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PATIENT BENEFIT FROM MODIFIED NUSS CORRECTION OF PEX EXCAVATUM IS PHYSICAL AS WELL AS COSMETIC

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ABSTRACT

Background: Patient benefit from repair of pectus excavatum has mainly been considered psychological although some evidence of physiological benefit has emerged. The purpose of this study was to investigate lung function, respiratory movement and physical fitness together with the patient's satisfaction with the look of the chest one year following modified Nuss procedure.

Methods: In this prospective cohort follow-up study eighteen boys and young men who underwent modified Nuss procedure participated. Measurements of lung volumes using spirometry, respiratory movements using Respiratory Movement Measuring Instrument, and physical fitness using Aastrand ergometer bicycle test were performed pre- and one year post-operatively. Assessment of participants' opinion of the looks of their chest was made at the same point in time.

Results: Participants mean age was 16±3 years, their mean BMI was 20.8±3.5 kg/m2 and mean Haller Index was 3.9±0.7. Significant changes one year following modified Nuss procedure were found in mean abdominal- and upper thoracic respiratory movements during deep breathing ($p \geq 0.001$, $p \geq 0.05$, respectively), mean physical fitness ($p=0.01$) and mean satisfaction with chest looks in general and without shirt ($p=0.03$, $p=0.001$ respectively).

Conclusion: Increased physical fitness, abdominal- and upper thoracic respiratory movements one year following modified Nuss procedure together with high level of patient satisfaction supports the argument that patients benefit from undergoing the repair is physical as well as cosmetic.

Keywords: Pectus excavatum, modified Nuss procedure, lung function, respiratory movement, physical fitness.

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INTRODUCTION

Pectus excavatum, PE is the most common congenital deformity of the chest with incident reported from 1:800 births to 1:300 births and is more common among boys than girls or 9:1 to 4:1[1-4]. The cause of PE is thought to be hypertrophy of the cartilage between sternum and ribs creating indentation in the chest wall of varying size and shape from few millimetres to several centimeters[5]. The deformity often increases during puberty and late teenage years causing the heart to be compressed, pushed to the left and compressing the left lung. Symptoms associated with PE are fatigue, pain, dyspnoea and increased heart rate and common complaints are decreased exercise tolerance and endurance. Furthermore, the patients often experience psychological distress and negative body image [1-5].

The method most frequently used to calculate the size of the indentation is Haller index, HI, where the inner diameter of thorax were largest is measured on a CT scan and divided by the distance between inner sides of sternum to anterior side of vertebra [1]. Individuals having Haller index 3.2 or more are given the option of surgical correction [6]. The surgical procedure currently used to correct PE is modified Nuss procedure named after the surgeon who first described it in the year 1998. The method involves changing the anterior wall of the chest by inserting a metal bar from one side of the chest, under sternum and out of the other side of the chest and strain the sternum to as normal position as possible [2,7]. The advantages of the modified Nuss procedure over open surgery (Ravich procedure) previously used are shorter surgery time, very small loss of blood and short time to full activity of the patient following surgery [2]. Not so long ago physiological value of the Nuss procedure was questioned and the procedure was even grouped among cosmetic surgeries. Few studies have been published on effects of the modified Nuss procedure, most of them are retrospective and show results on lung volumes, physical endurance and patient satisfaction [1,5,8]. When the present study started only one prospective study was found but since then more prospective studies have been published [9-13]. Results on the effect of modified Nuss procedure on lung volumes have been inconsistent from lung function of PE patient being within normal predicted values pre-operatively and no change 6 months after bar removal [8][8]to decreased lung function pre-operatively compared with reference values and increased function after removal of the bar [5,14] or decreased function both before and after bar placement and removal [15,16]. Initially no study was found investigating respiratory movement of PE patients, but since then two studies, where motion analysis is used, have been published[10,12]. Furthermore only one study was found on physical fitness of PE patients[1] but recently two more studies on cardiopulmonary exercise function have been released [11,13].

