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Long-term evaluation of patient satisfaction and quality of life in pectus excavatum repair

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Abstract

Background: The aim of our study was to evaluate long-term patient satisfaction and quality-of-life improvement in grown-up patients treated for pectus excavatum with the Nuss procedure in the pediatric age, searching for correlation between preoperative characteristics and long-term outcomes.

Methods: At first, we performed a retrospective analysis of pediatric patients undergoing the Nuss procedure in a 5-year period. We administered, at least 5 years after bar removal, a single-step questionnaire to focus on the assessment of patient satisfaction with operative results.

Results: Most patients stated general health and exercise tolerance were improved after the operation. High levels of overall satisfaction were reported after Nuss repair, with 95.6% of patients being either satisfied or very satisfied. Overall, 87.0% of patients stated they would have the operation again. The high overall satisfaction after surgery was not correlated with the deformity severity and the presence of physical symptoms before correction.

Conclusions: Patients expressed high levels of satisfaction in terms of self-image and quality of life. Improvement in cosmetic appearance and health in general translated in most patients in an improvement of social life. The degree of postoperative pain after the Nuss procedure is the overriding factor in the patient's perception of the quality of the postoperative course.

Keywords: Pectus excavatum, Chest wall deformity, Nuss, MIRPE, Patient satisfaction, Quality of life, Pediatric

Background

Pectus excavatum (PE) is the most common congenital chest wall deformity, occurring in about one per one thousand children and in boys 4 times more commonly than girls. The defect arises when excessive or unequal growth of the lower costal cartilages pushes the sternum posteriorly, resulting in a depression of the anterior chest wall. The severity of the defect ranges from a minor degree of deformity to a severe concavity that displaces mediastinal organs and is expressed as the Haller index [1–3]. Although cardiovascular or respiratory symptoms such as chest pain, dyspnea, and diminished exercise tolerance can occur, more often, instead, the deformity determines a poor self-image and avoidance of social interactions with a psychological impact that may be devastating for adolescents and young adults, resulting in a significant reduction of quality of life [4]. As a consequence, a proportion of patients with this condition pursue surgical correction for what is essentially a cosmetic problem. Thomas Rowland et al. study on the effects of pectus excavatum deformity on endurance fitness and cardiorespiratory functional reserve in adolescent boys showed that patients with pectus excavatum deformity have lower endurance fitness than controls associated with reduced cardiac output and tidal volume responses to exercise [5]. Also, Maj Lesbo et al. studied effect of pectus excavatum in exercising teenagers with pectus excavates. They found patients are not capable of increasing their stroke volume to the same extent as normal young people, concluding that habitual lack of exercise training among pectus patients could have an

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impact on the lower performance in this group [6]. Children and adolescents are considered the ideal candidates for minimally invasive repair of pectus excavatum (MIRPE), also known as the Nuss procedure, because cartilage is still malleable and elastic; thus, reconfiguration of the chest wall can be obtained by applying force from a convex metal bar against the depressed sternum. Successful permanent remodeling over time of the chest wall thanks to the Nuss procedure has been proved by extensive literature [2, 4]. Long-term patient satisfaction and overall quality-of-life improvement related to both physical and psychosocial functioning, in patients treated in the pediatric age, has been less investigated, especially once patients become adults. Herein, we report a single-institution experience in the management of pediatric PE with the Nuss procedure, evaluating patient satisfaction and quality-of-life improvement in grown-up patients that were treated in the pediatric age, at least 5 years after bar removal. For this purpose (long-term evaluation), we used a single-step questionnaire to focus on the assessment of patient satisfaction with operative results after bar removal. Correlation between sex, severity of the defect, the presence of cardiorespiratory involvement with patient satisfaction, and quality-of-life improvement after surgical correction was searched. In the end, the study aims at establishing whether or not these patients, aware of the whole surgical reconstruction process, would recommend it to other patients or themselves to undergo the procedure again.

