Minimally Invasive Repair of Pectus Excavatum

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Chapter 8

Sports activity in adolescents in the Netherlands with a pectus excavatum; the impact of surgery

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Abstract

Introduction

Pectus excavatum (PE) is the most common chest wall deformity in adolescents. The main complaint is cosmetic, but many patients also complain about exertional dyspnea. This may lead to the patient seeking surgery of the thoracic wall deformity (TWD). The assumption is that both, appearance and physical complaints will have a negative effect on being able or wanting to engage in sport activity.

Materials and Methods

In December 2011 a prospective registration of sport activity in pectus excavatum patients started. Sport activity was assessed using questionnaires (CHQ, SF-36 and PEEQ). Measurements were taken before corrective surgery (pre-operatively) and 12 months post-operatively.

Results

127 patients have been included. The number of patients who were active in sports pre-operatively and after 12 months remained steady. The type of sport activity, individual sport or team sport showed no significant change. The CHQ showed that physical activity caused fewer complaints ($p < 0.001$). The PEEQ showed a decrease in difficulties with sports activity performance after 12 months ($p< 0.001$).

Conclusions

Twelve months after surgical correction of PE there was no significant increase in the number of patients performing sport activities. However there was a significant decrease of complaints or difficulties during sport compared to pre-operatively.

Introduction

Pectus excavatum (PE) is the most common chest wall deformity and affects predominantly males, with an incidence of approximately 1 in 400 live born infants [1]. The most important complaint is cosmesis although a substantial part, up to 65% of the patients also complains of physical impairments, especially shortness of breath during exercise[2]. PE can be surgically corrected by two procedures, the Nuss procedure and the Ravitch procedure. Both procedures have been reported to give good cosmetic results [3]. There is an increasing number of studies which report physical improvement after correction of PE in both adolescents and adult patients [4,5,6]. Although patients with a deep PE (high Haller Index) were expected to have more pre-operative physical complaints than those with more superficial PE deformities, this has not been confirmed [7]. There is a positive effect of surgery reported on the general physical development measured by weight and length and computed tomographic index of the chest [8]. However a high Haller Index was associated with pulmonary dysfunction but no cardiac dysfunction [9].

Both physical and cosmetic complaints may lead to a decreased participation in sport activity by not being able to or not wanting to engage, especially in adolescents who are vulnerable to peer pressure. Being ashamed of their appearance they may be inclined to hide their chest and refrain from sports activities [10]. Less sport activity itself may lead to lower physical capability [11]. We hypothesized that adolescents with a pectus excavatum would have lower sport activity than peers and that participating in sports activities would increase after surgical correction.

Methods

In December 2011 a prospective multicenter registration started of sport activities in PE patients. Sport activity was assessed using questionnaires. Measurements were taken when patients visited the outpatient clinic to discuss the possibility of corrective surgery (T1) and 1 year post-operatively (T2), at this time the Nuss bar was still in place.

Patients

Since December 2011, patients who were referred with a PE to the outpatient clinic of one of the five participating centers were asked to participate in this

study. Exclusion criteria were age younger than 12 years and patients or parents with insufficient knowledge of the Dutch language in reading or writing. Patients with Marfan's syndrome or other associated connective tissue diseases were allowed to participate into the study.

Patients sixteen years or older gave informed consent. For patients under the age of sixteen informed consent was obtained from both patients and their parents. The medical ethics committee approved the study.

Questionnaires

Patients were divided into three groups based upon age: younger than 16 years, 16 - 18 years and older than 18 years of age. Appropriated questionnaires were used for each age group.

The Dutch version of the Child Health Questionnaire (CHQ-87) was used in patients younger than the age of 18 years. The CHQ-87 is a generic Quality of Life assessment tool that has good reliability and validity [12]. This questionnaire covers the physical, emotional and social well-being of children. Items are scored using a four to six point Likert scale and converted to a 0 to 100 point continuum, with higher scores indicating a better QoL. Normal values of the Dutch population are available and allowed for comparison with "healthy" children [13].

The Pectus Excavatum Evaluation Questionnaire (PEEQ) was developed to measure physical and psychosocial quality-of-life changes after surgical repair of pectus excavatum. It consists of nine questions asking the children how they feel or act about their bodies. The instrument has high test-retest reliability (Rho > 0.6 for all retained questions) [14].

The 36-Item Short Form Health Survey (SF-36) was developed for the Medical Outcomes Study (MOS) and was completed by patients over the age of 16 years. The instrument was designed for purposes of group comparisons involving multiple health dimensions [15].

This study focused on question 2.1 of the CHQ, question 5 and 10 of the PEEQ, question 3A from the SF-36 and a demographic questionnaire.

