field.[1-3] The trade-off between screening out irrelevant evidence whilst identifying relevant evidence (sometimes discussed in terms of a search's 'precision' and 'sensitivity') is a well known challenge for information scientists and researchers who work on systematic reviews. In this paper we present a worked example of how an empirical study comparing different ELS can be conducted to explore the effects that different search strategies may have on the identification of studies for a systematic review, and how this in turn may affect the review's conclusions.

Systematic reviews vary in terms of subject matter and approach[3] and this can have implications for how ELSs are designed. Some systematic reviews are based on comprehensive searches which aim to have high sensitivity and retrieve references to all relevant papers, whereas others are based on more restricted searches which may limit the number of relevant papers identified.[5] Search strategies that are insufficiently sensitive may risk encouraging potentially harmful decisions based on the findings of reviews that have failed to identify important evidence. Search strategies that aim to comprehensively identify all the relevant evidence can present challenges in situations where reviewers have limited time or other resources (e.g. as a result of research funding requirements, or because findings are considered to be needed urgently), or

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when extending a search fails to identify relevant evidence and might therefore represent an ineffective allocation of scarce resources.[3-5]

Previous research exploring how to improve the effectiveness and efficiency of search strategies has tended to focus on issues such as how to optimise search outputs from 'frontline' electronic databases (i.e. databases that are frequently searched for systematic reviews of medical interventions such as Medline and Embase), and how to identify randomised control trials (RCTs).[6-12] This research focus may in part reflect the influence of the Cochrane Collaboration, which has helped to stimulate considerable interest in systematic reviews of clinical trials.[1]

However, not all systematic reviews (nor indeed all Cochrane Reviews[13]) focus on RCTs of clinical interventions. Interest in broader, non-clinical systematic reviews has steadily increased within the social and public health sciences and other disciplines.[3 5] As some of these non-clinical reviews tackle relatively under-researched topics, they often combine a scoping and hypothesis testing function by asking relatively broad research questions that, for example, cover a range of outcomes (e.g. *what are the health impacts of intervention x?; what health outcomes are associated with risk-factor y?*).[14-27] Evidence-informed guidance on how to conduct searches for this broader range of systematic reviews is therefore an emerging priority.

There are few examples of research that can help guide information scientists and reviewers to develop efficient but effective search strategies for these broader / non-clinical systematic reviews. The research that is available illustrates how searches for such reviews can become lengthy and complex.[28] For example Greenhaulgh et al

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recommended the development of iterative search strategies to search for complex evidence (e.g. multiple study designs). Ogilvie et al suggested that cross-disciplinary reviews may necessitate searching databases across a range of disciplines rather than focusing on frontline health databases.[4]

From our own experiences of conducting systematic reviews of non-clinical, public health research, the authors of this paper can identify additional challenges that have led to large and complex ELS. For example, search terms that involve commonly used words are likely to identify large numbers of irrelevant papers and non-clinical public health reviews often rely on commonly used terms to describe everyday settings, activities and outcomes (e.g. 'walking', 'obesity', 'stress', 'workplace health', 'health promotion', and 'general health'). In comparison, an ELS for a clinical review will often involve very specific medical terminology that can help to focus the search on papers relevant to a particular field.[3]

Furthermore, the Cochrane Handbook[1] (section 6.4.2) states that a search strategy to identify studies for a Cochrane review "typically has three sets of terms: (1) terms to search for the health condition of interest, i.e. the population; (2) terms to search for the intervention(s) evaluated; and 3) terms to search for the types of study design to be included (typically a 'filter' for randomized trials)". Each of these sets of terms can help to filter out unwanted studies from the search, but it is not always appropriate or possible to structure an ELS in this way. Systematic reviews do not always include populations defined by a health condition (they may, for example, focus on studies of the general population). As stated earlier, not all systematic reviews are based on evaluations of interventions. Furthermore, not all systematic reviews focus on RCTs

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and some include a range of study designs. Systematic reviewers recognise that it is sometimes appropriate to deviate from this typical search structure: for example, the Cochrane Handbook states that in some circumstances it may be necessary to search "only for the population or the intervention" (Cochrane Handbook[1] section 6.4.2).

The chances of an ELS identifying irrelevant studies could be increased if the search includes both specialist and non specialist databases, or uses search terms based on unspecialised vocabulary, or cannot include terms for population types or interventions or study designs to help screen out irrelevant literature. Searches characterised by a large number of search results and low precision may be resource intensive and this could become a problem if the resources required for a search outstrip what is available for a particular review. In such circumstances, reviewers may look for alternative means of increasing precision. However, for the broader public health reviews of the kind we have described here, there is relatively little evidence based guidance on how greater precision can be achieved without compromising sensitivity (compared to the guidance on clinical/RCT systematic reviews).

