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ORIGINAL ARTICLE

Long term cardiopulmonary effects of closed repair of pectus excavatum

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Abstract The closed or ''Nuss'' repair of pectus excavatum is widely accepted for correction of moderate to severe deformities. Patients typically report significant subjective improvements in pulmonary symptoms, and short and medium term evaluations (up to 2 years with the bar in place) suggest modest improvement to cardiac function but a decrease in pulmonary function. This study examined the effects at 3 months post-bar removal of closed repair of pectus on pulmonary function, exercise tolerance and cardiac function. Patients were followed prospectively after initial evaluation for operation. All patients underwent preoperative and post-bar removal evaluation with CT scan, complete pulmonary function and exercise testing to anaerobic threshold, as well as echocardiogram. Twenty-six patients have completed the follow up protocol. Preoperative CT index was 4.5 ± 1.3, average age at operation was 13.2 years, and average tanner stage was 3.5 ± 0.5. At 3 months or greater follow-up post-bar removal, patients reported an improvement in subjective ability to exercise and appearance ($P < 0.05$ by wilcoxin matched pairs). Objective measures of FEV1, total lung capacity, diffusing lung capacity, O_2 pulse, VO_{2max}, and respiratory quotient all showed significant improvement compared to preoperative values, while normalized values of cardiac index at rest did not (All values normalized for height and age, comparisons $P < 0.05$ by student's paired t test). These results demonstrate a sustained improvement in cardiopulmonary function after bar removal following closed repair of pectus excavatum. These findings contrast with results from previous studies following the open procedure, or with the closed procedure at earlier time points; the long term physiological effects of closed repair of pectus excavatum include improved aerobic capacity, likely through a combination of pulmonary and cardiac effects.

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Methods

Over the past decade there has been a surge of interest in the correction of chest wall defects, which has been brought about by the innovations introduced by Nuss [1]. This technique utilizes a substernal bar to achieve a closed, minimally invasive repair of excavatum defects. With this increase in the number of patients presenting for chest wall repair, there is ongoing controversy regarding the cardiopulmonary effects of these interventions. While there has long been a perception of improved ability to exercise following repair by patients and the surgeons caring for them [2–4], the evidence relating to the open technique suggested in fact there is a deterioration in pulmonary function with some improvement in cardiovascular function following repair [5–9]. In previous reports we have described our cohort of patients, and showed that the effects of the closed (Nuss) repair paralleled those of the previous open repair [10]. Initially there is a deterioration of pulmonary function, but an immediate improvement in cardiovascular function, along with a self reported improvement in exercise tolerance. The present study extends the previous findings, reporting the outcomes of patients followed to 3 months after bar removal. We hypothesized that over time the previously noted deterioration in pulmonary function would reverse, and that with long-term follow up the patients would demonstrate an improvement in pulmonary function as measured by FEV1 and FVC, improved cardiac function with improved cardiac index, and improved exercise tolerance as manifest by improved VO_{2max}.

Materials and methods

All patients referred for evaluation of pectus excavatum were seen through a single centralized clinic. Patients were interviewed and a complete history and physical done. Those patients felt to be surgical candidates were screened prospectively using a standardized protocol of CT imaging, pulmonary function studies including exercise tolerance, and echocardiogram. The patients and their families were approached and consent obtained for study enrollment, which consisted of collection of the initial patient data, as well as ongoing monitoring with repeated pulmonary function studies and echocardiogram done 3, 18 months and post-bar insertion, and then at 3 months following bar removal. This protocol was approved by the conjoined Health Ethics Review Board of the Calgary Health Region. Patients with co-existing cardio-pulmonary disease or an underlying causative syndrome such as Marfan's were excluded.

The present study details the results from the assessments done pre operation and 3 months after bar removal. At each time point, patients were interviewed, and asked to rate both their appearance and their self reported ability to exercise using a 5 point Likert scale [10]. Data are presented from the initial preoperative and 3 month post-bar removal self reported impressions of ability to exercise (0 = unable to run any distance without shortness of breath, 5 = able to run the length of a soccer field and keep up with peers) and appearance (0 = chest looks ''terrible'', 5 = chest looks ''normal, and would go swimming without a shirt (males) or in a bikini top (females)''. Patient demographic data were obtained during the initial hospital evaluation. The cardio pulmonary evaluation protocol included a limited computed CT scan done at the point of maximal depression, which allowed for calculation of the Haller index [11] as measure of severity. Pulmonary function studies were done in conjunction with progressive exercise testing using a Sensormedics Vmax metabolic cart with a cycle odometer or treadmill. Continuous measurement of inspired and expired oxygen and carbon dioxide output were done using in-line sensors. A 10 or 25 W protocol was used depending on the patients age (Sensormedics Corp., Yorba Linda, CA, USA) [12]. Subjects exercised to the point of fatigue, or demonstrated increase in CO_2 output without further increase in O_2 uptake (anaerobic threshold). Subjects were encouraged to continue exercise until this threshold was reached. During testing continuous non-invasive monitoring of heart rate, blood pressure and pulse oximetry was monitored using a Marquette ECG system (Marquette Electronics, Marquette, WI, USA).

