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Systematic Reviews of Surgical Procedures in Children: Quantity, Coverage and
Quality

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Abstract

Aim: Systematic reviews have the potential to map those areas where children are under-represented in surgical research. We aimed to describe and evaluate the quantity, coverage, and the quality of conduct and reporting of systematic reviews of surgical procedures in children.

Methods: We searched four biomedical databases, a systematic review register, reference lists and conducted hand searching to identify relevant reviews. Two reviewers worked independently to critically appraise included studies and abstract data. We assessed reporting quality using the PRISMA statement and methodological quality using the AMSTAR tool.

Results: Fifteen systematic reviews were identified, representing 0.01% of all paediatric surgical citations in MEDLINE and Embase. Thirteen of the reviews were Cochrane reviews, and most reviews (12/15) addressed subspecialty interests such as otorhinolaryngology. The median number of included trials per systematic review was four (interquartile range 1 to 9.5), the median number of primary outcomes was 5.5 (interquartile range 3.5 to 7.5). In general, reporting and methodological quality was good although there were several omissions, particularly around completeness of reporting of statistical methods used, and utilisation of quality assessments in analyses. Outcomes were often not clearly defined and descriptions of procedures lacked sufficient detail to determine the similarities and differences amongst surgical procedures within the contributing trials.

Conclusion: Systematic reviews of surgical procedures in children are rarely published. To improve the evidence-base and guide research agendas, more systematic reviews should be conducted, using standard guidelines for conduct and reporting.

Key Words: AMSTAR, Bias, Outcomes, PRISMA, Quality, Systematic review

Introduction

An estimated 11 percent of the global disease burden can be treated with surgery.¹ Children represent an important part of this population because they differ both anatomically and physiologically from adults and because untreated disease has the potential to cause long-term disability.² To ensure children receive best possible care, it is important that surgical procedures are based on appropriate evidence which, for most procedures should come from randomised controlled trials (RCTs) and systematic reviews of RCTs. However, we know that few RCTs of surgical procedures in children are published.^{3,4}

Despite this, unless all pre-existing evidence is systematically located and synthesised, gaps in the evidence base are not easily highlighted. Without systematic reviews, it becomes difficult to generate research agendas, attract research funding, ethically justify the need for new research, design and interpret studies in the light of the totality of the available evidence or publish in some journals.^{5,6} A lack of trials in an area does not preclude the conduct of a systematic review; it merely highlights, in a scientific manner, the research-practice gap. Therefore, while systematic reviews undoubtedly represent a significant investment of time and energy and while their results are dependent on the quantity and quality of the primary studies included, they need to be done.

Currently, little is known about the quantity or quality of systematic reviews of surgical procedures in children. To better understand what systematic reviews of the evidence are taking place, we aimed to describe and evaluate the quantity, coverage, and quality of conduct and reporting of systematic reviews of surgical procedures in children.

Materials and methods

Identification and Selection of Studies

A systematic search of the literature was conducted to identify all published systematic reviews of randomised controlled trials of surgical procedures in children. We searched the Cochrane Database of Systematic Reviews (Issue 3, 2011), MEDLINE via OvidSP (1950-15/03/2011), Embase via OvidSP (1980-15/03/2011) and Database of Abstracts of Reviews of Effectiveness via OvidSP (Issue 1, 2011) for reviews. See the **supplementary file** for the search terms used. We also searched the reference lists of retrieved systematic reviews, contacted content experts, searched the register of systematic reviews PROSPERO (www.crd.york.ac.uk/prospero/, accessed on the 18/03/11) and performed hand searches of selected journals (Journal of Pediatric Surgery, Pediatric Surgery International, European Journal of Pediatric Surgery and Seminars in Pediatric Surgery) from 2000-2010, to identify additional relevant systematic reviews.

We defined a systematic review as an attempt to collate all empirical evidence using pre-specified eligibility criteria, to answer a specific research question, using systematic methods that were selected with a view to minimising bias.⁷ We excluded editorials, narrative reviews and reviews following case series as these did not fit our definition of a systematic review.