Including search terms that relate to health outcomes is one commonly used technique for increasing precision in broader reviews.[14-24] However, if a review question is broad enough to include multiple health outcomes it is not obvious how an ELS that includes health outcomes can best accommodate this breadth of scope. Some reviews have used generic health terms (e.g. 'health', 'illness', 'morbidity') to search for evidence that includes a range of health outcomes.[14-17] In other cases, reviewers have used more specific search terms to identify a number of diseases or symptoms

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considered to be of particular relevance to the review question.[18-21] Both approaches may be hypothesized to have risks. Generic search terms may either be too inclusive (virtually every study on Medline is about 'health') or may miss studies that only use more specialist vocabulary to describe a particular illness. Specific search terms are problematic if the reviewers want to avoid pre-specifying which health outcomes are relevant to the review (e.g. scoping reviews). Some reviews combine both generic and specific approaches[22-24], but the extent to which this either adds value to the search or merely adds to the workload is not known.

We know of no study that has compared the relative merits of ELS strategies that focus on either generic terms for health, or specific terms for particular health issues or illnesses. Nor do we know of any evidence to help reviewers determine whether these two approaches are likely to identify a similar or a different set of publications (both of the above observations are based on a non-systematic exploration of the literature rather than a systematic review). When the authors of this paper recently conducted a systematic review that included multiple health outcomes, we felt that guidance on this issue would have been helpful. As there was an absence of evidence upon which to base such guidance, we ran two separate literature searches for our review: one that included generic health terms and one that used more specific health terms. Our aim was to see which approach was most effective in identifying studies that were included in the final review.

Hence, we examined whether the included studies tended to be identified from the generic search only, the specific search only, or both searches. We also explored efficiency by comparing the size of the searches (i.e. the number of references initially

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identified from the ELS – sometimes referred to as the number of 'hits') for each approach. Finally, we explored the extent to which the 'generic search' and the 'specific search' identified studies with different or similar types of health outcome.

Our review was conducted within a limited time frame (originally planned as nine months and then extended to 18 months), and we believe the implications of this study are of particular relevance to reviews of broader public health topics and reviews with time or other resource limitations.

Methods

This paper focuses on one specific, but crucial, stage of a systematic review: the literature search. We developed two contrasting strategies for searching electronic databases and compared their effectiveness in identifying studies for a specific systematic review. The systematic review itself is summarised in Panel 1, and described more fully in the publically available Protocol document (available as a supplemental document online), and the full report of the review which will be published separately to this methodological paper.

Panel 1. Summary of the systematic review used as the basis of this methodological study.

Title: How robust is the evidence of an emerging or increasing female excess in physical morbidity rates between childhood and adolescence? Results of a systematic literature review.

Hypothesis: That the incidence of physical morbidity amongst children tends to be

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higher amongst males in pre-adolescent childhood, but this male excess is replaced by an emergence of higher rates in females during the transition to adolescence.

Inclusion / Exclusion criteria: These criteria are summarised using the PICOS statement below. For full details of the inclusion and exclusion criteria, see the protocol: supplemental document).

Included studies must have the following characteristics

Population: males and females between the ages of 4 and 17;

Intervention: none;

Comparator: sex and age (at least two age-groups);

Outcome: gender patterning, by age, in measures of physical morbidity;

Study design: longitudinal, cross-sectional and repeat cross-sectional studies (including analysis of study-specific data or routinely collected data).

Methods: The systematic review included methodological components suggested by the PRISMA guidelines (e.g. protocol, literature search, study selection, flow chart, data extraction, critical appraisal and synthesis), and was designed to meet the standards of that guidance. More details are provided in the protocol.

Data sources and search strategy

We searched five electronic databases (Medline, Embase, the Cumulative Index to Nursing and Allied Health Literature (CINAHL), PsycINFO, and the Education Resources Information Center (ERIC)) for studies published in English between 1992

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and the date of search (April 2010). As it was our intention to update a previous review conducted around twenty years previously,[29] we searched for studies published from 1992 to the present. Supplemental document 1 describes the review methods and search strategy in more detail. Following test-searches using pre-identified papers, an information scientist advised on database selection and search terms. As the review's timeframe was limited, the information scientist advised on a search strategy that limited the number of records retrieved by the searches so that they could be processed within the time frame. Prior to the electronic search we manually searched private collections (one of the reviewers has worked in the field of gender and adolescent health for several years and two for approximately two decades); conducted a relatively unstructured internet search and also identified papers that had cited the earlier review.[29] At the end of our study selection process we manually checked the bibliographies of included studies.