Children and teenagers with visible bodily deformation are prone to have negative body image and be teased, which can lead to them avoiding situations where the defect can be seen. Kelly and co-workers showed that problems associated with body image and limitations in physical performance experienced by patients are significantly decreased following corrective surgery of PE [17]. The overall aim of the study was to investigate the physiological benefit of modified Nuss procedure one year post-operatively together with patient's satisfaction with the look of the chest.

METHODOLOGY

We conducted this prospective cohort follow-up study to test our hypothesis that modified Nuss procedure improves lung volumes, respiratory movement and physical fitness of PE patients.

Subjects were 20 consecutive boys and young men undergoing modified Nuss procedure for correcting PE. Chest radiographs and computerized tomography were performed to calculate the HI and outcome of $\geq$3.2 were set as criteria for surgery [2]. Those who met the criteria were scheduled for surgery. Introductory letter was mailed to expected participants when they were called into preparation for surgery. The Data Protection Authority and the Ethical Committee (10-013) approved the study protocol.

At admittance to hospital the participant signed informed consent and a guardian of those younger than 18 years of age. Age, height, weight, body mass index, BMI, body surface area, BSA and participation in sports in school and outside of school was recorded. Then he answered questions adapted from Lawson and co-workers regarding effect of PE on satisfaction with chest appearance, activities and feelings [5]. Bar removal was scheduled 3 years later.

Lung function measurements were performed using Spiro 2000 v. 1.8 (Medikro Oy, Kupio Finland) pre-operatively and one year post-operatively.

Prior to the first measurement of respiratory movements patients answered standardized questions on diseases or trauma that could adversely affect chest movements such as ankylosing spondylitis, neuro-muscular diseases, chest surgery and trauma. Chest wall and abdominal motion-was measured with the Respiratory Movement Measuring Instrument, RMMI (MTT, Arleyni 8, 112 Reykjavik, Iceland), which is based on 6 infrared sensors connected to a PC computer through an analogue to digital converter. The RMMI measures posterior-anterior diameter with a measuring frequency of 21 per second and accuracy is 0.0003 mm and comes with specially designed software. Measurements using the RMMI have been found valid and reliable [18,19].

Respiratory movement and frequency was measured in the supine position with 20° inclination of the head rest. Positioning of the sensors was performed by drawing a vertical line from the medial 1/3 of the clavicle on each side of the thorax to the abdominal wall lateral to the umbilicus. The infrared beams were adjusted to fall on each of these vertical lines including one at the height of the 4^{th} rib, one at the 9th rib, and one lateral to the umbilicus. The sensors were then positioned 10 cm from these landmarks.

Patients were specifically instructed to "neither move nor talk during the measurement." Respiratory movements during quiet and voluntary deep breathing were then recorded during 60 seconds. The instruction to the patients prior to measuring deep breathing was: "When I say breathe deeply now, you breathe in and out as deeply as you can in a slow rhythm and keep doing so until I tell you to breathe normally again." These measurements were repeated one year post-operatively

All participants underwent an Aastrand endurance test on ergometric bicycle (Monark 818 E Ergometric, Monark-Cresent AB, Varberg, Sweden) pre- and one year post-operatively. The result is given in ml/kg/min and grouped into categories from "very poor" to "superior" according to a table of reference values for men aged 15 to over 65 [20].

Statistical analysis: Descriptive statistics were used on demographic data and all measurement results. Wilcoxon Signed Ranks Test was used to analyse significance of changes in measurements.Correlations between HI/FVC, HI/respiratory movements, HI/fitness and BSA/FVC, BSA/ respiratory movements BSA/fitness was calculated using bivariate correlation calculation. SPSS statistical computer program 11th edition was used for calculations. Significance was set at $p \leq 0.05$.

RESULTS

Subjects were 20 consecutive boys and young men undergoing modified Nuss operation for correcting pectus excavatum. Two patients did not turn up for measurements one year after the operation. Demographic data are shown in Table 1. All but 3 were active in some sort of sports, 12 in school and thereof were 8 also active in ports outside the school; 3 went to the gym 2-4 times a week. Demographic data is shown in Table 1.