We also excluded reviews that included animal studies and reviews that included non-randomised studies. We only included systematic reviews, where the primary procedure under review was surgical, that is not diagnostic, prognostic or aetiological and at least half of the included studies in the review involved children. We defined 'surgery' as the application of a manual or instrumental technique in the treatment of disease, injury or deformity. We defined a child as an age less than 18. As the scope of practice of paediatric surgeons varies internationally, we took a pragmatic approach and selected systematic

reviews of surgical procedures in children rather than select reviews based on a specialties typical caseload. We limited our search to studies published up to the end of 2010, as 2010 was the last complete year of publication. There were no restrictions on inclusion by language, publication status or review topic. RGM and TER worked independently to critically appraise included studies and abstract data with consensus agreement.

Data Extraction

We collected data on a broad range of descriptive, epidemiological and reporting characteristics from eligible reviews using a 97-item data collection form. Procedures were judged as adequately described if they provided information about who conducted the procedure, where the procedure was conducted and any steps that were crucial to the success of the procedure or differed amongst trials. Outcomes were judged as adequately described if it was clear how, when and for whom outcomes were assessed. The primary outcome(s) were those defined by the review authors.

Reporting Quality

The reporting quality of the included systematic reviews was assessed using the PRISMA statement. PRISMA is an internationally recognised reporting guideline that consists of 27-items and a flow diagram to guide optimal reporting in systematic reviews and meta-analyses.⁷ The quality in each PRISMA domain was graded as high, low or unclear for each review based on the degree to which it followed the criteria in the PRISMA statement.⁷

Methodological Quality

The methodological quality of included systematic reviews was assessed using the AMSTAR tool. AMSTAR stands for Assessment of Multiple SysTemAtic Reviews and is composed of

an 11-item checklist.⁸⁻¹⁰ The quality in each AMSTAR domain was graded as high, low or unclear for each review according to the criteria in the AMSTAR tool publication.⁹

Statistical Analysis

To estimate the proportion of all citations that were systematic reviews we divided the number of reviews obtained in MEDLINE and Embase by the total number of citations retrieved in MEDLINE and Embase on the 15/03/2011. To give a relative indication of the frequency with which systematic reviews of children undergoing surgical procedures are conducted in comparison to systematic reviews in other disciplines we divided the number of systematic reviews of children undergoing surgical procedures obtained from The Cochrane Library by the total number of systematic reviews available from The Cochrane Library on the 15/03/2011. 95% confidence intervals (95% CI) were calculated for all point estimates using Stata software (Stata11, StataCorp LP, Texas, USA).

Results

Included Reviews

Our search identified 24 633 unique records, see **Figure 1**. Initial screening excluded 24 490 records. The remaining 143 full-text articles were retrieved, of which 128 were ineligible. We therefore included 15 systematic reviews in our review,¹¹⁻²⁵ which represented 0.01% (15/110 885, 95% CI 0.01% to 0.02%) of all citations retrieved from MEDLINE and Embase.

Thirteen of the 15 reviews we retrieved were Cochrane reviews, while one was published in *Pediatric Surgery International*,¹⁶ and one in *Archives of Disease and Childhood*.¹⁷

On the 15/03/2011, there were 4 574 reviews in The Cochrane Library. Therefore, systematic reviews of children undergoing surgical procedures represented 0.3% (13/4574, 95% CI 0.2% to 0.5%) of all systematic reviews available in The Cochrane Library.

Characteristics of Included Reviews

The characteristics of the included systematic reviews are outlined in **Table 1**. The majority of the reviews were published in 2010 (5/15) and the earliest review was published in 2001. On average, there were four authors per review, from four different institutions and two different countries. The median number of included trials per systematic review was four (interquartile range 1 to 9.5), while three reviews did not identify any relevant trials. The median number of citations screened per review was 60 (interquartile range 15 to 226). Eight of the 15 first authors were male. The reviews covered a narrow range of topics; the most commonly assessed area was the use of surgical treatments for the management of otitis media (four reviews).

The most commonly assessed procedures were surgical techniques such as tonsillectomy, adenoidectomy and wound closure strategies, which were assessed in 73% (11/15) of the

reviews. The remaining 27% (4/15) of reviews assessed the efficacy of devices such as grommets or pressure equalising tubes and sutures. Of those reviews, which assessed surgical techniques, all lacked sufficient detail to determine the similarities and differences between the surgical procedures used in different trials. For example, the reviews lacked descriptions of who performed the procedure (consultant, registrar, years of experience), where the procedure was performed (outpatients, day-care surgery or elsewhere) and any differences between the performance of the procedure across trials.