Statistical Analysis

Data analyses were conducted using IBM SPSS 23 software (SPSS Inc. Chicago, IL, USA). Descriptive statistics for variables of interest in this study are presented as number, means and standard deviations (SD). Comparison between scores at measurement moment T1 and T2 for continuous variables from the study group were calculated using the paired Student T-test. Categorical variables were analyzed using Wilcoxon signed rank test. McNemar's test was applied on paired nominal data in 2 × 2 contingency tables. The cut off point for significance was set at $p < 0.05$.

Results

Between November 2011 and July 2016, 127 patients were included 110 males and 17 females. All patients underwent a Nuss bar procedure. The mean age was 16.2 years (SD 2.2) with 68 patients under the age of 16 years and 15 patients older than 18 years of age at time of surgery. Additional demographic data including pre-operative symptoms can be found in Table 1.

Table 1. Demographic data and pre-operative symptoms

General demographics:	
Mean age	16.2 years (SD 2.2)
Gender	87 % males
Mean Haller index	3.9 (SD 1.4)
Pre-operative symptoms:	
compromised cosmesis and/or	96.9%
pain chest wall	7.9%
exertional dyspnea	25,2%
palpitations	0%

Scores are represented in means (SD) or percentages of the study group.

Pre-operatively 67% of the study group was engaged in sports activities (Table 2). Twelve months post-operatively this percentage had remained almost the same with 65% of the patients after Nuss bar implantation involved in sports activities (p=0.85). Furthermore, the number of hours spend weekly on sports activities did not differ (p= 0.56) in the study group.

Table 2. Sport activity pre-operative (T1) versus 12 months post-operative (T2)

	Sport activity T2			
Sport activity T1	No	yes	Total	p-value
No	29	13	42	
Yes	15	70	85	
Total	44	83	127	0.85

Cross tabulation T1 versus T2 in numbers. McNemar test with p-value.

The type of sport activity, individual or team sport, was similar with respectively p= 0.09 and p= 0.16, between pre-operative and 1 year post-operative (Table 3).

Table 3. Type of sport activity at pre-operative (T1) versus 12 months post-operative (T2)

	Type of sport activity T2				
Type of sport T1	Team sport	Individual sport	No sport	total	p-value
Team sport	35	10	9	54	0.09
Individual sport	6	18	6	30	0.16
No sport	3	11	29	43	1.00
Total	44	39	44	127	

Cross tabulation T1 versus T2 in numbers. McNemar test with p-values.

There was however a significant improvement (Table 4) in our study population one year after PE correction with Nuss bar concerning coping with difficult physical exercise ($p<0.001$), difficult sport activity ($p< 0.001$) and less avoidance of daily activities ($p<0.001$).

Subgroup analyses based on the pre-operative complaints were limited to the primary outcome measure sport activity (table 4). Due to different possible combinations of pre-operative symptoms analyses per symptom were not possible.

Table 4: Comparison between scores on CHQ, PEEQ, SF-36 and demographics for T1 and T2

	T1 versus T2	p-value
CHQ		
2.1 sport activity difficulties	Significant less difficulties with sport activities on T2	<0.001
7.1c How good at sports?	Small improvement in sports at T2	0.213
PEEQ		
5 Avoids daily situations	Significant less avoidance at T2	<0.001
10 Physical exercise Difficulties	Significant less problems with physical exercise at T2	<0.001
RAND 36		
3A Vigorous activities	Small change in limitations with vigorous activity	0.388
Demographic Questionnaires		
3B hours sport a week	4.8 (2.6) versus 4.5 (3.6) hour	0.559
Sub-group analyses for sports-activity based on pre-operative complaints		
Haller index <3.2 versus >3.2	No differences in sports activity	0.70
Exertional dyspnea	No differences in sports activity	0.69
Cosmesis	No differences in sports activity	0.85
Pain	No differences in sports activity	1.0

Scores are represented in means (SD). Concerning scores: a higher score represents improvement. Wilcoxon signed ranked test for paired comparison of ordinal data. Paired t-test for continuous variable. McNemar test for subgroup analyses. The cut off point for significance was $p < 0.05$.

Discussion

The primary goals of pectus correction are improvement of cosmesis and quality of life. Quality of life and especially aspects such as body image, self-esteem and emotional limitations improve significantly after surgery [16]. Increasing exercise capacity, if that was an issue before operation, may be an important additional goal. Not everyone with a pectus excavatum complains of exercise intolerance pre-operatively. Kelly et al. reported approximately 65% of patients complained of exercise intolerance or shortness of breath and approximately 30 percent mentioned chest pains, even if they were not actively engaged in sports [2].