We searched each database twice: once using 'generic' health subject headings and keywords and once using 'specific' subject headings and keywords relating to the health conditions we had selected for review (see table 1). In this paper, we refer to these searches as the 'generic search' and the 'specific search'. The precise search strategy differed between databases if different search facilities and search engines made it necessary to adapt our approach.

Table 1: Search History

Database	Generic Search	Specific Search
Medline (Ovid	Date: 16/05/2010	Date: 06/05/2010

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interface)	(child, preschool or child or adolescent).sh. and (sex factors or sex distribution).sh. and (health status or attitude to health or health surveys or mental health[1]).sh. limit to (english language and humans and yr="1992 -Current") (.sh. = MeSH subject headings) Results – 3587	(child, preschool or child or adolescent).sh. and (sex factors or sex distribution).sh. and (asthma or epilepsy or headache disorders, primary or diabetes mellitus).sh. limit to (english language and humans and yr="1992 -Current") (.sh. = MeSH subject headings) Results – 1426
Embase (Ovid interface)	Date: 19/04/2010 (child or school child or adolescent or preschool child).sh. and (sex difference or gender).sh. and (adolescent health or health survey or health status).sh. limit to (human and English language and yr="1992 - Current")	Date: 06/05/2010 (child or school child or adolescent or preschool child).sh. and (sex difference or gender).sh. and (asthma or primary headache or migraine or diabetes mellitus or epilepsy).sh. limit to (human and English language and yr="1992 -

[1] Note: following this initial search, the decision was made to focus the review on physical rather than mental health.

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	(.sh.= subject headings) Results – 2652	Current") (.sh.= subject headings) Results – 1526
CINAHL (Cumulative Index to Nursing and Allied Health) (EBSCO Host interface)	Date: 19/04/2010 (adolescence or child or child, preschool).sh. and (sex factors).sh. and (health status or health status indicators or attitude to health or symptoms or morbidity or child health or adolescent health).sh. limit to (english language and yr="1992 -Current") (.sh.= word in subject heading) Results – 1467	Date: 07/05/2010 (adolescence or child or child, preschool).sh. and (sex factors).sh. and (asthma or diabetes mellitus or headache or epilepsy).sh. limit to (english language and yr="1992 -Current") (.sh.= word in subject heading) Results – 498
PsycINFO (EBSCO Host interface)	Date 19/04/2010 (child* or adolesc* or young pers* or teenage* or pupil* or school child* or minor*).kw. and (human sex differences or sex).sh. and (health or health attitudes or health complaints	Date: 07/05/2010 (child* or adolesc* or young pers* or teenage* or pupil* or school child* or minor*).kw. and (human sex differences or sex).sh. and (asthma or diabetes mellitus or headache or

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	or general health questionnaire or well being or self report or morbidity or symptoms).sh. limit to (english language and yr="1992 -Current") (.kw. – keywords, .sh.= subjects) Results – 1136	epilepsy).sh. limit to (english language and yr="1992 -Current") (.kw. – keywords, .sh.= exact subjects) Results: 38
ERIC (Education from US Department of Education, and Institute of Education Sciences) (Ovid interface)	(19/04/2010) ((children or young children or adolescents or early adolescents or late adolescents).sh. or (pupil* or school child* or minor*).ab.) and ((sex or gender differences).sh. or (sex or gender).ab.) and ((health or child health or adolescent health or well being or wellness).sh. or (morbidity or symptom*).ab.) limit to (english language and yr="1992 -Current") (.sh.= ERIC subject headings, .ab. = abstract)	Date: 07/05/2010 ((children or young children or adolescents or early adolescents or late adolescents).sh. or (pupil* or school child* or minor*).ab.) and ((sex or gender differences).sh. or (sex or gender).ab.) and ((asthma or headache or migraine).ab. or (diabetes or epilepsy).sh.) limit to (english language and yr="1992 -Current") (.sh.= ERIC subject headings, .ab. = abstract) Results: 22

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	Results: 593	

Study Selection

One reviewer (AM) screened all the publications identified by both literature searches to exclude obviously irrelevant titles. The remaining (i.e. not excluded) publications were retrieved and, on reading, AM screened out those that were clearly not eligible for inclusion in the review (see Figure 1, 'First Sift'). Studies of uncertain eligibility were checked by two other reviewers (KH and HS) so that a decision to exclude or retrieve the full paper could be reached (see Figure 1, 'Second Sift'). Some retrieved papers were excluded at the initial reading ('Third Sift'), whilst others were excluded at the data extraction and appraisal stage (based on agreement from all the reviewers). At this final stage we also excluded studies that only explored asthma-related outcomes after finding a review that already applied our research question to this health outcome.

Outcomes

Our main outcomes measures for this analysis were the number and proportion of studies included in the systematic review that were identified from each ELS. We also collected data on (i) the number of studies identified by each ELS at all stages of the reviews' search and selection process; (ii) the types of health outcomes identified by each ELS; and (iii) the number of studies identified by manual searches.