Table 1: Demographic data of Nuss patients pre-operatively

Pre- operative	Age in Years	Height in cm	Weight in kg	BMI kg/m2	HI	BSA in m2
Mean± StDev	16±3	182±7	71.50± 13.27	20.8± 3.5	3.9± 0.7	1.91± 0.17
Range	12–24	166 – 191	57 -102	17.7-30.5	2.8-5.4	1.65-2.32
Post-operative						
Mean± StDev	17±3	183± 7.3	73.50± 12.10	21.9± 3.5		
Range	13–25	169 – 192	57 -102	17.3-30.5		

BMI = body mass index, BSA = body surface area, HI = Haller index, StDev = standard deviation.

Results of questions on effect of PE on satisfaction with chest looks, activities and wellbeing are shown in Table 2.

Table 2: Results of questions on effect of PE on satisfaction with chest looks, activities and wellbeing before undergoing modified Nuss procedure

Questions	Mean± StDev	Range
Satisfaction with looks in general	8.0±2.6	3-10
Satisfaction how looks without shirt	4.8±3.6	0-10
Spending rest of life as chest is now	4.6±2.7	0-10
Made fun of because of chest	2.2±3.1	0-8
Avoids activity because of chest	3.5±4.0	0-10
Hides chest	4.5±3.9	0-10
Bothered because of the way chest looks	4.1±3.8	0-10
Has trouble exercising	5.2±3.3	0-10
Chest causes shortness of breath	4.7±3.1	0-9

Average lung volumes did not change significantly from pre-operative values, but showed slightly lower figure than pre-operatively Table 3.

Table 3: Mean and standard deviations of lung function measurements among patients undergoing modified Nuss procedure for correction of PE

Pre-operative	FVC% predicted	FEV_1% predicted	PEF % predicted
Mean ± StDev	92.28±13.44	87.06±17.21	79.59±19.32
Post-operative			
Mean ± StDev	88.83±12.59	84.83±17.66	72.89±19.32

FVC = forced vital capacity, FEV_1 = forced expiratory volume in one second, PEF = peak expiratory flow, StDev = standard deviation. All differences in pre-and post-operative measurements were non-significant.

Abdominal and upper thoracic respiratory movements during quiet breathing showed small but not significant increase one year following Nuss procedure Figure 1.

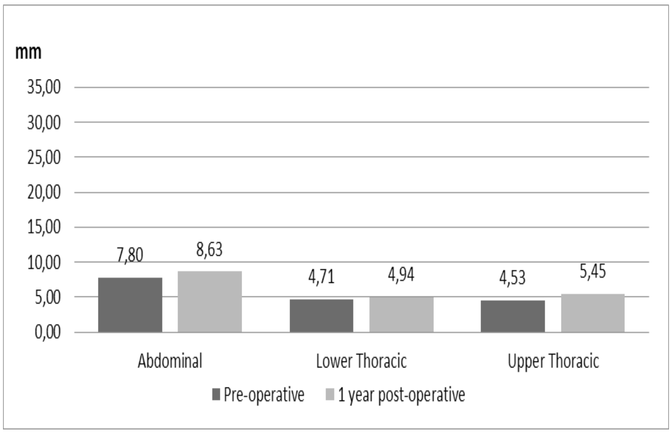

Figure 1: Average respiratory movement during quiet breathing before and one year after modified Nuss procedure for correction of PE.

During voluntary deep breathing these same movements increased significantly one year after modified Nuss procedure for correcting PE Figure 2.

