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Early cost-utility estimation of the surgical correction of pectus excavatum with the Nuss bar

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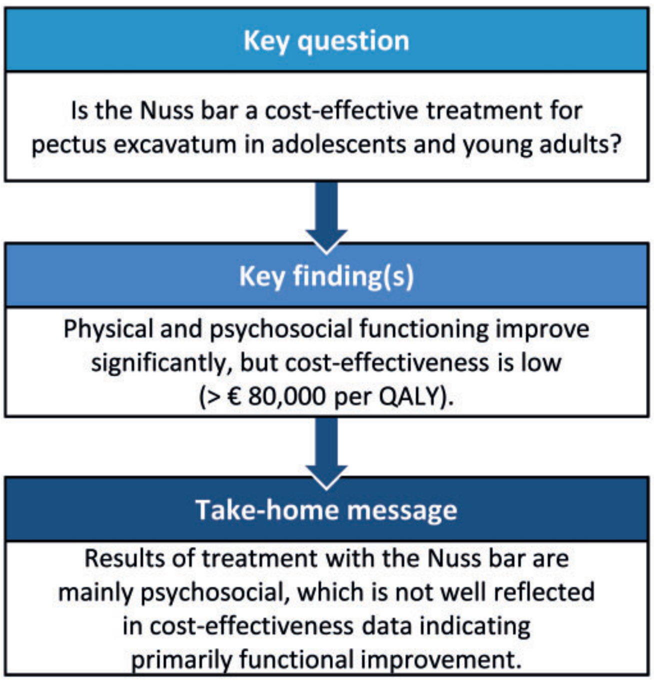

Abstract

OBJECTIVES: The surgical correction of pectus excavatum (PE) with a Nuss bar provides satisfactory outcomes, but its cost-effectiveness is yet unproven. We prospectively analysed early outcomes and costs for Nuss bar placement.

METHODS: Fifty-four patients aged 16 years or older (6 females and 48 males; mean age, 17.9 years; range 16.0–29.4 years) with a PE filled out a Short Form-36 Health Survey (SF-6D) preoperatively and 1 year after a Nuss procedure. Costs included professional fees and fees for the operating room, materials and hospital care. Changes in the responses to the SF-36 or its domains were compared using the Wilcoxon signed rank test and the utility test results were calculated preoperatively and postoperatively from the SF-6D. The quality-adjusted life years (QALYs) were calculated from the results of these tests.

RESULTS: Significant improvements in physical functioning, social functioning, mental health and health transition (all $P < 0.05$) were noted. The other SF-36 subgroups showed improvement; however, the improvement was not significant. The SF-6D utility showed improvement from 0.76 preoperatively to 0.79 at the 1-year follow-up ($P = 0.096$). The mean direct costs were €8805. The 1-year discounted QALY gain was 0.03. The estimated cost-utility ratio was €293 500 per QALY gained.

CONCLUSIONS: Despite a significant improvement in many domains of the SF-36, the results of the SF-6D cost-utility analysis showed only a small improvement in cost-effectiveness (> €80 000/QALY) for patients with PE 1 year after Nuss bar placement. Based on this discrepancy, general health outcome measurements as the basis for cost-utility analysis in patients with PE may not be the best way forward.

Keywords: Cost-effectiveness • Cost utility • Quality-adjusted life-years • Nuss bar • Pectus excavatum

INTRODUCTION

The ever-increasing technological possibilities and associated rising costs in health care systems make choices based on the limitations of treatment options inescapable [1]. A possible way to direct the limited resources towards treatments that have been shown to improve the quality of a patient's life is cost-utility analysis. This method can be used to assess the value of an intervention in terms of improving both quality and quantity of life [2]. Taking this approach results in cost-effectiveness studies gaining in importance as stakeholders try to determine how to fairly distribute limited health care funds. In pectus excavatum (PE), an important anterior chest wall deformity occurring predominantly in boys, a frequent treatment is surgical correction with a Nuss bar. This intervention does not increase life expectancy but is rather aimed at improving the quality of life of these patients.

To describe the health improvement resulting from this kind of surgical correction, one could use quality-adjusted life years (QALYs) to indicate which health effects of an intervention are reasonable compared to its cost [3]. The utility score gives the opportunity to correct and therefore use this score in patients with a great variation in health states [4]. Many different health-related quality-of-life (HRQOL) instruments are used. All these questionnaires have in common the use of responses to different domains in the areas of pain and physical, social and mental health [5]. The Short Form-36 Health Survey (SF-36) is a commonly used HRQOL instrument. The scores of this questionnaire can be converted to a Short Form 6-Dimension (SF-6D) algorithm, which can be used for utility measurements [6]. The SF-6D consists of 6 different health domains: physical function, role limitations, social function, pain, mental health and vitality and uses 11 questions of the SF-36 to assess these domains. For the SF-6D, the score ranges from 0.29 (lowest score in a living person) to 1 (perfect health). The SF-36-6D is a standard validated means to translate the scores from the questionnaire into utilities, which can then be used for cost analysis [7]. The SF-36-6D is commonly used for this purpose.

