

A Pilot Study of the Impact of Surgical Repair on Disease-Specific Quality of Life Among Patients With Pectus Excavatum

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Background: This study was conducted to determine the ability of 2 questionnaires (ie, child and parent versions) to measure physical and psychosocial quality-of-life changes after surgical repair of pectus excavatum.

Methods: The authors administered these questionnaires by telephone interviews with 22 parents and 19 children (ages 8 to 18) before surgery and 6 to 12 months after repair by the Nuss procedure.

Results: The instruments had high test-retest reliability ($Rho > 0.6$ for all retained questions). Children reported significant improvements in exercise intolerance, shortness of breath, and tiredness. Of 9 questions asking the children how they feel or act about their bodies, all but one question showed significant improvement after surgery. Parents also

reported significant improvements in their child's exercise tolerance, chest pain, shortness of breath, and tiredness and decreases in the frequency of the child being frustrated, sad, self-conscious, and isolated.

Conclusions: These questionnaires appear to be more than adequate to measure disease-specific quality-of-life changes after surgery. These data confirm for the first time that surgical repair of pectus excavatum has a positive impact on both the physical and psychosocial well-being of the child. *J Pediatr Surg* 38:916-918. © 2003 Elsevier Inc. All rights reserved.

INDEX WORDS: Pectus excavatum, surgical outcomes, quality of life, body image, instrument validation.

PECTUS EXCAVATUM (PE), or sunken chest, is the most common congenital chest wall disorder, occurring in about one per one thousand children and in boys 4 times more commonly than girls.¹ In this disorder, the sternum and adjacent chest wall are displaced posteriorly toward the spine, creating a depression of the anterior chest wall. Anecdotally, children with severe PE report physiologic and psychologic limitations that are improved after surgery, but to date no systematic research has evaluated these outcomes after pectus repair with the Nuss procedure.

As part of a multicenter study of outcomes of the surgical repair of PE, 2 questionnaires were developed to assess the impact of surgical repair of the disorder on the

quality of life related to both physical and psychosocial functioning. One instrument was for the children's self-reports, the second was for their parents. The purpose of this study was to evaluate the reliability and validity of the instruments, including an investigation of their ability to measure postoperative changes in quality of life.

MATERIALS AND METHODS

The Pectus Excavatum Evaluation Questionnaire (PEEQ) consists of both pre- and postoperative telephone questionnaires for children and their parents. They were developed by a psychologist and a group of clinical experts having years of experience with this disorder. IRB approval was obtained through Eastern Virginia Medical School (IRB # X-00-080). For all phases of the research, a convenience sample of parents were contacted initially, and their children were interviewed if they met age requirements and were available for interview.

In the first phase of this research, 10 qualitative interviews were conducted to assess face validity of the questions: 2 parents at presurgery, 4 parents at postsurgery, one child presurgically, and 3 children postsurgically. Interviewers also collected qualitative data about the instrument during the reliability studies. A planned interrater reliability study was cancelled when the first series of interrater tests showed absolutely no variability between interviewers.

In the second research phase, 24 parents and 19 children participated postsurgically in a test-retest reliability study. Approximately one week elapsed between the first and second administration with slight variations in time because of scheduling conflicts. Spearman's Rho correlation coefficient was used to assess the repeatability of answers across administrations. Spearman's Rho is a statistic used to determine how well answers to the same question at 2 different times correlate with each other. The closer Rho is to one, the more likely the respondents were to answer the question the same way at different times. To assess construct validity, questions were grouped according to the construct

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Table 1. Child Pectus Excavatum Evaluation Questionnaire

Component (Cronbach's Alpha) (N = 18)	Test/Retest Correlation [†]	Question Stem	Median Presurgery Score	Median Postsurgery Score (N = 19)
Psychosocial (0.8)	1.0	Looks in general [‡]	2	1*
	1.0	How looks without shirt [‡]	3	1*
	0.7	Spending rest of life as chest looks now [‡]	3	1*
	0.6	Kids make fun of child because of chest [§]	3	4*
	0.6	Avoids doing things [§]	4	4
	0.9	Hides chest [§]	2	4*
	0.7	Bothered because of the way chest looks [§]	3	4*
	0.7	Feels shy/self-conscious because of chest [§]	3	4*
	1.0	Feels bad about self [§]	3	4*
	Physical (0.9)	0.4	Has trouble exercising [§]	3
0.9		Chest caused shortness of breath [§]	3	4*
0.9		Chest caused child to be tired [§]	2	4*

*Significant improvement from pre to postsurgery at alpha = 0.05 using Wilcoxon Signed Rank Test.

[†]Spearman's Rho on collapsed responses to test-retest questions before revision. Value of one indicates complete agreement.

[‡]Scale: 1 = very happy; 2 = mostly happy; 3 = mostly unhappy; 4 = very unhappy.

[§]Scale: 1 = very often; 2 = often; 3 = sometimes; 4 = never.

being measured (ie, physical versus psychosocial symptoms), and Cronbach's alpha was used to test internal consistency of these items. Cronbach's alpha is a statistic used to determine if responses on several questions are similar to each other, and, again, the closer to one the more likely these questions were to measure the same idea. Based on the results of the reliability analyses and the qualitative data, specific items were by objective criteria scrutinized to decide whether to remove, alter, or include each item.