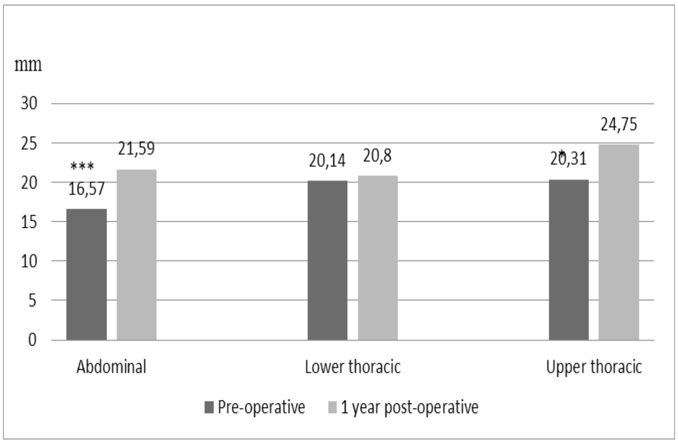

Figure 2: Respiratory movement during voluntary deep breathing before and one year after modified Nuss procedure for correction of PE. *** p≥0.001, *p≥0.05.

Pre-operative respiratory movement pattern was thoracic dominant as average abdominal movement was 18.41% less than average lower thoracic movement and 17.73% less than average upper thoracic movement. One year post-operatively differences between measuring sites were not significant.

Mean fitness pre-operatively was 42.94±7.15 ml/kg/minand was 46.44±7.49 ml/kg/minone year post-operatively, which was significant (p=0.01) increase. Grouping of patients into categories of fitness before and one year after surgery is shown in Table 4.

Table 4: Number of patients in each category of fitness before and one year after PE corrective surgery [20].

Time	Very poor	Poor	Fair	Good	Excellent	Superior
Pre-op	1	6	4	5	2	0
1 year	0	2	6	5	4	1

The only correlations found were low (0.58) but significant (p =.01) correlation between HI and FVC and low negative (-0.52) but significant (p =.03) correlation between BSA and 7physical fitness.

Patients satisfaction with "how the chest looks without shirt on" and with "spending rest of life with the chest looking how it looks now" showed highly significant (p=0.001) increase from pre-operative values and satisfaction with "looks in general" also increased significantly (p=0.03). Table 5.

DISCUSSION

The main results from this study are significant increase in abdominal and upper thoracic respiratory movement, physical fitness and patient satisfaction one year after modified Nuss procedure for correcting PE. Changes in lung volumes were not significant.

The average pre-operative lung volumes among our PE patients were decreased compared with reference values, which is in agreement with several previous studies[5, 8, 10, 14] and where unchanged post-operatively as in some studies[8, 21] but in disagreement with others[5, 14]. Three studies investigating lung function before and after bar removal found unchanged lung volumes between measurements [15, 16]. Although average pre-operative lung volumes among our PE patients were lower than predicted, lung volumes for some of them fell within the normal ranges like in the study by Malik et al [1]. Twelve of our 18 patients had FVC within the range for reference values pre-operatively and eight of them were active in school gymnastics plus at least one endurance sport such as soccer and skiing, which could explain their relatively good lung function and high level of physical fitness. Redlinger and co-workers reported 13% decreased FVC and FEV_1 compared to reference values, while in the present study the average decrease was 6% and 13% respectively and PEF was 26% less than reference values [10]. One year post-operatively FVC was 11% less than reference values, FEV_1 15% and PEF 27%. The reason for slightly decreased pre-operative average lung volumes of PE patients is not difficult to comprehend, but it could be expected that increased space within the chest as demonstrated with opthaloscopic plethysmography in two recent studies [10,22] and the significantly increased abdominal and upper thoracic respiratory movement shown in our study, that FVC would increase. PEF is the lung volume measurement that has the closest relationship with muscle strength of the expiratory muscles followed by FEV_1[23]. It is, therefore, tempting to speculate if inadequate strength of the respiratory muscles could be an explanation for reduced forced expiration, as PEF and FEV_1 had decreased further compared to reference values one year following modified Nuss procedure. Placing muscles in changed starting position changes their ability to contract forcefully and it takes time for them to adapt to a new position which can be reduced by specific training. The abdominal muscles are particularly of interest in this respect as they are important respiratory muscles in forceful in- and expiration as in FVC, FEV_1 and PEF and are postural muscle as well. It is therefore important to investigate this aspect of the results of correcting PE to find out if specific training of these muscles post-operatively is warranted to prevent future back problems.

