

Quality of life of patients who have undergone the minimally invasive repair of pectus carinatum

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Received 12 November 2011; received in revised form 23 January 2012; accepted 13 February 2012

Abstract

OBJECTIVES: Several studies previously demonstrated an improvement in the quality of life (QoL) of the patients undergoing a minimally invasive repair of pectus excavatum, but there are no data about such improvement following the minimally invasive repair of pectus carinatum (PC) deformity. The purpose of this study was to investigate the effects of the minimally invasive repair of PC deformity on the psychosocial and physical functioning of the patients.

METHODS: Among 40 patients who underwent minimally invasive repair for PC deformity from July 2008 to March 2011, 35 patients accepted to answer the QoL questionnaires, and 30 of them who had completed the postoperative 6th month were evaluated in this study. The modified two-step Nuss questionnaire was used for the QoL assessment. All patients and their parents completed the appropriate questionnaires regarding the patients' preoperative psychosocial and physical functioning, and they were asked to answer the same questions on the postoperative 6th month. The results from these questionnaires were analysed using Wilcoxon signed rank test to investigate the effects of the minimally invasive repair of PC deformity on psychosocial and physical functioning of the patients.

RESULTS: The questionnaires used in the study confirmed the positive impact of the surgical correction on psychosocial and physical well-being in the patients and their parents. Spearman's ρ correlation coefficient determined how well the answers to the same question at two different times correlated with each other, and Cronbach's alpha demonstrated the internal consistency of these answers. These two parameters showed that the statistical results of the study were reliable enough. Statistical analysis of the scoring of the individual questions and the total scoring of individual patients revealed a statistically significant improvement ($P < 0.05$) following surgery. Similar significant improvements were observed in the total scoring of individual parents and in most scoring of the individual questions (10 of 13, 77%) in the parental questionnaire ($P < 0.05$).

CONCLUSIONS: The results of this study confirm for the first time that minimally invasive repair of PC deformity has a positive impact on both psychosocial and physical functioning of the patient, which is supported by parental assessment.

Keywords: Pectus carinatum • Quality of life • Body image • Nuss questionnaire

INTRODUCTION

After pectus excavatum (PE), pectus carinatum (PC) is the second most common congenital deformity of the sternum and anterior chest wall. Both these deformities predominantly affect males. In some patients, the deformed thoracic cage may cause symptoms related to the thoracic viscera, mostly the heart and the lung [1]. However, most patients have serious loss of self-esteem and affected social behaviour because of the cosmetic disfigurement [2].

Chest wall deformities can have a negative impact on social interaction. Patients often experience feelings of shame and they hide their chests. Social withdrawal and features of depression may occur. As the patient matures, he/she thinks that the

deformity is an obstacle to his/her future relationships. Characteristically, these patients have reduced self-confidence and self-esteem. Besides the studies that showed good cosmetic results [3–5], several authors also demonstrated an improvement in the quality of life (QoL) and body image following both the open and minimally invasive repair of PE [2, 6–10].

In a very recent study, Steinmann *et al.* [11] published their results about the QoL and impaired body image of the patients with PC together with PE, but this study is based only on the data prior to surgical correction. Apart from this study, there are no data about the QoL of the patients with PC in the literature. The purpose of this study was to investigate the effects of the minimally invasive repair of PC deformity on the psychosocial and physical functioning of the patients based on questionnaires

performed twice (pre- and post-operatively) on both patients and their parents.

PATIENTS AND METHODS

Patients

A total of 40 patients were operated on at our clinics for minimally invasive repair of PC deformity from July 2008 to March 2011, and 35 patients answered the QoL questionnaires. Among them, 30 patients who had completed the postoperative 6th month were evaluated in this study.

Written consent was obtained from all patients, and their parents, according to the patient's age. The study design was approved by the institutional board and ethics committees of the university.