There is no existing information concerning the cost-effectiveness of the surgical correction of PE with a Nuss bar. Therefore, we prospectively analysed the outcomes and cost-utility of a serial cohort of Nuss procedures during a 1-year follow-up period.

MATERIALS AND METHODS

Patients and data

The study is based on a longitudinal cohort of patients with PE. The study cohort included 54 subjects (6 females, 48 males) with an average age of 17.9 years, whose PE was corrected with a Nuss bar. The number of patients included was not based on a sample-size calculation since we did not make a comparison between possible treatments but were only interested in the cost-effectiveness of this particular treatment.

Inclusion criteria were age 16 years or older and PE suited for treatment with the Nuss bar. Marfan syndrome or other associated connective tissue diseases were not exclusion criteria; however, in this study population, no patients with connective tissue disease were included.

The exclusion criterion was insufficient knowledge of the Dutch language in reading or writing. All patients gave informed consent. The medical ethics committee approved the study.

Patients scheduled for PE surgery, above the age of 16 years, with implantation of a Nuss bar between 2011 and 2016, were asked to fill in the SF-36 questionnaire preoperatively and 12 months after surgery. The survey was completed at additional times after surgery (at 6 months and at 1, 2, 3 and 5 years), but for this analysis, we used only the results obtained preoperatively and after 1 year. The SF-36 Health Survey questionnaire is a 36-item self-report inventory with 8 dimensions of physical and mental domains, including physical functioning, role–physical, bodily pain, general health, vitality, social functioning, role–emotional and mental health [8]. The SF-36 questionnaire has a robust predictive validity for health-related outcomes [9].

The scores from those questions on the SF-36 that comprise the SF-6D were used to compute this score. This method is standard and is accepted worldwide as a tool for cost-effectiveness analysis.

The acquired data were stored in a protected SPSS database (SPSS Inc., Chicago, IL, USA).

In a previous paper, we reported the association between pain and QoL in the first small group of this study population [10].

Cost measures

Total direct medical costs were obtained for all included patients. These costs included a diagnosis-treatment combination that also included a certain percentage of complications during first admission. If patients had a complication that required redo surgery or readmission, these separate costs were taken into account for the analyses presented here. The indirect costs were not estimated because nearly all patients were still students when the measurements were made. Fifty-four patients with PE diagnosed and treated from 2011 to 2016 were included. In the Netherlands, patients with PE can be treated in paediatric surgical departments or thoracic surgical departments. The procedure does not differ per department and includes videothoracoscopy for all patients. Costs were adjusted by the consumer price index till 2017.

Cost data were retrieved from the hospital financial database. These included preoperative outpatient clinic consultations and the cost of inpatient hospital care (laboratory, radiology, operative procedures including implant, staff salaries, pharmacy, physiotherapy and, if necessary, other medical consultations). There were no significant differences observed between paediatric surgical and thoracic surgical departments.

Patient responses to the SF-36 questionnaire were transformed to SF-6D using SF-6D preference-based algorithm licensed software (University of Sheffield, UK) to calculate the health utility value preoperatively and 1 year postoperatively. This programme gives a preference-based utility index in which 1 is perfect health and 0 is equal to death [11]. QALYs were then calculated with the utility scores translated from the SF-6D [6].

Statistical analyses

Data analyses were conducted using IBM SPSS 23 software (SPSS Inc.). Descriptive statistics for variables of interest in this study are presented as percentages. Comparisons between paired scores at measurement moment T1 and T2 for variables from the study group were calculated using the Wilcoxon signed rank test. The cut-off point for significance was set at *P*-value <0.05.

Because no comparison was made between different treatment options, a sample size calculation was not performed. The true costs of correcting PE with a Nuss bar were calculated. The calculated difference in health utility value, preoperatively to 12 months postoperatively using the SF-6D, was presumed to be totally caused by the surgical procedure. From that, a QALY calculation was performed.

In addition, we described 4 often used (maximum) valuations per QALY to give a better oversight worldwide: €33 000, €42 000, €60 000 and €80 000, based on published acceptable costs per QALY in, respectively, England and Wales, the United States, Italy and the Netherlands. We calculated the gain per QALY for treating PE by Nuss bar implantation at 1 year post-surgery.

RESULTS

Fifty-four patients were available for follow-up. SF-36 subgroups showed significant improvement in physical functioning, social functioning, mental health and health transition (all $P < 0.05$), although all domains showed improvement (Table 1).