A small number of outcomes were measured per review, with a median of one primary outcome and 5.5 outcomes in total assessed (interquartile range: 1 to 3, 3.5 to 7.5 respectively). The outcomes assessed among reviews were diverse. The most commonly assessed outcomes were mortality, adverse events and quality of life, which were each assessed in five reviews. A definition was provided for these outcomes in only three reviews.

Reporting Quality

Figure 2 illustrates the results of reporting quality as assessed by the PRISMA statement. Most items were well reported across reviews. However, analytical and statistical results (items 19-23) tended to be less well reported than other components. In addition, reviews only infrequently included the terms ‘systematic review’ or ‘meta-analysis’ in their titles (although it should be noted that Cochrane reviews never use these terms in their titles as they are implied, given they are collated in a database called “The Cochrane Database of Systematic Reviews”), most reviews did not adequately specify that they checked risk of bias across studies and approximately one-third of included reviews did not declare their funding sources. No review utilised the PRISMA flow diagram to illustrate the results of their literature search and the process of screening and selecting studies for inclusion however, one review utilised the QUOROM (PRISMA’s predecessor) flow diagram.

Methodological Quality

Figure 3 illustrates the results of the methodological quality assessment using the AMSTAR tool. While most methodological items were well conducted, several were conducted inadequately. In particular, the scientific quality of included studies was often not used appropriately in formulating conclusions, publication bias was frequently not assessed and while conflicts of interest were often presented by the authors of the systematic review, they were not presented for the authors of trials included in the systematic reviews.

Discussion

Despite the importance of systematic reviews in informing healthcare policy, practice, and research agendas, most areas are not informed by any systematic reviews of surgical procedures. In total, there were 15 systematic reviews available, which at the time of our search represented only 0.01% of the entire child related surgical literature. Most of the reviews represented subspecialty interests, which may reflect our pragmatic approach to selecting studies. This also reflects the dichotomy of the way published literature is organised compared to the pragmatic reality of surgical practice internationally. For example, four reviews evaluated surgical treatments of otitis media, which limits their usefulness to the majority of general paediatric surgeons who would not routinely perform these procedures. Reporting and methodological quality, as assessed by PRISMA and AMSTAR respectively, was generally adequate although there were several omissions, particularly around completeness of reporting of statistical methods used, and use of quality assessments in analyses. Descriptions of procedures often lacked specific details, which would enhance their reproduction for example highlighting similarities and differences between the surgical procedures used in the trials included in the review. Reported outcomes were diverse and loosely defined. Less than a third of reviews assessed outcomes important to patients and clinicians such as adverse events.

Relative to other surgical and paediatric specialties, the quantity of systematic reviews of children undergoing surgical procedures remains low. In 2007, 2,500 systematic reviews were being indexed annually in MEDLINE; of which one-fifth were Cochrane reviews.²⁶ In Neonatology, there were 61 Cochrane reviews available in 2009,²⁷ and within Orthopedic surgery, 40 reviews were located in 2001.²⁸ We were only able to locate 15 systematic reviews of children undergoing surgical procedures in total, of which 13 were Cochrane

reviews. Compared to other Cochrane reviews, systematic reviews of children undergoing surgical procedures included a similar number of studies and outcomes per review.²⁹

One solution to improve the evidence base for children undergoing surgery is to require a systematic review to be conducted before any research project is started.⁵ Undoubtedly this would require a significant investment of time and energy, but it would improve the evidence base whilst also providing guidance on the design of the proposed study and allow results to be interpreted in the light of the totality of the available evidence.⁵ While such an initiative may produce a large number of empty reviews, this will highlight the necessity of the proposed study and help in the development of strategic research agendas.