All but one patient in the study were male. The median age was 14.5 years (range, 10–27 years) at the time of operation. All patients presented with cosmetic complaints. The deformity involved the lower sternum (chondrogladiolar type PC), and the sternum was compressible in all patients, 12 of them (40%) having asymmetrical deformity. Postero-anterior and lateral chest X-rays, pulmonary function tests, electrocardiograms and echocardiography were routinely undertaken. Chest computed tomography was not taken for every patient and we did not calculate sternum-vertebral ratio indices as part of our preoperative assessment. The indication for surgery in all patients was the persistent request to relieve the cosmetic complaints, and each patient was informed about the potential risks and complications of the surgery.

Surgical technique

All operations were performed by the same surgeon (M.Y.). The minimally invasive repair technique mainly consisting of a pre-sternally placed metal bar compressing the sternum was based on the principles of the operation defined by Abramson *et al.* [12], and later modified by Yüksel *et al.* [13], using a newly designed bar and stabilizing system made of 316L steel (Hipokrat

Medical Devices Manufacturing and Marketing Inc., Izmir, Turkey). An epidural catheter was placed in all patients prior to the operation. No chest drain was used after the surgery. We obtained a postoperative X-ray of the chest to control the placement of the bar and the presence of a pneumothorax. Patient-controlled analgesia using epidural fentanyl and bupivacaine in the first 48 h postoperatively, followed by oral tramadol and etodolac was the method of pain control for each patient.

We tend to keep the bar in place for at least two years for a satisfying result. Early bar removal may cause recurrence of the deformity, while keeping the bar in place for an extended period of time may end up with iatrogenic PE, especially during puberty. For patients older than 18, bar removal can be postponed till the end of the 3rd or 4th year depending on the follow-up.

QoL assessment

All patients and their parents completed the appropriate questionnaires regarding their preoperative condition, and they were asked to answer the same questions in the postoperative 6th month. The questionnaire was administered by a senior assistant, who was blinded to the development of the questionnaires and to the endpoints of the study.

Lawson *et al.* [6] first published and validated the two-step Nuss questionnaire (NQ) for a paediatric population (including questionnaires for both patients and their parents). The questionnaire assesses the effect of the surgical procedure on the psychosocial and physical functioning of the patients. Krasopoulos *et al.* [2] modified the NQ for patients (but not for parents) for use also in adults, reversing the scoring of first three questions for better assessment. We modified the NQ for parents also, according to the principles of Krasopoulos *et al.* [2] and translated all questionnaires into Turkish. The modified two-step questionnaires were used for patients (NQ-mP) and for their parents (NQ-mPr) to evaluate the QoL of patients. The questionnaires included 12 items for patients and 13 items for parents (about the patients) with values between 1 and 4 [2]. The questions and the scoring system are given in Table 1 and Table 2.

Table 1: Modified Nuss questionnaire for patients

N ^a	Question	Scoring
Q1	Looks in general	Very happy: 4, mostly happy: 3, mostly unhappy: 2, very unhappy: 1
Q2	How chest looks without shirt	Very happy: 4, mostly happy: 3, mostly unhappy: 2, very unhappy: 1
Q3	Spending rest of life as chest looks now	Very happy: 4, mostly happy: 3, mostly unhappy: 2, very unhappy: 1
Q4	Others make fun of him/her because of chest	Very often: 1, often: 2, sometimes: 3, never: 4
Q5	Avoids doing things	Very often: 1, often: 2, sometimes: 3, never: 4
Q6	Hides chest	Very often: 1, often: 2, sometimes: 3, never: 4
Q7	Bothered because of the way chest looks	Very often: 1, often: 2, sometimes: 3, never: 4
Q8	Feels shy/self-conscious because of chest	Very often: 1, often: 2, sometimes: 3, never: 4
Q9	Feels bad about self	Very often: 1, often: 2, sometimes: 3, never: 4
Q10	Has trouble exercising	Very often: 1, often: 2, sometimes: 3, never: 4
Q11	Chest causes shortness of breath	Very often: 1, often: 2, sometimes: 3, never: 4
Q12	Chest causes him/her to be tired	Very often: 1, often: 2, sometimes: 3, never: 4

The format is adapted from that of the Nuss questionnaire for children, as published by Krasopoulos *et al.* [2].