Materials and methods

After institutional review board approval, a retrospective review was performed to identify patients who underwent PE repair by the Nuss procedure at a single institution from 2007 to 2014. Included in the analysis were adult patients aged 18 years or older who underwent primary PE repair during the pediatric age, with at least a 5-year follow-up after the bar removal (Fig. 1). Excluded from the analysis were patients with any history of connective tissue disorders and recurrent PE and patients who underwent combined cardiothoracic procedures. Data on demographics, operative details, and operative outcomes were collected from the medical charts. Preoperative evaluation included clinical examination, cardiac evaluation through EKG and cardiac ultrasounds, static pulmonary function tests, and computed tomography scan. The indication for PE repair was a severe pectus deformity as defined by a Haller index greater than 3.5 on computed tomography scan imaging or the presence of symptoms related to impairment of cardiopulmonary function or body image issues regardless the Haller index, in patients aged 11–15 years. Pre- or postoperative psychological support was not systematically given but only offered if required by patients and family.

All patients underwent a conventional video-assisted Nuss procedure. Intraoperatively, if one metal bar did not satisfactorily correct the deformity, a second bar was placed superiorly. The bar was then stabilized with one stabilizer.

At the end of the procedures, patients were admitted to the pediatric intensive care unit (ICU) for the first 24 postoperative hours. In all patients, an epidural catheter was placed during the induction of anesthesia and kept for 4 days. All patients were carefully instructed to always sleep supine and to avoid sudden twisting and bending thoracic movements, performing any kind of sport during the first 4 weeks after surgery was not permitted, and walking a lot and going on with respiratory physiotherapy exercises learnt during the hospital stay were instead recommended. Association of codeine and acetaminophen was prescribed as home analgesia, to take only if needed. Bar removal was performed between 24 and 36 months after primary repair. Patients were systematically followed up in outpatient clinic at 4 weeks after both the operations and then on an annual basis until coming of age (18 years old).

Patient satisfaction and quality-of-life assessment

For long-term evaluation focusing on the assessment of patient satisfaction with operative results after bar

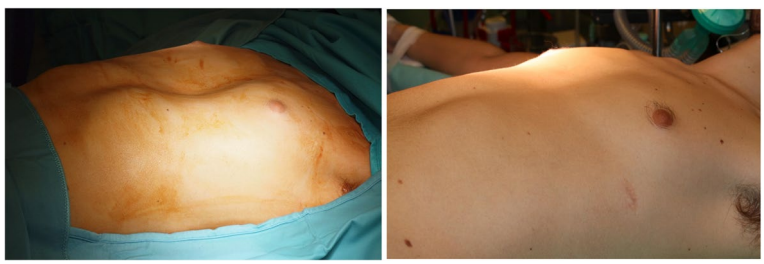

Fig. 1 Preoperative and postoperative result

removal, we used the single-step questionnaire, introduced by Krasopoulos et al. and modified by Sacco Casamassima et al. The questionnaire consisted of 11 items (Table 1) that measured the following domains: general health perception/physical functioning, social belonging, chest pain/discomfort, and satisfaction. We further enriched the questionnaire by repeating question no. 7 and no. 8 to better understand timing of chest pain/discomfort.

Patients were contacted by telephone for the study presentation, all participants received written information about the study and the assurance of confidentiality, and upon acceptance to enrollment, questionnaires were then delivered via e-mail. Filled questionnaires were collected by a research fellow who was not part of the surgical team and was blinded to patient characteristics and prior course.

Statistical analysis

Categorical values were presented with descriptive statistics. Association between patients' postoperative satisfaction, represented by questions 5, 10, and 11 and preoperative HI, was assed via linear regression analysis. Correlation between the presence of symptoms and patients' postoperative satisfaction was studied with Mann-Whitney *U*-test. The association between influence on everyday life of postoperative appearance and postoperative satisfaction was evaluated with Spearman rank test. A p-value < 0.05 was considered statistically significant.

Results

During the study period, a total of 36 patients with PE underwent Nuss minimally invasive repair at our institution. Preoperative characteristics of patients are reported in Table 2.