Validity of the revised instrument was assessed in a third study of the PEEQ with a new cohort of patients and parents to determine if the instrument was responsive in the measurement of postoperative change. Twenty-two parents and 19 children (ages 8 to 18) were interviewed preoperatively and reinterviewed at least 6 months after surgery by the Nuss procedure. The Wilcoxon Signed Rank test was used to determine statistical significance of differences, with a 0.05 level of significance. The Wilcoxon Signed Rank test is similar to a paired *t* test, but is used for noncontinuous and nonnormally distributed data such as these.

RESULTS

The test-retest reliability of the PEEQ is summarized in Table 1 (for children) and Table 2 (for parents). More than half (53%) of the parents' questions had test-retest correlation coefficients of 0.7 or above. Conversely, only one third of the children's questions had coefficients above 0.7. Collapsing children's response categories (eg, letting "very happy" = "mostly happy") improved reliability, with over half (57%) of the questions achieving a correlation at least 0.7.

Questions with coefficients less than 0.7 were either reworded based on qualitative data or were dropped from the final questionnaire used in the multicenter study. For example, because the word *exercise* was interpreted by

Table 2. Parent Pectus Excavatum Evaluation Questionnaire

Component (Cronbach's Alpha) (N = 24)	Test/Retest Correlation [†]	Question Stem How Often Does/Is the Child [‡]	Median Presurgery Score	Median Postsurgery Score (N = 22)
Psychosocial (0.8)	0.7	Irritable	3	4*
	0.9	Frustrated	2.5	4*
	0.6	Sad/depressed	3	4*
	0.7	Restless	4	4*
	0.5	Isolated	3	4*
	0.5	Made fun of	4	4*
	Self-consciousness [§]	0.7	Reluctant to wear bathing suits	1
		How often parent is concerned about		
Caregiver concern	0.8	effects of pectus on child's life	1	3*
Physical (0.7)	0.7	Have trouble exercising	2	4*
	0.4	Have chest pain	3	4*
	0.7	Have shortness of breath	2	4*
	0.7	Feel tired	2	4*
	1.0	Have problems gaining weight	3	4

*Significant improvement from pre to postsurgery at alpha = 0.05 using Wilcoxon Signed Rank Test.

[†]Spearman's Rho on responses to test-retest questions prior to revision. Value of one indicates complete agreement.

[‡]Scale: 1 = very often; 2 = often; 3 = sometimes; 4 = never.

[§]Remaining items in this component removed after reliability studies because of inapplicability to most participants, see text.

most participants to mean only organized activities, it was changed to *being physically active* for parents and *running around or exercising* for children. Questions about limitation of activity in gym, camp, and daycare were found to be inapplicable to over 50% of respondents and thus were dropped. For both child and parent forms of the PEEQ, the items assessing psychosocial symptoms (ie, body image and emotional experiences) and physical symptoms were internally consistent, as illustrated by Cronbach's alphas above 0.7 in all cases.

Tables 1 and 2 further summarize responses to the PEEQ pre- and postoperatively in our evaluation of the impact of PE repair by the Nuss procedure. There were no negative postoperative changes reported by the children or parents. *Children* indicated significant improvements in frequency of experiencing exercise intolerance, shortness of breath, and fatigue. *Parents* also reported significant improvements in their child's exercise tolerance and decreases in the frequency of chest pain, shortness of breath, and fatigue. All indicators of psychosocial functioning improved, including body image satisfaction; feeling frustrated, sad, restless, and isolated; and experiences of social ridicule. Of 9 psychosocial questions asked of the children, all but one item showed significant improvement after surgery. Similarly, their

parents confirmed significant improvements on all psychosocial items and also conveyed a significant reduction in their own concerns about the effects of PE on their child's life.

DISCUSSION

Our research with the PEEQ provides clear and consistent evidence of the reliability and validity of this instrument. This tool holds promise in further documenting the utility of PE repair. Statistical significance in our relatively small sample size was caused by the uniformly substantial improvements that occurred, not only as experienced by the children themselves, but also by their parents. These data confirm for the first time that surgical repair of PE has a positive impact on the well-being of the child. The significant quality-of-life improvements in both physical and psychosocial functioning should encourage physicians to approach PE repair in the same manner as they do other deformities that have a deleterious impact on the physical and psychological well-being of the individual.

REFERENCE

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Discussion

D. Nuss (Norfolk, VA): Anecdotally, I am sure all surgeons who do pectus surgery have noted that the children always say they feel so much better. The trouble is that these studies don't always show the improvement. This was a different way of attacking this problem. We got tremendously positive reports from the parents about how much better the children are feeling about them-

selves, how much better their exercise tolerance is, how much better they are behaving, how much better they are doing in sports. We hear comments like, "Johnnie used to try to play soccer, but he always dropped out. Now he's the number one player on the team." That was anecdotal, so this method of measuring improvement was developed, and, as you can see, it has been remarkably successful.