N: number; Q: question.

^aQ1–9: For psychosocial; Q10–12: For physical functioning of the patients.

Table 2: Modified Nuss questionnaire for parents

N ^a	Question	Scoring
Q1	Irritable	Very often: 1, often: 2, sometimes: 3, never: 4
Q2	Frustrated	Very often: 1, often: 2, sometimes: 3, never: 4
Q3	Sad/depressed	Very often: 1, often: 2, sometimes: 3, never: 4
Q4	Restless	Very often: 1, often: 2, sometimes: 3, never: 4
Q5	Isolated	Very often: 1, often: 2, sometimes: 3, never: 4
Q6	Made fun of him/her	Very often: 1, often: 2, sometimes: 3, never: 4
Q7	Reluctant to be in public while wearing bathing clothes that would show the chest	Very often: 1, often: 2, sometimes: 3, never: 4
Q8	How often parents are concerned about effects of the deformity on patient's life	Very often: 1, often: 2, sometimes: 3, never: 4
Q9	Have trouble exercising	Very often: 1, often: 2, sometimes: 3, never: 4
Q10	Have chest pain	Very often: 1, often: 2, sometimes: 3, never: 4
Q11	Have shortness of breath	Very often: 1, often: 2, sometimes: 3, never: 4
Q12	Feel tired	Very often: 1, often: 2, sometimes: 3, never: 4
Q13	Have problems gaining weight	Very often: 1, often: 2, sometimes: 3, never: 4

The format is adapted from that of the Nuss questionnaire for children, as published by Lawson *et al.* [6].

N: number; Q: question.

^aQ1–8: For psychosocial; Q9–13: For physical functioning of the patients.

Statistical analysis

Spearman's ρ correlation coefficient was used to assess the repeatability of answers across administrations. This coefficient is used to determine how well answers to the same question at two different times correlate with each other. To assess construct validity, questions were grouped as physical versus psychosocial functioning, and the internal consistency of these items was tested using Cronbach's alpha. It is a statistic used to determine whether responses to several questions are similar to each other, and the closer to one the more likely these questions were to measure the same idea [6]. The distribution of all data was analysed using the Kolmogorow–Smirnov test. The results from the NQ-mP, and NQ-mPr were analysed using the Wilcoxon signed rank test to determine statistical differences. A $P < 0.05$ was considered significant.

RESULTS

Clinical characteristics

Minimally invasive repair of PC using a single bar and two stabilizers was performed in every patient. The median duration of surgery was 65 min (range, 45–110 min). The median in-hospital stay was 5 days (range, 2–10 days). Tolerance of the implant was good; 27 patients (90%) were off analgesics on postoperative day 14, and three patients required prolonged usage of oral analgesics for a total of 4–6 weeks.

QoL assessment

Patients. The mean scores of the patients given for both psychosocial (19.9 ± 5.7 preoperatively, and 31.1 ± 2.9 postoperatively) and physical functioning questions (9.7 ± 1.9 preoperatively, and 10.9 ± 0.9 postoperatively) revealed a statistically significant improvement following the operation ($P < 0.0001$, and $P = 0.001$, respectively). Analysis of the total

scores of the individual patients demonstrated a significant improvement. Total scores obtained per patient for NQ-mP demonstrated a statistically significant improvement following the minimally invasive repair of PC (29.6 ± 6.4 preoperatively, 42.0 ± 2.9 postoperatively, $P < 0.0001$, $\rho = 0.39$). There was also a statistically significant improvement in the analysis of the mean scores obtained for each question in the NQ-mP (Table 3).