Our assessment of reviews against current standards for methodological and reporting quality did not identify substantial problems but these standards fail to address important issues around the reporting of procedures and outcomes. Firstly, the similarities and differences between surgical procedures used in different trials should be discussed, so that important steps and acceptable variations can be identified. Unlike pharmacological interventions, which are usually standardised, the implementation of surgical procedures depends in part on their reproducibility.³⁰ Our cohort of reviews did not provide enough practical details about the variations in surgical procedures across trials. Systematic reviews are not designed to, nor should they provide step-by-step instructions for completing a surgical procedure, but they should include information on those factors that are likely to affect a procedure's efficacy. To aid authors and readers in this process it has been suggested that procedures be classified using standardised taxonomies and collated into compendiums of procedures similar to pharmacopoeia.^{31,32} Secondly, sets of core outcomes should be developed which will help authors with the conduct of reviews and ensure that important effects are not overlooked. Recently, the COMET (Core Outcome MEasures in Trials) initiative has highlighted the importance of reporting core outcomes in trials.³³ The principle behind this project remains

the same for systematic reviews; namely, the use of core outcomes reduces reporting bias, misinformation and wasted resources.³²⁻³⁴ The development and endorsement of core outcome sets and procedure taxonomies will depend on the support and encouragement from relevant Colleges and Societies.

As a descriptive study, there are potential limitations to our work. We did not design our study to identify the causes and mechanisms behind our results and so can only speculate on the reasons for our findings. We have attempted to assess the totality of systematic reviews of surgical procedures in children. However, bibliographic databases are not comprehensive and so we may have overlooked reviews. We attempted to overcome this limitation by searching a range of databases, as well as searching reference lists, contacting content experts, hand searching selected journals and searching a systematic review register. Nevertheless, given the small number of included reviews, the data must be interpreted with caution, as our results may be underpowered to detect even large differences in quality estimates. In addition, while we have assessed the quality of systematic reviews, we have not assessed the quality of the trials included in those reviews. Finally, this review intentionally focuses on systematic reviews of RCTs. The quantity and quality of alternative systematic reviews for example systematic reviews of diagnostic test accuracy studies remain unknown.

Systematic reviews are important for a wide variety of reasons including the practice of evidence-based surgery, for guideline developers and for those constructing research agendas. Our findings suggest that this type of research is not being conducted as frequently as it could be and set a benchmark for progress. Although the reporting and methodological quality of reviews in general was adequate, there remains room for improvement. The development of core outcome sets and procedure taxonomies will assist authors in the conduct of reviews and ensure that effects are less likely to be missed and are more replicable. These findings should

challenge researchers and funding bodies to improve the evidence base for children undergoing surgery by promoting the conduct of high quality systematic reviews.

What is already known on this topic

1. Systematic reviews collate and summarise all the available evidence on a topic.
2. Systematic reviews are important for a wide variety of reasons including the practice of evidence-based surgery, for guideline developers and for those prioritising research agendas.

What this paper adds

1. To our knowledge, this is the first study of systematic reviews of surgical procedures in children.
2. It shows that few systematic reviews are being published.
3. This has implications for surgical practice involving children and those setting research agendas.

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Figures legends

Figure 1. Process for the selection of systematic reviews of surgical procedures in children.

Figure 2. Quality of reporting of systematic reviews of surgical procedures in children, using the PRISMA checklist (<http://www.prisma-statement.org/statement.htm>).⁷

*ROB: Risk of bias

Note: The area of the star plot represents the proportion of studies that adequately reported each PRISMA item.

Figure 3. Quality of conduct of systematic reviews of surgical procedures in children, using the AMSTAR tool (<http://www.biomedcentral.com/1471-2288/7/10/table/T2>).⁹

Note: The area of the star plot is proportional to the percentage of reviews, which adequately conducted each AMSTAR item

Table 1: Characteristics of included studies

Category	N=16 (%)
Journal	
Cochrane Database of Systematic reviews	13 (86)
Pediatric Surgery International	1 (7)
Archives of Disease and Childhood	1 (7)
Year of publication	
2010	5 (33)
2009	2 (13)
2008	3 (20)
2007	1 (7)
Pre 2007	4 (27)
Number of authors	
2	3 (20)
3	3 (20)
>3	9 (60)
Gender of first author	
Male	8 (53)
Female	7 (47)
Topic area	
Oncology	2 (13)
Orthopedics	1 (7)
Otorhinolaryngology	8 (53)
Other	4 (27)
Funding source	
Investigator	10 (67)
Not stated	5 (33)