Resting cardiac function was evaluation using standard supine echocardiography (Agilent 5500 ultrasound machine, Agilent Technologies Andover, MA, USA). Stroke volume and cardiac output and cardiac indices were computed using standard methods [13].

Surgical techniques

Surgical techniques and early postoperative complications are as previously reported [10]. In brief, the closed repair as originally described by Nuss was successful in all cases. In these 26 patients, 22 had one bar placed and 4 patients had two bars placed. Statistical comparisons were done using a student's paired t test on data normalized as to patients age, height and weight. Nonparametric values (appearance self rating, exercise self rating, and Tanner stage) were compared using Wilcoxin sum rank test. A P value < 0.05 was considered significant.

Results

A total of 26 patients completed the study protocol between the years 1999 and August of 2006. Demographic and descriptive data are presented in Table 1. As previously reported, the procedures were well tolerated with an average hospital stay of 5.0 ± 1.4 days for the bar insertion, and less than 24 h for bar removal. There were three perioperative problems related to bar movement which required reoperation. The results for these patients are included in the present studies, there were no other patients requiring reoperation during the study period. The patients reported a subjective improvement in both appearance, and their perceived ability to exercise, which did not change over the 2 year follow-up period or with bar removal [10]. Pre and postoperative pulmonary func-

Table 1 Demographics and self-rating of outcomes

	Preoperative	Post bar removal
N	26	26
Age (years)	13.2 ± 2.1	15.8 ± 0.5
Weight (kg)	49.0 ± 10.3	61.0 ± 11.0
Height (cm)	163 ± 11	172.3 ± 7.0
Tanner scale	3.5 ± 0.5	4.0 ± 0.2
Pectus severity (Haller index)	4.5 ± 1.3	
Appearance	2.9 ± 1.7	4.5 ± 1.1*
Exercise tolerance	1.9 ± 0.9	4.5 ± 1.2*

Data: mean ± SD

Self-rating determined at clinic visit

Appearance: 0 (looks terrible) to 5 (looks normal)

Exercise tolerance: 0 (unable to run 10 m) to 5 (able to run soccer field as fast as peers) (10)

*$P < 0.05$ by Wilcoxon matched pairs test

Table 2 Pulmonary effects of closed repair of pectus excavatum

	Preoperative	Post bar removal
N	26	26
FVC (l)	3.28 ± 1.04	3.77 ± 1.08
FVC (% expected)	89.5 ± 18.6	92.4 ± 20.6
FEV1 (l)	2.67 ± 0.81	3.2 ± 0.91
FEV1 (% expected)	78.4 ± 16.0	84.2 ± 18.4*
TLC (l)	4.47 ± 1.22	5.20 ± 0.96*
TLC (% expected)	95.3 ± 16.0	99.3 ± 13.7*
VC (l)	3.34 ± 1.05	3.81 ± 1.1
VC (% expected)	91.1 ± 18.6	93.3 ± 21.1
Diffusing lung capacity (ml/mm Hg/min as % expected)	97.8 ± 13.6	103.2 ± 14.9*

Data: mean ± SD

Values of pulmonary function studies as measured at rest. % values are normalized for age, height and weight (12)

*$P < 0.05$ by students paired *t* test

tion study results are included in Table 2. Comparative statistics are done only on the normative data, because of the interval growth of the patients. These showed significant improvements in FEV1, total lung capacity and the diffusion capacity (Table 2). Exercise testing to the anaerobic threshold was successful in all patients preoperatively, and in 23 of 26 patients postoperation (Table 3). There was a significant improvement in VO_{2max}, and the respiratory quotient, but a decrease in maximal minute volume of breathing (Table 3). The total work done at exercise, and the anaerobic threshold did not change over the study period (data not shown). Specific testing of cardiac function at rest showed that stroke volume was improved over the study period; however, when this was controlled for height and weight and expressed as cardiac index, the change was not significant (Table 4).