The median age at operation was 14.2 (age 11–15 years old). Mean Haller index was 4.8 +/− 1.75.

Although the primary indication for surgery was cosmetic in every case, 16 (44.4%) of the patients also had signs of cardiac compressions by the deformed sternum; furthermore, 14 (38.9%) patients complained of shortness of breath on exertion with occasional sternal pain. Mean operative time was 74 min +/− 16 min. In all patients but one, only 1 bar was placed. No situations

Table 2 Preoperative characteristic of patients

Variable	Value
Male	31 (86.1%)
Age, years, mean +/− SD	126 +/− 2.0
Haller index (CT scan), mean +/− SD	4.8 +/− 1.75
Electrocardiogram abnormalities, *n* (%)	11 (30.6%)
Cardiac ultrasound abnormalities, *n* (%)	16 (44.4%)
Restrictive syndrome at pulmonary function test, *n* (%)	8 (22.2%)
Depression, *n* (%)	1 (2.8%)
Preoperative symptoms, *n* (%)	14 (38.9)
- Dyspnea on exertion	14 (38.9%)
- Chest pain	1 (2.8%)
- Palpitations	1 (2.8%)

Table 1 Modified single-step questionnaire

Question	Scoring	Median
1. Health in general after operation	Much better now, 5; somewhat better now, 4; about the same, 3; 4 somewhat worse now, 2; much worse now, 1	5
2. Exercise capacity after operation	Much better now, 5; somewhat better now, 4; about the same, 3; 4 somewhat worse now, 2; much worse now, 1	5
3. Extent that chest appearance interferes with social life before operation	Extremely, 5; quite a bit, 4; moderately, 3; slightly, 2; not at all, 1.4	4
4. Extent that chest appearance interferes with social activity after operation	Not at all, 5; slightly, 4; moderately, 3; quite a bit, 2; extremely, 1.4	1
5. Overall satisfaction with postoperative chest appearance	Extremely satisfied, 5; very satisfied, 4; satisfied, 3; dissatisfied, 2; 4 very dissatisfied, 1	5
6. Bothered by surgical scars	Not at all, 5; very slightly, 4; slightly, 3; a little bit, 2; a lot, 1.4	5
7. Pain during hospital stay	None, 5; very mild, 4; mild, 3; moderate/severe, 2; very severe, 1.2	1
8. Chest pain with bar in place	None, 5; very mild, 4; mild, 3; moderate/severe, 2; very severe 1.2	3.5
9. Pain (now) without bar	None, 5; very mild, 4; mild, 4; moderate/severe, 2; very severe, 1.5	5
10. Overall satisfaction (cosmetic and functional) with final result	Extremely satisfied, 5; very satisfied, 4; satisfied, 3; dissatisfied, 2; 4 very dissatisfied, 1	5
11. Going back, would you have the operation again?	Yes, 10; unsure, 5; no, 0.10	10

of cardiac or great vessels perforation occurred during the study period.

The mean length of hospital stay was 8.2 +/− 2.5. Early and late postoperative complications are summarized in Table 3. Notable early complications included pneumothorax in 1 patient (2.8%), requiring chest tube, pleural effusion (3 patients; 8.3 %) that did not require drainage, noninfectious seroma in 2 patients (5.5%) requiring would drainage, and pneumonia in 1 patient (2.8%).

Bar displacement occurred in 1 patient (2.8 %). Bar removal was anticipated because of persistent pericarditis in 1 patient (2.8%).

Of the 36 patients treated and included in the study, 63.8% patients (23 out of 36) responded to the survey. Most patients stated general health and exercise tolerance were improved after the operation: 95.6% (22 of 23 patients) noted a subjective improvement in their chest wall appearance, with improvement in social interaction reported by 86.9% of responders (20 of 23). High levels of overall satisfaction were reported after Nuss repair, with 95.6% of patients (22 of 23) being either satisfied or very satisfied. Overall, 87.0% of patients (20 of 23) stated they would have the operation again (Table 4).