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Comparing the two searches

We produced a series of Venn diagrams for each stage of the review process, showing the number of studies identified only by the specific literature search, the number identified only by the general literature search, and the number identified by both searches (see Figure 1). The purpose was to see if the two searches identified similar or different sets of documents. Studies that were included in the final review were then tabulated in more detail to help us assess whether there was any systematic variation in the types of health outcome identified by the different searches. Comparisons involved the calculation of frequencies and percentages.

Results

Figure 1 shows for each stage of the review the number of studies identified exclusively by either the specific or the generic search, and (in each intersect) the number of studies identified by both searches.

The diagram makes two points apparent. Firstly, there was relatively little duplication between the two searches. For example, out of the 11509 total hits identified from both literature searches, only 413 (3.6%) were duplicates between the two searches. Throughout each stage of the study selection process, duplication between the two searches remained low, so that only three (7.3%) of the 41 studies selected for final inclusion in the review were identified by both search strategies (further details of the 41 included studies are available in a supplemental document).

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Secondly, we note that the specific search led to less than half the number of initial hits, compared to the generic search (3299 vs. 8210, respectively), but both searches identified a similar number of studies included in the final review (17 vs. 21, and 3 duplicates).

Four final inclusion studies were identified from our initial manual search but the generic ELS also identified each of these four studies. Further bibliographic checking revealed that one of the studies identified from both the manual search and the generic ELS could also have been found by checking the bibliographies of included studies identified from the specific search. One study identified from the specific ELS could also have been found by checking the bibliographies of included studies identified from the generic search . This means that the generic ELS in combination with the manual search and bibliography check would have identified 25 of the 41 included studies. The specific ELS in combination with the manual search and bibliography check would have identified 24 of the 41 included studies.

We then examined the 41 studies included in the final review, categorising them by the health outcomes each one investigated (see Table 2). The findings suggest some systematic differences in the health outcomes of studies identified using each of the two search strategies. The specific search tended to be the more successful at identifying studies that focused on a single type of health outcome (i.e. those that related to the search terms). The opposite was found for the generic search strategy, which tended to be more successful at identifying studies with multiple health outcomes.

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Table 2: Studies included in the systematic review (n = 41) by summary health outcomes and by the search strategy used to identify each study.

Outcomes	Generic Search	Specific Search	Both Searches
Abdominal Pain		1	
Back pain	2		
Diabetes	1	6	
Epilepsy		3	
Headache	2	7	1
General physical health / wellbeing	5		
Multiple physical health outcomes*	11		2
Total (for each search)	21	17	3

* A range of health outcomes were included in these studies: usually involving measures of general health and bodily pain. See tables in supplemental document 2.

Most notably, we found that the specific ELS alone (i.e. not the generic ELS or manual search) identified all three included studies of epilepsy and all but one of the seven studies on diabetes. Therefore, failure to run the specific search would have meant that our review would have missed most of the evidence relating to these two outcomes. Within the context of our review's findings, this omission would have been important because, whilst the evidence for the other health outcomes presented in Table 2 tended to support our review's main hypothesis, findings for diabetes and

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epilepsy uniquely suggested a counter-hypothesis. Failing to identify evidence to support the counter-hypothesis would have directly affected our review's conclusions.

The tables in supplemental document 2 describe the studies identified by the different ELS by summarizing information on health outcome, journal, study design, appraisal score and country. Three longitudinal studies and six studies classed as higher scoring following the study appraisal were amongst those identified by the generic ELS (although three of these were also identified using the manual search). Five higher scoring studies (but no longitudinal studies) were amongst those only identified by the specific ELS. Both searches identified evidence from a similar (but not identical) range of European countries but only the generic search identified any North American studies. All the studies identified were published in medical/health journals.

Discussion

We have compared two strategies for conducting an electronic literature search for a systematic review. One strategy used generic health terms, whilst the other used more specific health terms. The purpose was to explore whether literature searches with relatively broad inclusion criteria (in terms of health outcomes) are better served by generic or specific health terms, or whether both are needed.

We found that both specific and generic health terms were necessary. They each uniquely identified some of the review's more robust studies. They also identified different types of health outcome. Failure to identify some of those outcomes would have directly affected our review's conclusions. Had we only used generic health terms in our search we would have missed around half the studies that we finally

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included in the review. Likewise, focusing exclusively on specific health terms in the literature search would have failed to identify around half the included papers. A small proportion of these studies would have been identified by our manual search and bibliography check but failing to conduct either of the ELS approaches would still have led to a serious 'loss' of data (or, more correctly, a failure to find data) that would have compromised the integrity and accuracy of our review's findings.

