Cost-Effectiveness of Neonatal Surgery: A Review

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Background/Purpose: Technological developments have revolutionized both diagnosis and treatment in neonatal surgery. However, it has been increasingly recognized that financial resources might become insufficient to provide all the medical care that is technically feasible or that patients and families might desire. The purpose of this study is to apply the theory of health economics to neonatal surgery and to explore the extent and the kind of economic evaluation done in neonatal surgery.

Methods: To explore the work done so far, the authors undertook a literature search aimed at costs and effects of surgical interventions in newborns with Ravitch’s surgical index diagnoses of congenital anomalies. Common keywords in cost-effectiveness analysis were used to search Medline.

Results: Evidence about the cost effectiveness of neonatal surgery is largely lacking. This is probably because of difficulties in long-term tracking of the patients and to the problem that most generic quality-of-life measures are not applicable in children yet.

Conclusions: Further cost-effectiveness research in neonatal surgery is warranted to settle priority discussions in health care when neonatal surgery is part of such discussions. Methodology for generic quality-of-life measurement in children is badly needed.


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Cost-effectiveness of neonatal surgery 589

When costs are not an issue, values are only important from a clinical perspective for future patients and current payers of health care. This means that the general public in the sense that they are possible societal values. After all, health care decisions also affect societal value of their quality of life. For instance, if one expects that the quality of life after pediatric surgery is notably different from the quality of life after cardiovascular surgery at an older age. In that case the additional years may not represent the same value for society.

In a cost-effectiveness analysis, life years are corrected for the societal value of their quality of life. For instance, if one expects that the quality of life after surgical intervention is notably different from the quality of life after cardiovascular intervention, the additional life years may not represent the same value. The outcome of a cost-effectiveness analysis therefore is expressed as cost per quality-adjusted-life-year (cost per QALY). There are several elaborately tested methodologies to value the quality of life.5 Whatever valuation method is used, it is important to realize that in economic evaluation it is most appropriate to use societal values. After all, health care decisions also affect the general public in the sense that they are possible future patients and current payers of health care. This means that the general public should value health. Patient values are only important from a clinical perspective when costs are not an issue.9

Economic evaluation is a decision analytic tool that could support medical decision making.5,10 In this review article we explore the extent and the kind of economic evaluation done in the field of neonatal surgery.

Materials and methods

A Medline literature search was done aimed at costs and effects of surgical interventions in newborns with the following Ravitch' surgical index diagnoses of congenital anomalies:11: (1) esophageal atresia, (2) omphalocoele and gastrochisis, (3) anal atresia, (4) intestinal atresia, (5) congenital diaphragmatic hernia, and (6) Hirschsprung’s disease. Keywords used were (1) cost effectiveness, (2) cost analysis or cost(s), (3) QALY, (4) quality of life, (5) survival or long-term follow-up. These keywords, common in cost-effectiveness analysis,5,6,12,13 were combined with neonatal or pediatric surgery or with one of the 6 index diagnoses.11 The keywords were used in different combinations to be sure that the number of hits was maximal.

Because the purpose is to analyze the costs and effects of standard protocols, we excluded case studies of rare complications. We also excluded studies in which for part of the operative or follow-up procedures the influence of uncommon techniques is analyzed. With respect to survival and long-term follow-up, only review articles were included. Only articles published in English in the period from 1989 to 1998 are included.

Results

Our literature search procedure yielded 36 hits. Table 1 summarizes the outcomes research done for the different congenital anomalies. Subsequently, the table sorts the studies to complete and partial analysis. If in a study both costs and effects are analyzed (as in cost-effectiveness analysis or cost-utility analysis), this is a complete economic evaluation. Partial economic evaluation refers to studies that analyze either the effects or the costs of neonatal surgery.

In neonatal surgery only 2 complete economic evaluations have been performed.14,15 When focused solely on “costs,” 5 additional hits were found. Focusing on only “effects” resulted in the majority of hits; there were no hits on “QALY,” 17 additional hits on “quality of life” or “long-term follow-up,” and 12 additional hits on survival. More hits are mentioned in the table, because some of the hits on quality of life included mortality figures. Congenital diaphragmatic hernia has been studied most frequently (15 hits).