The mean presurgical health utility value (SF-6D) was 0.76 and 1 year post-surgically it was 0.79, an average gain in health in 1 year of 0.03 QALY. This value approximates what is considered the minimally important difference threshold [12]. Patients with PE displayed a reduced HRQOL, both preoperatively and postoperatively, compared to patients from a healthy reference group [13].

The mean direct cost was €8805 (Table 2). The estimated cost-utility ratio was €293 500 per QALY gained [14]. We then calculated the economic gain after 12 months at QALY valuations of €33 000, €42 000, €60 000 and €80 000, which brought the gain per QALY to €990, €1260, €1800 and €2400, respectively.

Surgery

Patients underwent surgery by a dedicated paediatric or thoracic surgeon. The mean operating time was 54.8 min (standard deviation 14.4) and the mean hospital stay was 6.6 days (standard deviation 1.4).

Table 1: Improvement preoperatively to 1 year postoperatively in the domains of the SF-36 ($N = 54$)

	Mean preoperatively	Mean 1 year postoperatively	*P*-value
Physical functioning	86.67	92.41	0.03
Social functioning	82.41	88.43	0.04
Mental health	71.63	76.30	0.01
Bodily pain	83.94	79.74	0.13
Vitality	66.30	67.96	0.46
General health perceptions	73.70	77.04	0.18
Role limitations from physical health	80.56	86.57	0.16
Role limitations from emotional problems	79.63	80.86	0.90
Health transition	47.22	63.89	<0.01
SF-6D index (utility) score	0.76	0.79	0.10

Means preoperatively and 1 year postoperatively of SF-36 domains. *P*-value calculated with Wilcoxon signed ranks test.
SF-36: Short Form-36 Health Survey; SF-6D: Short Form-6 Dimension.

Table 2: Costs associated with surgical correction of pectus excavatum with the Nuss bar

Cost category	Percent of total ($N = 54$)	Cost in euros
Outdoor clinic	1.0	91.00
Hospitalization and medication	60.4	5321.00
Surgical procedure and implant	34.4	3029.00
Radiology	1.4	124.00
Laboratory	1.3	115.00
Miscellaneous	1.5	125.00

Complications

The total complication rate was 26%. There were 2 small perforations of the pericardium perioperatively, which were managed conservatively, and 1 bleed from the mammary vessel, which necessitated a minithoracotomy. A wound infection and 1 case of pneumonia were both treated with antibiotics. There were 3 reoperations within 1 year: 1 for correction of an early dislocation of the Nuss bar and 2 for chronic pain near the location of the stabilizer, for which 1 stabilizer was removed. All other complications (sensibility changes) were due to the use of epidural pain management and were self-limiting after discontinuation of the epidural. There were no deaths 1 year postoperatively.

DISCUSSION

Overall knowledge and skills seem to have improved over the recent decades in using the Nuss bar for the correction of PE [15]. Little is known, however, about the cost-effectiveness of this surgical treatment. One study addressed the reduction of hospitalization costs in patients whose PE was surgically corrected using a minimally invasive technique [16]. A cost-utility analysis could address this void. Therefore, we analysed a cohort of patients with PE undergoing this procedure.

Cost-utility analyses are increasingly used to compare the costs of medical treatments to decide which procedure is economically acceptable. To take patient preferences into account, it is expressed in calculations as utility or QALY.

For our study, individual treatment costs were identified for all patients in the study group. Direct costs were obtained from the financial systems of the hospitals. Although small differences in specific costs may exist due to local contracts with health insurance companies, these differences are too small to have any real impact. Costs did not include indirect costs, such as transportation or loss of production and patient income; however, it should be noted that the patients in the group were mainly high school students. Theoretically, any indirect cost would lead to an increase in total costs and subsequently to a higher estimated cost-utility ratio.

The patients filled in the SF-36 before surgery and after 1 year. All SF-36 subgroups showed improvement in previous limitations due to physical health and emotional problems, bodily pain, vitality and general health. Only the areas of physical functioning, social functioning, mental health and health transition were significantly improved. SF-36 is a highly validated questionnaire widely applied in medical evaluations; it reflects a broad, general appreciation of physical and mental functioning. The improvement on all levels in our study did not lead to a large increase in the calculated SF-6D index score. Earlier reports showed lower results with the SF-6D than with the Euro-Qol-5 dimensions, which also gives a larger improvement in utility in patients who start out in a low disability state before having surgery [17]. The result of this relatively low SF-6D score leads to high costs per QALY, suggesting a small change with this intervention for this group of patients. This finding is actually contrary to the larger changes taking place in the different domains of the SF-36, where there are significant positive changes in physical, mental and social function, which should lead to an improvement in self-esteem and body image of the patients. Because body image is low before surgery in a population with PE compared to that in a matched healthy population and, in adolescents, is directly related to their well-being, restoration of body image seems to be an important goal [18–20]. The same applies to self-esteem, particularly because restoring self-esteem leads to positive affect and a better capability to cope with life and its challenges [21, 22]. However, the SF-36 is an HRQOL tool and, although it is very good in detecting functional changes, it is less effective in detecting changes in patient satisfaction. In particular, the improvement in domains as markers of patient satisfaction (body image, self-esteem) might be more important than overall improvement in HRQOL or in the health utility score itself. As long as cost-effectiveness is an expression of (improvement of) physical functioning, it will be difficult to prove the utility of the use of surgery/treatment with a main focus on body image and/or self-esteem when looking at the costs or the QALYs.