Parents. The mean scores of the parents given for both psychosocial (22.2 ± 3.5 preoperatively, and 28.7 ± 2.7 postoperatively) and physical functioning questions (15.3 ± 2.1 preoperatively, and 17.1 ± 1.1 postoperatively) revealed a statistically significant improvement following the operation ($P < 0.0001$ for both scores). Analysis of the total scores of the individual parents also demonstrated a significant improvement. Total scores obtained per parent for NQ-mPr demonstrated a statistically significant improvement following the minimally invasive repair of PC (37.6 ± 4.5 preoperatively, 45.7 ± 3.0 postoperatively, $P < 0.0001$, $\rho = 0.54$). There was also a statistically significant improvement in the analysis of the mean scores obtained for most questions in the NQ-mPr, except for question numbers 6, 9 and 10 (Table 4). Overall, there was a significant improvement in 77% of the questions in the NQ-mPr (10 out of 13).

DISCUSSION

The two-step questionnaires for patients and their parents used in this study confirmed that there was a positive impact of the minimally invasive repair of PC deformity on psychosocial and physical well-being of the patients.

Cronbach's alpha is a coefficient of reliability that normally ranges between 0 and 1. According to the rules proposed by George and Mallery [14], the reliability is acceptable when Cronbach's alpha is between 0.70 and 0.79, and questionable when it is between 0.60 and 0.69 and unacceptable if it is less than 0.5. We can say that the scales used in this study showed nearly adequate internal consistency reliability. Only Cronbach's alpha of the physical functioning question scales in NQ-mPr was

Table 3: Changes in means for the individual patient question scores

Component (Cronbach's alpha)	Test/retest correlation ^a	Q ^b	Presurgery score (mean ± SD)	Postsurgery score (mean ± SD)	P-value
Psychosocial (0.84)	0.36	Q1	2.2 ± 0.9	3.1 ± 0.4	0.0002
	0.23	Q2	1.4 ± 0.6	3.2 ± 0.5	<0.0001
	0.13	Q3	1.1 ± 0.3	3.2 ± 0.6	<0.0001
	0.36	Q4	3.6 ± 0.7	3.9 ± 0.2	0.004
	0.24	Q5	2.6 ± 1.0	3.4 ± 0.6	0.0003
	0.61	Q6	2.3 ± 1.3	3.7 ± 0.5	<0.0001
	0.31	Q7	2.0 ± 1.0	3.4 ± 0.6	<0.0001
	0.43	Q8	2.4 ± 1.2	3.6 ± 0.5	<0.0001
	0.13	Q9	2.4 ± 0.9	3.6 ± 0.5	<0.0001
Physical (0.70)	0.17	Q10	3.0 ± 0.6	3.3 ± 0.5	0.01
	0.39	Q11	3.3 ± 0.7	3.7 ± 0.4	0.005
	0.13	Q12	3.4 ± 0.9	3.9 ± 0.3	0.01

Q: question; SD: standard deviation.

^aSpearman's ρ correlation coefficient.

^bFor questions in detail, refer to Table 1.

Table 4: Changes in means for the individual parents question scores

Component (Cronbach's alpha)	Test/retest correlation ^a	Q ^b	Presurgery score (mean ± SD)	Postsurgery score (mean ± SD)	P-value
Psychosocial (0.70)	0.45	Q1	3.0 ± 0.8	3.5 ± 0.5	0.002
	0.41	Q2	2.8 ± 0.8	3.3 ± 0.5	0.001
	0.24	Q3	2.8 ± 0.6	3.5 ± 0.5	<0.0001
	0.38	Q4	3.1 ± 0.5	3.6 ± 0.4	0.0001
	0.25	Q5	2.8 ± 0.9	3.6 ± 0.4	0.0001
	0.24	Q6	3.8 ± 0.4	3.9 ± 0.2	0.16
	0.28	Q7	2.1 ± 1.2	3.6 ± 0.6	0.0001
	0.26	Q8	1.7 ± 1.0	3.4 ± 0.6	<0.0001
Physical (0.66)	0.37	Q9	3.1 ± 0.7	3.4 ± 0.5	0.06
	0.42	Q10	3.3 ± 0.6	3.8 ± 0.4	0.07
	0.46	Q11	3.6 ± 0.7	3.9 ± 0.3	0.02
	0.28	Q12	3.2 ± 0.7	3.8 ± 0.3	0.0001
	0.85	Q13	1.8 ± 1.1	2.1 ± 1.0	0.004