Respiratory movement during normal quiet breathing in the supine position both pre- and post-operatively was abdominal dominant as found among none symptomatic individuals (Figure 1) [24]. However, during voluntary deep breathing type of respiratory movement was thoracic dominant pre-operatively opposite to what is found among none symptomatic males, Figure 2 [24]. The explanation for smaller pre-operative abdominal range of motion during voluntary deep breathing could be the shape of the lower thorax at the origin of the diaphragm placing it in unfavourable position compared with the position after repair of the defect. One study was found reporting pre-operative respiratory movement in volume increase during deep breathing, where percent increase was greater at the abdominal and lower thoracic compartment than at the upper thoracic compartment. The authors suggested that patients were compensating for reduced thoracic movement by increasing the abdominal movement [10]. One year post-operatively type of respiratory movement at the level of the bar was unchanged but abdominal and upper thoracic movement had increased significantly ($p \geq 0.001$, $p \geq 0.05$ respectively), Figure 2. The unchanged range of lower thoracic respiratory movements likely to be due to the bar hindering movement at this level of the thorax and the increased abdominal movement could be caused by changed position of the origin of the diaphragm. The upper-thoracic movement however increased the most, which is in agreement with two recent studies[12, 13]. The diaphragm is normally the principal and least energy consuming/most efficient respiratory muscle. The increased diaphragmatic excursion could therefore increase their physical fitness even more when the diaphragm has adapted to the new position and consequently uses less of the total energy consumption during physical exertion.

Physical fitness among our patients measured with Aastrand ergometer bicycle test increased significantly ($p=0.01$) one year after modified Nuss procedure compared with pre-operative values. Before correction of their PE 61% had fair or below physical fitness, but 44% one year post-operatively and mean increase in physical fitness was 3.5 ml/kg/min. This improvement might be considered small, but it should be remembered that the patients were not training fitness during the hospital stay and for some time thereafter, therefore, they had to regain pre-operative fitness level before they could start improving it. The explanation for their improved fitness cannot be attributed to increased lung volumes but we speculate that the relieved pressure to the heart might result in increased stroke volume which is in line with two recent studies showing improved cardiopulmonary function during vigorous exercise [21,25]. Although many of our patients were still growing during the year between measurements (mean height 1 cm and mean gain in weight 2 kg) it cannot explain the increase in physical fitness as measurement are made by kg body weight.

The only correlations found in this study were low (0.58) but significant ($p = .01$) correlation between HI and FVC and low negative (-0.52) but significant ($p = .03$) correlation between BSA and physical fitness.

Patients´ satisfaction with chest look one year after modified Nuss procedure in our study improved significantly ($p = .001$) from pre-operative values. Previous studies have also found excellent patient satisfaction and excellent cosmetic results [5,26]. Kang et al compared patient satisfaction among those operated with Nuss and Ravitch procedure and found that the long-term satisfaction was determined by a complete correction without recurrence or need for re-intervention rather than by the operation type [27]. The fact that the upper range of scores for satisfaction with looks was 10 indicates that some of the patients' motive for undergoing the procedure was not entirely cosmetic but the prospect to enhance their performances in their sport.

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Limitations of the study and further studies

The limitations of the study are mainly a small sample size and the lack of age and gender matched non-symptomatic subjects as age of subjects in the reference values available is 20 to 60 years. Measuring posterior – anterior diameter increase of the chest wall only can also be considered limitation as the movements of the ribs are three dimensional. However, in a study on chest expansion measured with tape measure and modified Breysky pelvimeter Moll and Wright stated that "the increase in posterior- anterior and lateral diameter during inspiration is very similar and therefore use of either one is suitable in clinical trials" [28].

Further studies are needed on respiratory movements among PE patients and to investigate the effect of PE and PE repair on posture and postural-, respiratory muscles.

CONCLUSION

Increased physical fitness, abdominal- and upper thoracic respiratory movements one year following modified Nuss procedure together with high level of patient satisfaction found in this study supports the argument that patients benefit from undergoing the repair is physical as well as cosmetic.

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