No correlation was found between patients' postoperative satisfaction and preoperative HI (question 5: $p = 0.56$, r-square = 0.02; question 10: $p = 0.25$, r-square = 0.09; question 11: $p = 0.52$, r-square = 0.03).

No association was found between the presence of symptoms before surgical correction and postoperative satisfaction (question 5: $p = 0.21$; question 10: $p = 0.42$; question 11: $p = 0.36$).

Everyday life influence of postoperative appearance correlated well with question 5 (rs = −0.60, $p < 0.01$) and question 10 ($rs = -0.45$, $p = 0.03$), while no statistically significant association was found with question 11 (rs = −0.39, $p = 0.07$).

Table 3 Postoperative complications of patients ($n = 36$)

Pneumothorax	
Not required chest tube	-
Required chest tube	**1 (2.8%)**
Pleural effusion	
Not required chest tube	**3 (8.3%)**
Required chest tube	**-**
Pericarditis	**1 (2.8%)**
Wound seroma	
Not required drainage	**2 (55%)**
Required drainage	**-**
Bar displacement	**1 (2.8%)**
Early bar removal	**1 (2.8%)**
Allergy	-

Discussion

The initial report by Lawson et al. [2] highlighted the positive impact that the Nuss correction of PE could have on the physical and physiological well-being of a pediatric population.

Roberts et al. [7] have also shown that this operation can also have an important impact on the patients' perception of quality of life. The authors administered a questionnaire before surgery and 6 to 12 months after repair by the Nuss procedure, that it is to say in a short- and midterm. A multicenter study on a very large population (247 responders) found that surgical repair of PE can significantly improve the body image difficulties and limitation on physical activity experienced by patients [4]. Krasopoulos et al. confirmed a similar impact on young male adults, always in the short term [8]. Like in the abovementioned studies, questionnaires were administered only 1 year after surgical repair of PE, leaving the doubt on the long-term patient satisfaction and quality of life after MIRPE. We believe results can be skewed by the relatively short period of time passed between surgery and the survey, considering the interviewed/

Table 4 Impact of operation on various aspects of quality of life

Change after operation	**Much worse**	**Worse**	**The same**	**Better**	**Much better**
Health in general	-	-	4%	22%	74%
Exercise capacity	-	-	22%	13%	65%
Satisfaction with appearance	-	4%	13%	22%	61%
Difference in negative social impact	**Not at all**	**Slightly**	**Moderately**	**Much**	**Extremely**
Preoperative negative social impact	-	13%	13%	26%	39%
Postoperative negative social impact	4%	4%	5%	17%	70%
Pain after operation	**None**	**Mild**	**Moderate**	**Severe**	**Very severe**
Pain in hospital	-	8	13%	17%	61%
Pain at follow-up	69%	13%	9	9%	-

surveyed patients are still children. In fact, having passed such a short period of time and being the bar still positioned, the worse aspects of the surgical correction process (hospitalization, postoperative pain, immobilization, ICU, etc. could influence the patient's opinion on the surgery. Other authors explored satisfaction and quality of life after PE repair in the long run but only on adult patients operated during adulthood. Sacco Casamassima et al. in 2006 and Hanna et al. in 2013 reported favorable long-term results achieved with the Nuss procedure in adults with a high rate of patient satisfaction, significant improvement in self-image, and excellent midterm cosmetic result. It is to remember that previous studies found an increased rate of early postoperative complications in adults compared with children and adolescents, the more frequent complications being pulmonary embolism (specific of the adult population), pleural effusion, and bar displacement. Furthermore, because of difference in costal cartilage flexibility, there is a biomechanical disadvantage in performing the Nuss procedure in adults [9]. We then thought to interview patients operated during childhood but become adults, with a minimum of a 5-year follow-up. So, we administered the questionnaire introduced by Krasopoulos et al. [10] and modified by Sacco Casamassima et al. [9].