Although complete economic evaluation is largely lacking in the field of neonatal surgery, it might be possible to derive an economic appraisal of neonatal surgery.

Table 1. Complete or Partial Economic Evaluation Performed in Neonatal Surgery

<table>
<thead>
<tr>
<th>Diagnoses</th>
<th>Reference Numbers</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Partial Economic Evaluation Complete Economic Evaluation</td>
</tr>
<tr>
<td></td>
<td>Effects</td>
</tr>
<tr>
<td>Congenital diaphragmatic hernia</td>
<td>17, 22-32</td>
</tr>
<tr>
<td>Intestinal atresia</td>
<td>17, 35-37</td>
</tr>
<tr>
<td>Esophageal atresia</td>
<td>17-19, 39-42</td>
</tr>
<tr>
<td>Imperforate anus</td>
<td>16, 17, 36, 43, 44</td>
</tr>
<tr>
<td>Hirschsprung’s disease</td>
<td>17, 45, 46</td>
</tr>
<tr>
<td>Omphalocele/gastrochisis</td>
<td>17, 47-49</td>
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</tbody>
</table>
surgery on the basis of the partial evaluation. Then the outcome measures that have been used should meet the demands of economic evaluation. To explore to what extent the 36 hits provide information that could be used in economic evaluation, Table 2 orders the 36 studies by outcome.

Table 2 shows that the few articles that deal with costs only touch on the direct medical costs of the surgery in the immediate postoperative period. This means that only a small amount of all possible costs is measured. This information is feasible for decision making at the institutional level but not at the societal level. The latter requires a broader scope, because savings in one sector might lead to much higher costs in other sectors. Therefore, the net effect should be studied in the societal perspective.

Furthermore, Table 2 shows that the term quality of life covered a lot of different outcome measures. Only 3 studies use a generic outcome measure and hence have the potential to be used in economic evaluation across disease categories. Most of the investigators present natural units and use those to draw conclusions about quality of life. Although studies in which effects are expressed in natural units might indicate impairments of quality of life, they do not indicate how much quality of life is impaired. For instance, Baeten et al16 stress the importance of dynamic graciloplasty because it improves fecal continence, hence, it improves quality of life. However, societal values for quality of life are lacking, hence, we do not know how important incontinence is with respect to quality of life, or whether it is worth the cost. Such judgements are made validly and explicitly with the use of a generic instrument, as in the studies of Takayanagi et al17 and Ure et al.18 For instance, Ure et al18 compared primary anastomosis and colon interposition. They found differences in morbidity rates between the 2 groups receiving different treatments. However, these differences did not reach statistical significance on a visual analogue scale. This means that these differences in morbidity are not an important aspect of quality of life.

Table 2 also shows that in the cost-effectiveness analysis of Hackam et al15 and Roberts14 only natural units have been used. Note that only under specific circumstances disease-specific outcomes can be used in economic evaluation. Hackam et al15 performed an economic evaluation of 2 interventions for Hirschsprung’s disease: the multistage and the single-stage procedure.15 The treatment costs, exclusive of operating room–related expenditures, were calculated. The investigators suggest that adopting the single-stage procedure could reduce the costs, because the multistage procedure is not more effective, whereas the procedure is twice as expensive. On the basis of natural units they concluded that the 2 interventions were equally effective. Therefore, the differences in costs determine the difference in cost effectiveness between the 2 interventions; the economic evaluation takes the form of a so-called cost minimalization study.

Roberts14 performed an economic evaluation of extra-corpooreal membrane oxygenation (ECMO) in full-term babies with severe respiratory failure. Among the most common diagnoses in this group was congenital diaphragmatic hernia. The cost-effectiveness analysis was based on survival at 1 year of age without severe disability. ECMO would both increase overall survival and the costs when compared with conventional management. The additional costs per additional survivor without severe disability at age 1 year was about £51222. After a 4- to 7-year follow-up period QALYs will be estimated. Expecting a near-normal life expectancy and quality of life that is expected to be only slightly impaired by the underdevelopment of the lungs, the authors hypothesize that ECMO is relatively cost effective.