The costs of improvement after surgery can be calculated, but acceptable cost-effectiveness levels differ around the world. Certain countries have an established threshold of what they consider to be cost-effective per QALY such as £20 000–£30 000 in the UK, USD50 000 in the United States, €60 000 in Italy and €80 000 in the Netherlands. The World Health Organization, however, uses a different formula based on the gross domestic product (GDP) per capita. Threshold values of less than the GDP per capita are highly cost-effective, whereas values between 1 and 3 times the GDP per capita are considered cost-effective, and health interventions costing more than 3 times the GDP per capita are considered not cost-effective [23]. Others recommend that analysts use USD50 000, USD100 000 and USD200 000 per QALY as a more reasonable figure [24]. However, other countries, for example Belgium, Denmark, Norway, Sweden and Finland, do not define a threshold value, because they find the basis for these thresholds unclear [25]. For our calculations, we defined the 4 general levels in euros.

We prospectively gathered costs and outcome scores for a specific group of patients undergoing PE correction. Although the QALY improved at 1 year when we used the SF-6D calculation, it missed statistical significance ($P = 0.096$). We have not found any other study that examined the cost-utility of Nuss bar placement or the real costs. Our mean direct costs were calculated at €8805. Although differences in costs among hospitals in the Netherlands may exist, they are very small.

We also calculated the economic increase using different levels of QALY values used around the world of €33 000, €42 000, €60 000 and €80 000, which brought the gain per QALY on the basis of a 0.03 increase in SF-6D score to €990, €1260, €1800 and €2400. Since it could be reasoned that this increase would account for the whole period that the Nuss bar was in situ (3 years) by the same SF-6D score, the costs could be spread over 3 years. Under the theoretical assumption that there would not be any other change, the gain per QALY would increase to €2970, €3780, €5400 and €7200, respectively.

One should keep in mind that the placement of the Nuss bar is usually followed, approximately 3 years postoperatively, by removal of the Nuss bar, usually as an out-patient procedure. Having this procedure after 3 years adds to the cost of the treatment and thereby affects the overall cost of treating PE with a Nuss bar. However, the removal of the bar also may have an additional effect on the functional outcome and thus lead to a small increase in cost-utility. The study is designed such that the patients do complete the SF-36 after removal of the bar. We decided not to wait to include these results since we do not expect a significant increase in functioning after removal.

Estimation of the cost-utility ratio under assumption of a 3-year period would lead to costs of €97 833 per QALY gained. However, since the patient population is very young and has a long life expectancy, they could profit for decades from the surgical improvement in their chest wall, something that is not clearly visible in the results of the utility gain or in the calculations [26].

Although the cost analysis in this study reflects the health-related economics in the Netherlands, which are different from those in other countries, the study does provide data about the SF-36 and SF-6D, which should allow surgeons around the world to use these tools and to adjust for the measurement of cost-utility in their specific surroundings. This study does show that correction of PE with a Nuss bar is not cost-effective after 1 year in the Netherlands or in the high-cost environment of the United States; nor it is cost-effective in large parts of the rest of the world if QALYs are calculated on basis of the SF-6D.

The percentages from Table 2 reflect real costs from the care of patients with PE undergoing the Nuss procedure. It will be interesting to re-evaluate our patients after approximately 3 years when the Nuss bars are removed, to determine whether the improvements in the SF-6D scores (QALY) are truly long lasting.

CONCLUSION

Surgical treatment of PE in adolescents or young adults with a Nuss bar renders a health benefit for a range of domains of the SF-36. In the short term, the costs exceed the acceptable costs per QALY in the Netherlands as well as elsewhere around the world based on the SF-6D. However, since the benefit should be life-long, the measured gain in quality of life after only 1 year makes it difficult to draw firm conclusions about the cost-effectiveness of the procedure. To appreciate and evaluate the important improvements in self-esteem and body image, a tool that is better than the well-known HRQOL questionnaires to measure patient satisfaction in QALYs is desirable.

Conflict of interest: none declared.

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