We found that the specific search tended to miss studies with general or multiple health outcomes, whilst the generic search tended to miss studies with single, specific health outcomes. This may appear intuitive, but we contend that the finding is actually surprising. It suggests, for example, that studies that look specifically at young people's diabetes, epilepsy and headache tend not to be identifiable by search terms such as "health status", "health surveys", "child health", "adolescent health", "health status indicators", "symptoms", "morbidity", "health complaints", etc. It also suggests that some studies that, for example, included headache as one of a number of different health outcomes may be identified by a search strategy that includes generic health terms, but could be missed by an ELS that specifically focuses on the term 'headache.'

This finding is at odds with what some authors of this paper initially expected. Prior to our exploring this issue, the authors assumed that the generic health search would identify the vast majority of included studies whilst the specific search would mainly identify a subset of those studies. If other systematic reviewers also make this assumption, then their reviews are at risk of being based on poor quality (highly insensitive) searches.

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Strengths and limitations

We have conducted a prospective comparative study of two electronic literature search strategies that have been field tested whilst we conducted a systematic review. This kind of study is uncommon and hence novel, whilst the prospective and comparative design is a key strength.

The main limitations of this study are that it is based on a single review and the search was not sensitive (i.e. lacking in the use of truncation, synonyms and related terms). The review that we based the study on does not focus on the effectiveness of an intervention, which means that precision cannot be easily increased by including simple study design search terms, and the outcomes are also very complex, which probably increases the difficulty of sensitive and specific searching. These may be regarded as unusual features affecting the generalisablity of our findings but we have argued in our introduction that 'unusual' (i.e. not clinical intervention) reviews are becoming more common and hence are an emerging priority in terms of review methods. The same may be said about time-limited reviews. Ours took eighteen months to complete – not an unusual timeframe in our experience - but we are aware that some systematic reviews (e.g. many Cochrane and Campbell reviews) take longer and involve more comprehensive searches.

It may also be hypothesised that conducting a more extensive ELS and manual search could have led to a greater number of, and possibly more overlap between, studies identified by each component of our search strategy. Ways to achieve a more extensive search could have included using more electronic databases and other

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relevant data sources; identifying a wider number of synonyms for both the health outcomes and other concepts included in the review; using both subject headings and words in the title and abstract to search for every concept in the search strategy; and minimising reliance on the accuracy of database indexers. There is also some existing evidence that the effectiveness of different search strategies may vary depending on the subject of the review.[5] Therefore, it is worth testing our findings in the context of other reviews and different types of literature search, including more sensitive searches. Missing out health outcomes altogether is an alternative means of increasing search sensitivity but we note that our initial search identified well in excess of 10,000 hits. Given the broad review question, attempts to vastly expand the search risked increasing the number of hits to unmanageable levels.

Implications and conclusions

Literature searching has a vital role to play in evidence-informed policy and practice, and it is plausible to theorise a direct pathway by which a poor search may lead to harmful decisions. Conducting research that may assist information scientists and reviewers to improve their search strategies should therefore be a priority. Such research can be nested within the processes of conducting systematic reviews: from our own experience this requires minimal additional resources to the cost of the overall review and can therefore be considered an inexpensive way of conducting useful research in an important field. We therefore hope that other reviewers will make use of similar opportunities to explore how best to optimise electronic searching.

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In light of our findings, we recommend that future systematic reviews of topics that involve multiple health outcomes include both generic and specific health terms in their literature search (if a health outcome search is considered necessary), along with manual searching. Choosing only one of these search components could, based on our findings, increase the risk of reviewers missing robust evidence and making misleading conclusions.

Authors' interests

(1) All authors have completed the Unified Competing Interest form at www.icmje.org/coi_disclosure.pdf (available on request from the corresponding author) and declare that ME, AM, HS, KH have support from the Medical Research Council and Chief Scientist Office for the submitted work (5TK50; 5TK40); (2) ME, AM, HS, KH have no relationships with any company that might have an interest in the submitted work in the previous 3 years; (3) their spouses, partners, or children have no financial relationships that may be relevant to the submitted work; and (4) ME, AM, HS, KH have no non-financial interests that may be relevant to the submitted work.

Ethics

As this study focused on searching for literature that was already in the public domain, it involved no patients, consents, nor other issues that required formal approval from an ethics committee.

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Data sharing

The review protocol (including search strategies) and a list of studies included in the final review are available in the supplemental documents submitted with this article. Further data related to the searches are available from the corresponding author at matt@sphsu.mrc.ac.uk.

Details of contributors

ME helped plan and conduct the study, analyse the findings, led on writing the manuscript and is guarantor for the study. AM, HS and KH helped plan the study and conduct the study, analyse the findings and provide content and comments on the manuscript. All authors, external and internal, had full access to all of the data (including statistical reports and tables) in the study and can take responsibility for the integrity of the data and the accuracy of the data analysis.