Participation in sports has a number of advantages. Both boys and girls performing individual sport or fitness activities have lower depression and anxiety levels and higher well-being than those who do not participate in sports activities. Boys involved in individual sport/fitness activity and boys involved in team sports showed no significant differences on mental health status, while for girls, there were significant differences in depression, anxiety and wellbeing between those involved in individual sport or fitness activity and those involved in team sports, with team sport associated with higher well-being and lower anxiety and depressive symptoms. Overall, adolescents (30.2 %) reported participation in at least one team sport, while 37.0 % reported participation in an individual sport or fitness activity but not team sports. Boys were more active than girls [17].

Concerning sports participation the Dutch Trend rapport sport and health 2000-2014 showed membership of a sport club in healthy 12-17 year old adolescents over the period 2006-2014 to be stable, with two third having a sport club membership. In 2012-2014, 85% engaged in sport monthly and 74% on a weekly basis [18]. The proportion of persons above 15 years not engaging in any moderate physical activity in the Netherlands in 2005 was 13%. The 2002 proportion of sufficiently active persons above 15 years was 44% [19].

In our cohort 67% of the patients engaged in sports activities, which is equivalent to the average for their age group. In addition no significant increase in patients engaging in sports one year after PE correction was found. Thus, surgery did not lead to an increase in the average hours of sport activity of patients who did engage in sport activity, neither did it change their preference for team sports or individual sport activity.

The effects of surgical correction of PE and their impact on the cardio-pulmonary system is still reason for discussion, whereby different mechanisms by which PE may reduce cardio-pulmonary function are brought forward. There may be an association between PE and diminished static pulmonary function. Correction with Nuss bar implantation procedure results in improvement in chest wall motion; this improvement in the breathing action is accompanied by improvement in pulmonary function tests [20]. Furthermore surgical correction of a deep PE may improve cardiac function by relieving inflow restriction [21]. The issue of improved cardiac function after chest wall reconstruction is also still debated, Guntheroth et al. found no improvement after surgery [22]. Lesbo et al. showed a lower stroke index without heart rate changes in pre-operative pectus

patients compared to healthy age matches [23]. Jeong et al. found that the heart returns to normal position and shape with improved cardiac function after PE correction [24].

The change of pulmonary function after surgery is less contradictory. Maagaard et al. noticed that the forced expiratory volume in 1 second (FEV1) normalized 1 year after PE surgery, and that 3 years after Nuss bar removal, cardiopulmonary function in patient during exercise had normalized and was comparable to peers [25]. Kelly et al. showed significant improvement in lung function measurements during rest and in VO2 max and O2 pulse after operation in pectus excavatum patients with a computed tomography index (Haller Index) >3.2. However, the number of patients in the study who actually had pre – and post-operative measurements was limited to 20 of 327 patients [26].

Neviere et al. suggested that impaired inspiratory muscle strength to generate negative pulmonary pressure in patients with PE and so limiting venous return and cardiac output was the underlying problem of reduced cardio-pulmonary function, and responded well to surgery [27]. Redlinger et al. demonstrated that chest wall motion dysfunction disappeared after surgical correction of the PE with a Nuss bar [28].

Although many studies observe cardio-pulmonary changes after surgical correction of PE, it is still not clear what the effects are on daily life for the patients. The studies give diverse conclusions which do frequently contradict each other.
In this study changes in physiological functioning are solely subjective patient reported outcomes (PROMS). It was not confirmed with exercise tests. It may be that the subjective changes are not reflected when actually testing the patients. However, we feel that the subjective improvement is what counts for these adolescents, because that makes them feel better whether or not it is reproducible in tests.

Another limitation of the study may be that the second questionnaires were administered when the Nuss bar was still in place. It may be that the patient reported outcomes change after removal of the bar. In the studies that tested patients before and after surgery for pectus excavatum only 1 study performed tests before, during (after 1 year), and after Nuss bar treatment (6 months later) [25] and 1 study tested before placement and 3 months after bar removal [29]. Both studies showed improvement in exercise function after surgery comparable to controls and Nuss bar removal did not influence this improvement.

Follow-up in our study is currently 1 year post-surgery. It may be that the reported outcomes change over time. In this study additional measurement moments are planned, however considering the fact that no study has a follow up longer than three years and none of the aforementioned studies has included PROMS we feel it is important to share these results in order to show that pectus excavatum repair leads to subjectively improved physical conditions.

We could argue that in our study the number of patients involved in sport activity did not change one year after surgical PE correction with Nuss bar. However there was a significant decrease in avoidance behavior, increased physical exercise capability and physical activity tolerance after PE correction surgery. The physical capability increased even without regular sport activity. Moreover the study group did not contain large numbers of patient with a deep pectus excavatum (30% Haller Index >3.2), the patients in whom the largest benefit could be expected.

Less avoidance, less difficulties with sports activities and less perceived problems with physical exercise. These findings, suggest an improvement of the cardio-pulmonary function in our patients, although this was not translated into a higher percentage of sport participation which remained equivalent to the percentage of the study group before surgery.

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