Q: question; SD: standard deviation.

^aSpearman's ρ correlation coefficient.

^bFor questions in detail, refer to Table 2.

below 0.70. Similar low scores (<0.70) in internal consistency reliability have been reported in several other studies and are explained by the developers as 'deliberately designed to be heterogeneous' [7, 15, 16].

Multiple individual, social and environmental factors interact in the creation of one's own assessment of body image. It is known that patients who are dissatisfied with their body image can experience an improvement in QoL, and psychosocial and physical functioning when they receive appropriate treatment [17–19]. In the case of PE, there are several studies in the literature demonstrating the improvement in QoL and body image following the minimally invasive repair of the deformity [2, 6–10]. In the study of Steinmann *et al.* [11], there were 71 patients with PE and 19 patients with PC deformity. Of patients with PC deformity, 26.3% had both cosmetic and health-related reasons to undergo surgery; 68.4% sought surgical correction for solely cosmetic reasons (the remaining 5.3% did not answer). Social

activities were more frequently impaired in PC than in PE patients. Compared with the control group, QoL scores in PC patients were significantly reduced, and physical difficulties were insignificantly ($P=0.096$) increased. Compared with PE group, patients in PC group demonstrated insignificantly higher degrees of body image impairment ($P=0.07$). This is a very informative study, but since there were no assessments after surgical correction, one cannot guess the impact of the surgical repair on the psychosocial and physical functioning of the PC patients. In our study, all patients (100%) had cosmetic reasons to undergo surgery.

We have shown that the minimally invasive repair of PC has a beneficial impact on the QoL of the patients. There were significant improvement of the psychosocial, physical and the total scores in both NQ-mP (for patients) and NQ-mPr (for parents). When each question is investigated separately, all the patients' results indicated significant improvement in psychosocial and

physical functioning. Parents' results also reported similar significant results, except for three questions where the improvement was not statistically significant.

As mentioned earlier, there is no specific study evaluating the impact of the surgical repair of PC on the QoL of the patients, unlike the reports on such impact in patients with PE deformity [6–10]. Compared with these reports in the literature, the study population in our study was small ($n=30$). This is simply because PC is not as common as PE [1]. However, further work is needed to verify that the information obtained from a small number of participants is representative of the larger population.

Another important point this study is unable to address is that we do not know the long-term effects of the minimally invasive repair of the deformity has on patients' physical and psychosocial functioning. The beneficial effects demonstrated in this study may be lost in time. The current study was done 6 months after the surgical correction of the deformity, which is a time when the novelty and investment of discomfort is still quite fresh in the minds of the participants. It is quite true that one can expect an enthusiastic placebo-like response to this experience. The same point has been marked by Krasopoulos *et al.* [2] and Roberts *et al.* [20] concerning the QoL in patients with PE deformity. We agree with their proposal that long-term studies with a longer follow-up period should be undertaken in future studies to confirm that the positive effects remain.

In conclusion, the questionnaires used in this study appear to be adequate to measure disease-specific QoL changes after the surgical correction of PC deformity. The results of this study confirm for the first time that the minimally invasive repair of PC has a positive impact on both psychosocial and physical functioning of the patient, which is supported by parental assessment.

Conflict of interest: none declared.

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