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What is already known on this subject

Literature searching has a vital role to play in systematic reviews that inform policy and practice. Evidence to help reviewers conduct effective literature searches tends to be based on reviews of randomised controlled trials. There is relatively little evidence to help guide literature searches for other types of systematic review.

What this study adds

Whilst conducting a systematic review that included a range of health outcomes, we compared two electronic literature search strategies – one that used generic terms for (ill)health and another than used terms for specific illnesses. Our findings suggest that systematic review searches that use *only* generic *or* specific search terms (rather than a combination of the two) for health outcomes risk missing a large proportion of the relevant evidence, potentially leading to erroneous conclusions that may, in turn, misinform policy and practice.

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References

1. Higgins JPT, Green S. *Cochrane Handbook for Systematic Reviews of Interventions Version 5.0.2 [updated September 2009].* : The Cochrane Collaboration, 2009.
2. NHS Centre for Reviews and Dissemination. *Systematic Reviews: CRD's Guidance for Undertaking Reviews in Healthcare* York: CRD University of York, 2009.
3. Petticrew M, Roberts H. *Systematic Reviews in the Social Sciences: A practical guide.* Oxford: Blackwell Publishing, 2006.
4. Ogilvie D, Hamilton V, Egan M, et al. Systematic reviews of health effects of social interventions: 1. Finding the evidence: how far should you go? *J Epidemiol Community Health* 2005;59:804-08.
5. Bambra C. Real world reviews: a beginner's guide to undertaking systematic reviews of public health policy interventions. *J Epidemiol Community Health* 2011;65:14-19.
6. Glanville JM, Lefebvre C, Miles JN, et al. How to identify randomized controlled trials in MEDLINE. *J Med Libr Assoc* 2006;130-136:130-36.
7. Golder S, McIntosh HM, Duffy S, et al Centre for Reviews and Dissemination and UK Cochrane Centre Search Filters Design Group. Developing efficient search strategies to identify reports of adverse effects in MEDLINE and EMBASE. *Health Info Libr J* 2006;23:3-12.
8. Montori VM, Wilczynski NL, Morgan D, et al. Optimal search strategies for retrieving systematic reviews from Medline: analytical survey. *BMJ* 2005;330:68.
9. Wong SS, Wilczynski NL, Haynes RB. Developing optimal search strategies for detecting clinically sound treatment studies in EMBASE. *J Med Libr Assoc* 2006;94:41-47.
10. Dickersin K, Scherer R, Lefebvre C. Identifying relevant studies for systematic reviews. *BMJ* 1994;309:1286-91.
11. Eisinga A, Siegfried N, Clarke M. The sensitivity and precision of search terms in Phases I, II and III of the Cochrane Highly Sensitive Search Strategy for identifying reports of randomized trials in MEDLINE in a specific area of health care - HIV/AIDS prevention and treatment interventions. *Health Info Libr J* 2007; 24:103-09.
12. Royle P MR. Literature searching for randomized controlled trials used in Cochrane reviews: rapid versus exhaustive searches. *Int J Technol Assess Health Care* 2003;19:591-603.
13. Joyce K, Pabayo R, Critchley JA, et al. Flexible working conditions and their effects on employee health and wellbeing. *Cochrane Database Syst Rev*, 2010.
14. Porta D, Milani S, Lazzarino A, et al. Systematic review of epidemiological studies on health effects associated with management of solid waste. *Environ Health* 2009;8(60):doi:10.1186/476-069X-8-60.
15. Uscher-Pines L. Health effects of relocation following disaster: a systematic review of the literature. *Disasters* 2009;33(1):1-22.
16. D'Onise K, Lynch J, Sawyer M, et al. Can preschool improve child health outcomes? A systematic review. *Soc Sci Med* 2010;70(9):1423-40.
17. Egan M, Petticrew M, Ogilvie D, et al. New Roads and Human Health: A Systematic Review. *Am J Public Health* 2003;93(9):1463–71.
18. Dangour A, Aikenhead A, Hayter A, et al. Comparison of putative health effects of organically and conventionally produced foodstuffs: a systematic review:

