



Review Articles

Quality assessment of economic evaluation studies in pediatric surgery: A systematic review



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ABSTRACT

Purpose: To assess economic evaluation studies (EES) in pediatric surgery and to identify potential factors associated with high-quality studies.

Methods: A systematic review of the literature using PubMed and Cochrane databases was conducted to identify EES in pediatric surgery published between 1 June 1993 and 30 June 2013. Assessment criteria are derived from the Drummond checklist. A high quality study was defined as a Drummond score ≥ 7 . Logistic regression analysis was used to determine factors associated with high quality studies.

Results: 119 studies were included. 43.7% ($n = 52$) of studies were full EES. Cost-effectiveness analysis was the most frequent (61.5%) type of full EES. Only 31.6% of studies had a Drummond score ≥ 7 and 73% of these were full EES. The factors associated with high quality were identification of costs (OR: 14.08; 95% CI: 3.38–100; $p < 0.001$), estimation of utility value (OR: 8.13; 95% CI: 2.02–43.47; $p = 0.005$) and study funding (OR: 3.50; 95% CI: 1.27–10.10; $p = 0.02$).

Conclusion: This review shows that the number and the quality of EES are low despite the increasing number of studies published in recent years. In the current context of budget constraints, our results should encourage pediatric surgeons to focus more on EES.

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Increasing healthcare expenditure has led to the development of health policies based on the optimization of resources in many countries. This optimization involves the use of medico-economic evaluation. The main objectives of economic evaluation studies (EES) [1] are to compare health programs or health strategies on a monetary basis. A number of countries require that EES be performed systematically through national agencies for any new treatment or medical device. This is the case for example in the United Kingdom, with the National Institute for Health and Care Excellence (NICE) and in Canada, with the Canadian Agency for Drugs and Technologies in Health (CADTH).

Taking economic parameters into account in the assessment of increasingly expensive diagnostic or therapeutic strategies has emerged as a critical issue in overall health policies. Over the last few years, there has been a steady increase in the number of EES reported in the

literature. These studies use a variety of methods, which do not always comply with the consensus on standard methodology [1,2].

Early reviews of the overall quality of EES in the management of adult or pediatric hematological diseases showed that the quality of these studies was generally quite low [3–6]. We sought to investigate whether the same is true for EES of surgical management of diseases in children, and whether EES in this area met international quality standards.

To answer these questions, we conducted a systematic review of the literature over the past 20 years to identify and assess the quality of EES relating to surgery in children. We also identified potential factors associated with high quality EES.

1. Methods

1.1. Study design and selection criteria

A systematic review of the literature was performed to identify EES in the field of surgical management of diseases in children. The eligibility criteria were:

Economic evaluation study;

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Surgical management of diseases in children (patients aged <18 years old);
All surgical specialties.

1.2. Search strategy

We performed a search in Medline through PubMed and the Cochrane Library, with search limits set from 1 June 1993 to 30 June 2013, and languages limited to English and French. The following search words (or combinations thereof) were used: “economic evaluation” or “cost analysis” or “cost-benefit” or “cost-effectiveness” or “cost-minimization analysis” and “child” or “children” and “surgery”. Two reviewers screened the titles and abstracts of all reports identified by the search strategy. We followed the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines for this systematic review [7]. Articles were selected as follows: the first step was to merge the references from different sources into a single database and to eliminate duplicates. In a second step, titles and abstracts were screened and analyzed to determine whether the article was on the topic of pediatric surgery and economic analysis. In the third step, the full text of selected articles was retrieved, read and analyzed. Additionally, reference lists of selected studies were screened to identify potentially overlooked studies.

1.3. Data collection and extraction

Data extraction and quality assessment were undertaken by one reviewer (A. F. K.) and checked by a second reviewer (V. N.) The following data were extracted: authors' names, publication year, country of origin, journal, corresponding author, specialty and academic affiliation, type of economic evaluation, study objectives, perspective, sources of cost and effectiveness data, source of utility values, outcome measure for effectiveness (disability-adjusted life years, quality-adjusted life years, life years gained), discount rate, and results. The perspective is the point of view from which the study is conducted. For example, when the societal point of view was taken, direct healthcare costs and indirect costs or out-of-pocket expenses for the patient and family were included in economic evaluations. Utility represents the cardinal values that reflect an individual's preferences for different health outcomes.