In our long-term assessment evaluation of patient satisfaction and quality-of-life improvement after surgery, respondents to the survey expressed high levels of satisfaction in terms of self-image and quality of life.

Moderate-severe prolonged chest pain was the main complaint in this study. Nevertheless, being most patients satisfied with the final cosmetic result, pain perception did not interfere with overall satisfaction and their willingness to undergo the operation again. Improvement in cosmetic appearance and health in general is translated in most patients in an improvement in social life.

Moreover, we found the high overall satisfaction after surgery was not correlated with the deformity severity and the presence of physical symptoms before correction. This could be an expression of deformity experienced transversely in the same way regardless of its severity. The discomfort is not only common to the whole population but such as to make everyone equally happy regardless of whether they were symptomatic or not. Perhaps preoperative psychological discomfort affects patients more than physical symptoms do.

Finally, there is a statically significant correlation between incidence of postoperative appearance on everyday life and postoperative satisfaction: the more this aspect affects their lives, the less the satisfaction is and vice versa.

Anecdotally, apart from the questionnaire and the shown results, we were enthused because of the tremendously positive comments received in the e-mails and phone calls to the patients. It was not rare to hear how much better the patients are feeling about themselves, how much better their exercise tolerance is, how much better they are behaving, and how much better they are doing in sports. However, a common complaint, apart from the pain, was the impossibility of practicing sports during the bar stay.

The degree of postoperative pain after MIRPE has been shown to be the overriding factor in the patient's perception of the quality of the postoperative course. This result was found on an adult population as well by Casamassima et al. [9]. Postoperative pain may require a more aggressive analgesic regimen. Currently, the two most common pain control strategies following the Nuss procedure are thoracic epidural infusion and intravenous patient-controlled analgesia (PCA). In addition, a variety of multimodal regimens utilizing chest wall indwelling catheter infusions and local or regional nerve blocks have recently been proposed to lessen pain and decrease perioperative opioid use. Great effort must be spent in order to improve pain management after surgery; with this goal, studies should be encouraged in order to compare different analgesic regimens. Perioperative pain management has always been a prerogative of anesthesiologists, but great interest has been evoked by the introduction of intraoperative intercostal nerve cryoablation, which temporarily ablates peripheral nerves, during the Nuss procedure [11–13]. Cryoanalgesia is a promising adjunct in the care of pediatric patients undergoing MIRPE and would make the surgeon part of the pain management equipe.

Conclusion

This study is limited by its retrospective nature and by the small sample size, limiting our ability to draw generalized conclusion. Furthermore, not all patients who underwent the Nuss repair responded to the survey. Despite the limitation, this study is the first one that investigated long-term patient satisfaction and quality of life on adults who undertook MIRPE when children/during childhood.

Concluding MIRPE is a safe and well-tolerated procedure. Favorable long-term results can be achieved with the Nuss procedure in pediatric patients.

Abbreviations

MIRPE: Minimally invasive repair of pectus excavatum; PE: Pectus excavatum; ICU: Pediatric intensive care unit.

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Authors' contributions

Conceptualization, NDS, ET, MDM, and GP; methodology, NDS, ET, GP, and MDM; software, GP; validation, NDS and ML; formal analysis, GP; investigation,

NDS, ET, and MDM; resources, NDS, GP, and ET; data curation, NDS, ET, MDM, and GP; writing—original draft preparation, NDS; writing—review and editing, NDS, ET, and ML; supervision, ML; and project administration, NDS. The authors read and approved the final manuscript.

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Availability of data and materials
Please contact author for data requests.

Declarations

Ethics approval and consent to participate
The study was conducted according to the guidelines of the Declaration of Helsinki and approved by the Ethics Committee of IRCCS Sant'Orsola Malpighi University Hospital (CHPED-02-20-pe). Informed consent was obtained from all subjects involved in the study.

Consent for publication
Written informed consent has been obtained from the patient(s) to publish this paper.

Competing interests
The authors declare that they have no competing interests.

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