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Report for the Food Standards Agency. London: Nutrition and Public Health Intervention Research Unit London School of Hygiene & Tropical Medicine, 2009.
19. Vickers AJ. Can acupuncture have specific effects on health? A systematic review of acupuncture antiemesis trials. *J R Soc Med* 1996;89:303-11.
20. Daykin N, Orme J, Evans D, et al. The Impact of Participation in Performing Arts on Adolescent Health and Behaviour: A Systematic Review of the Literature. *J Health Psychol* 2008;13:251-64.
21. Rogers G, Elston J, Garside R, et al. The harmful health effects of recreational ecstasy: a systematic review of observational evidence. *Health Technol Assess* 2009;13(6):DOI: 10.3310/hta13060.
22. Passchier W, Knottnerus A, Albering H, et al. Public health impact of large airports. *Rev Environ Health. 2000 Jan-Jun;15(1-2):* 2000;15(1-2):83-96.
23. Egan M, Petticrew M, Ogilvie D, et al. "Profits before people"? A systematic review of the health and safety impacts of privatising public utilities and industries in developed countries. *J Epidemiol Community Health* 2007;61:862-70.
24. McCartney G, Thomas S, Thomson H, et al. The health and socioeconomic impacts of major multi-sport events: systematic review (1978-2008). *BMJ* 2010;340:c2369.
25. Paradies Y. A systematic review of empirical research on self-reported racism and health. *Int J Epidemiol* 2006;35(4):888-901.
26. Vartanian LR, Schwartz MB, Brownell KD. Effects of soft drink consumption on nutrition and health: a systematic review and meta-analysis. *Am J Public Health* 2007;97(4):(4):667-75.
27. Bambra C, Whitehead M, Sowden A, et al. "A hard day's night?" The effects of Compressed Working Week interventions on the health and work-life balance of shift workers: a systematic review. *J Epidemiol Community Health* 2008;62(9):764-77.
28. Greenhalgh T, Peacock R. Effectiveness and efficiency of search methods in systematic reviews of complex evidence: audit of primary sources. *BMJ* 2005;331:1064-65.
29. Sweeting H. Reversals of fortune? Sex differences in health in childhood and adolescence. *Soc Sci Med* 1995;40(1):77-90.

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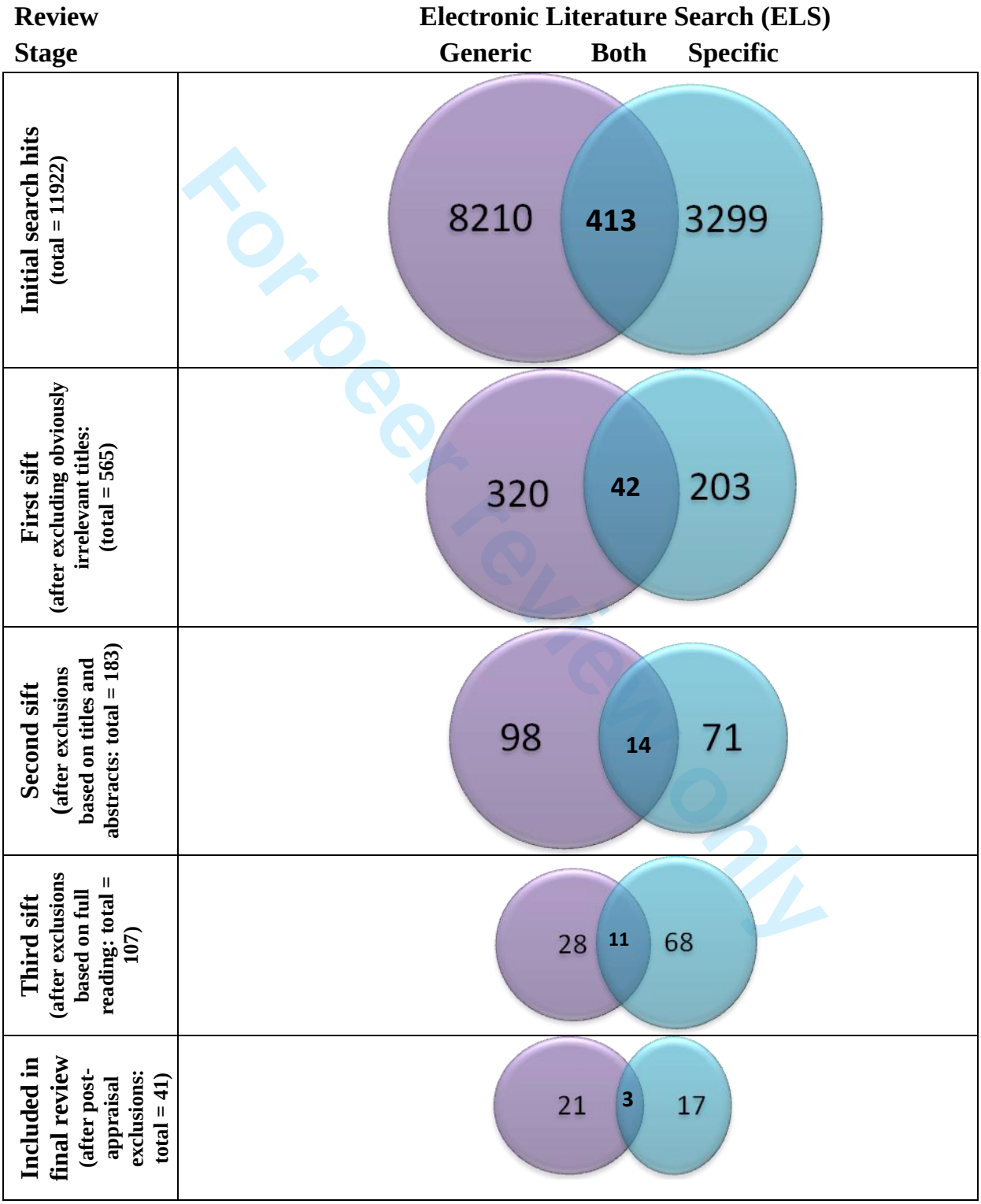
Figure 1. Review study selection flow chart: studies identified by the 'generic' search only (purple circle); 'specific' search only (light blue circle); and by both searches (dark blue intersect).