DISCUSSION

This study finds that little attention has been given to the relationship between costs and effects of neonatal surgical interventions. Only 2 complete economic evaluations have been performed in neonatal surgery.14,15 No generic outcomes were used in these 2 studies. This means that it is possible to find the most cost-effective treatment for a specific condition, but it is not possible to determine how cost effective the treatment is compared with interventions for other congenital anomalies or

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**Table 2. Outcome Measures Used in the Evaluation of Neonatal Surgery**

<table>
<thead>
<tr>
<th>Outcome Measures</th>
<th>Reference Numbers</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Survival</strong></td>
<td>18, 22, 24, 25, 28-32, 35-37, 40, 41</td>
</tr>
<tr>
<td><strong>Natural units</strong></td>
<td></td>
</tr>
<tr>
<td>Neonatal morbidity</td>
<td>14, 15</td>
</tr>
<tr>
<td>Disease-specific functioning at</td>
<td>16, 18, 19, 23, 24, 27, 39, 41-46, 48, 49</td>
</tr>
<tr>
<td>long-term follow-up</td>
<td></td>
</tr>
<tr>
<td>Psychosocial adjustment</td>
<td>43, 45, 47, 49</td>
</tr>
<tr>
<td>Neurodevelopmental status at</td>
<td>24, 45, 48, 49</td>
</tr>
<tr>
<td>long-term follow-up</td>
<td></td>
</tr>
<tr>
<td>Generic quality of life</td>
<td>17-19</td>
</tr>
<tr>
<td>Utilities</td>
<td>—</td>
</tr>
<tr>
<td>Costs</td>
<td></td>
</tr>
<tr>
<td>Health care costs</td>
<td>14, 15, 33, 34, 38, 50, 51</td>
</tr>
<tr>
<td>Costs of family/patients</td>
<td>—</td>
</tr>
<tr>
<td>Costs in other sectors</td>
<td>—</td>
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</tbody>
</table>

**NOTE.** This table does not provide an overview of all outcomes research performed, but only of articles that were found with a search on basis of the keywords that are common in health economics. This means that, for instance, hits on neonatal morbidity are only included when neonatal morbidity was a primary or secondary outcome in economic evaluation or quality of life assessment.
interventions outside neonatal surgery. The analysis of partial evaluations showed that most studies use natural units like neonatal morbidity to draw conclusions about quality of life. Only in 3 studies generic outcomes have been used, thus, only in these 3 studies quality of life is quantified or valued.\textsuperscript{17-19} The available information is, however, insufficient to answer the question whether on a societal level the costs can be accounted for by the health benefits.

The lack of data probably could be explained partially by the clinicians’ long tradition of using disease-specific symptoms, whereas measuring outcomes suitable for economic evaluation is relatively new. Another explanation could be that 2 major problems make collection of cost-effectiveness data difficult. First, information about the survivors in adulthood might be hard to collect because of the long follow-up period after surgery; it might be difficult to track down the patients.\textsuperscript{17} Second, generic quality-of-life measurement in children has not been given much attention until recently. Measuring quality of life in children differs from measuring quality of life in adults; children lack the cognitive ability, language skills, and long-term view that adults have, hence, often proxy measures need to be used. Furthermore, one has to determine which domains suit the quality-of-life perception of children and how these should be phrased. Most existing quality-of-life measures are, therefore, not applicable in children. In this respect it is a promising development that efforts are undertaken to fill in the gap.\textsuperscript{20,21}

The profession of neonatal surgery often is criticized with arguments about the relation of the high costs of an operation with the possibility of low quality of life after survival. This typically is a circumstance for economic evaluation. If evidence about the cost effectiveness of neonatal surgery remains absent, this discipline will be vulnerable, because it remains impossible to tell something about the efficiency of the treatment. Cost-effectiveness analyses have the potential to settle priority discussions for the benefit of society. Therefore, despite the practical problems, efforts should be made to apply the methods of economic evaluation to neonatal surgery.

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REFERENCES