In accordance with Drummond et al. [1], the classification of EES is usually based on the ability of studies to answer the following two questions: First, is there a comparison of two or more strategies? Second, are both costs (inputs) and consequences (outputs) of alternatives strategies examined? (Table 1).

If both the costs and the consequences of two or more therapeutic strategies or programs are not compared, the studies are considered as partial EES. These studies include cost analysis (CA), cost description (CD), and cost–outcome description (COD). The studies were classified as a CA if only the costs of two or more competing treatments were compared, a CD if only the costs were assessed, an OD if only outcomes were examined for a single program and a COD if cost and outcome were determined for a single program.

By contrast, studies were considered as full EES if both the costs and consequences of two or more strategies were compared. These studies include cost-effectiveness analysis (CEA), cost-minimization analysis (CMA), cost-utility analysis (CUA) and cost-benefit analysis (CBA).

The results of CEA are expressed in terms of monetary unit per outcome, i.e., disease averted, life years saved. CMA is a particular form of CEA whose aim is to identify the least costly strategy by considering that the strategies are of equivalent effectiveness. The results of CUA are expressed in terms of monetary unit per quality-adjusted life-years (QALYs) or disability-adjusted life-years (DALYs). In CBA, the cost and benefit are expressed in monetary units.

All monetary units were discounted and converted to 2013 United States dollars (USD) using the Consumer Price Index monetary units (http://www.bls.gov/data/inflation_calculator.htm). When the article did not state the monetary unit's original year, the year before the year of publication was assumed. If a range of dates was presented, the midpoint year was chosen as the reference year.

For CEA and CUA studies, the incremental cost effectiveness ratio (ICER), which represents the additional cost of one unit of outcome gained by one strategy compared to another, was recorded if included in the report. ICER was defined using the following equation:

$$ICER = \frac{C_x - C_y}{E_x - E_y}$$

where C_x is the cost of treatment X, E_x is the mean effect; C_y is the cost of treatment Y and E_y is the mean effect.

1.4. Qualitative assessment of economic evaluation studies

The qualitative assessment of the studies was performed using the checklist questions proposed by Drummond et al. [1] (Table 2). This previously validated checklist identifies ten key elements (items) that address all major principles of economic evaluation and allow the users to assess the methodological quality of an EES. Each item is scored as “yes”, “no” or “can't tell”. One point was given only for each “yes” (zero points for each “can't tell” or “no”). All items were then summed to define a score between 0 and 10 for each article. A high quality study was defined as a score ≥ 7 [8]. Discrepancies were resolved through discussion between the two reviewers (A. F. K. and V. N.) until consensus was reached.

1.5. Statistical analysis and predictive factors of high quality EES

Quantitative data are presented as mean \pm standard deviation (SD) and qualitative data as number and percentage.

The analysis of predictors of high quality EES (defined as a Drummond score ≥ 7) was performed using a two-step approach. First, the association of potential factors with observed high quality (yes/no) was examined by univariate analysis. Quantitative and qualitative variables were transformed, whenever possible, into dichotomous variables using different successive cut-off points. Second, all variables with a p value < 0.15 by univariate analysis were entered into a stepwise logistic

Table 1
Classification of economic evaluation studies.

Examination of one or two alternative strategies?	Examination of both the costs and consequences of alternatives strategies?		
	No	No	Yes
No	Costs only	Consequences only	Partial economic evaluation: Cost–outcome description
	Partial economic evaluation: Cost–description	Outcome description	
Yes	Partial economic evaluation: Cost analysis	Full economic evaluation: Cost-minimization analysis Cost-effectiveness analysis Cost-utility analysis Cost-benefit analysis	

Adapted from Drummond et al. [1].

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