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Note: from the above figures the following can be calculated.

Generic ELS: sensitivity = 58.5%; precision = 0.3%; number needed to read = 112 (1.3%).

Specific ELS: sensitivity = 48.8%; precision = 0.5%; number needed to read = 85 (2.3%).

An initial manual search identified ten articles for full reading, of which 4 were included in the final review. These studies were also identified from the generic ELS, and are included as such in the figure above.

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STROBE Statement—checklist of items that should be included in reports of observational studies, followed by a table showing how the current study conforms to the STROBE statement.

	Item No	Recommendation
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found
Introduction		
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported
Objectives	3	State specific objectives, including any prespecified hypotheses
Methods		
Study design	4	Present key elements of study design early in the paper
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection
Participants	6	(*a*) *Cohort study*—Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up *Case-control study*—Give the eligibility criteria, and the sources and methods of case ascertainment and control selection. Give the rationale for the choice of cases and controls *Cross-sectional study*—Give the eligibility criteria, and the sources and methods of selection of participants
		(*b*) *Cohort study*—For matched studies, give matching criteria and number of exposed and unexposed *Case-control study*—For matched studies, give matching criteria and the number of controls per case
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group
Bias	9	Describe any efforts to address potential sources of bias
Study size	10	Explain how the study size was arrived at
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding
		(*b*) Describe any methods used to examine subgroups and interactions
		(*c*) Explain how missing data were addressed
		(*d*) *Cohort study*—If applicable, explain how loss to follow-up was addressed *Case-control study*—If applicable, explain how matching of cases and controls was addressed *Cross-sectional study*—If applicable, describe analytical methods taking account of sampling strategy
		(*e*) Describe any sensitivity analyses

Continued on next page

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Results		
Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed
		(b) Give reasons for non-participation at each stage
		(c) Consider use of a flow diagram
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders
		(b) Indicate number of participants with missing data for each variable of interest
		(c) *Cohort study*—Summarise follow-up time (eg, average and total amount)
Outcome data	15*	*Cohort study*—Report numbers of outcome events or summary measures over time
		Case-control study—Report numbers in each exposure category, or summary measures of exposure
		Cross-sectional study—Report numbers of outcome events or summary measures
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included
		(*b*) Report category boundaries when continuous variables were categorized
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses
Discussion		
Key results	18	Summarise key results with reference to study objectives
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence
Generalisability	21	Discuss the generalisability (external validity) of the study results
Other information		
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.

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STROBE Checklist: How to avoid missing half the evidence: comparing the use of generic and specific electronic search terms used to identify health outcomes for a systematic review.

STROBE Item No.	Authors comment
1 (a)	Done – see abstract, main article
1 (b)	Done – see abstract, main article
2	Done – main article
3	Done – main article
4	Done – main article
5	Done – a literature search is internet based rather than set in a specific location so we gave details of the databases searched and the review that the searches were conducted for. Main article
6 (a)	Done – main article
6 (b)	Not applicable – the study includes no matching of the kind described in STROBE.
7	Done – main article
8	Done – main article
9	Done – use of multiple reviewers during search and selection process.
10	Not applicable - the study did not require a power calculation as it includes no participants (in the conventional use of the terms). The text does state that the two searches were field tested during an actual systematic review – main article and protocol.
11	Done – main article.
12 a to e	Done – main article.
13	Done – figure 1 and text in main article.
14 a	Done – table 1, main article.
14 b and c	Not applicable
15	Not applicable
16	Done in so far as applicable (the study does not involve estimates, statistical adjustment or missing data as described by STROBE). Main article.
17	Done – main article.
18	Done – main article.
19	Done – main article.
20	Done - main article.
21	Done - main article.
22	Done - main article.

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