Table 3 Exercise tolerance after closed repair of pectus excavatum

	Preoperative	Post bar removal
N	26	23[a]
VO_{2max} (l/min)	1.70 ± 0.45	2.08 ± 0.45*
VO_{2max} (% predicted)	70.8 ± 11	76.6 ± 10.7*
VO_2/kg (ml/kg/min)	34.1 ± 6.1	35.5 ± 68
Minute volume breathing at maximal exercise (l/min)	67.0 ± 15.3	71.0 ± 14.3
Minute volume breathing at maximal exercise (as % expected)	71.7 ± 19.5	64.5 ± 16.2*
Heart rate (beats/min)	178 ± 11	176 ± 12
Heart rate (% expected)	86 ± 6	86 ± 5
Respiratory quotient	1.07 ± 0.07	1.10 ± 0.07*
O_2 pulse (% predicted)	77.1 ± 9.5	82.5 ± 9.2*

Data: mean ± SD

Comparisons done on normalized data using students paired *t* test

*$P = 0.05$ versus preoperative values

[a] Patients not exercising to anaerobic threshold were excluded

Table 4 Cardiac function after closed repair of pectus excavatum

	Preoperative	Post bar removal
N	26	26
Stroke volume (ml)	69.0 ± 21.2	83.9 ± 24.5*
Cardiac output (l/min)	4.66 ± 1.39	5.38 ± 1.48*
Cardiac index (l/m^2)	3.08 ± 0.75	3.29 ± 0.82

Data: mean ± SD

Discussion

These results show for the first time that at long-term follow-up after closed repair of pectus excavatum with the Nuss procedure there is a sustained improvement in both pulmonary function and aerobic exercise tolerance. However, in contrast to our findings in the early postoperation phase, we did not see a sustained increase in cardiac index. Improvements in cardiac output and stroke volume were noted, but these may have been associated with the general growth of the patients. Overall, these results do support our hypothesis that operative correction of the chest shape abnormality in patients with non-syndromic pectus excavatum improves pulmonary function, and the capacity for aerobic exercise.

These results corroborate the growing body of evidence which shows that patients who have a moderate to severe pectus excavatum have an associated decrease in pulmonary function. Preoperatively, our patients exhibited a reduction in lung volumes, primarily in the dynamic components of FEV1 and FVC (Table 2), which are very similar to those reported in previous studies of patients from a variety of study groups [6, 8, 10, 14, 15]. This contradicts the long established dogma in the general pediatric literature that pectus excavatum is not associated with significant pulmonary changes [16]. However, in contrast to previous reports detailing the long term effects of closed repair of pectus excavatum, and our own studies done at earlier time points with the bar in place, the present cohort of patients exhibited an improvement in FEV1 and TLC following repair and removal of the bar. This is similar to the results reported by Lawson et al. [15], and suggests that the closed technique results in a significantly different long term outcome from the classical Ravitch technique. While the results are arguably modest, they do represent a normalization of function that would not be expected to occur without intervention [17].

The effects of correction of the pectus defect provide interesting insights into the components of pulmonary function. The action of the thoracic cavity to act as a bellows to create air movement comes from the combined activity of the extrinsic musculature of the chest wall, and the diaphragm. However, in situations where the chest wall confirmation is abnormal, the extrinsic musculature effect may be compromised, and diaphragmatic efficiency is likely reduced [18]. Conceptually then correction of the chest wall abnormality should improve this by improving both the intrinsic and diaphragmatic function. That this occurs is evidenced by the selective improvement of the most dynamic component of pulmonary function, FEV1. However, total lung capacity and diffusion capacity (measures of the combined action of the chest wall and diaphragm, and the efficiency of alveolar ventilation and perfusion matching, respectively) were also improved.

The observed improvement in VO_{2max} is an important observation; this is in agreement with the findings after open repair, and the results of a recent meta-analysis of both open and closed repairs [6–8]. It is difficult to delineate the specific factors which may have led to this improvement; however, we would argue that it is due to a combination of improved efficiency of ventilation, and increased stroke volume (and secondarily cardiac output) at exercise. The perceived shortness of breath with intense exercise, which patients describe preoperatively is likely due to the reduced stroke volume at load caused by the pectus compression [6, 17, 19]. In normal subjects, maximal exercise capacity (and the shortness of breath which is the factor usually given as limiting by subjects) is directly correlated with oxygen delivery, which in turn relates primarily to cardiac output [20, 21]. In our subjects the resting measures of cardiac output did increase, but when normalized for growth, this was not significant. Nonetheless, an improvement in stroke volume and a resultant increase in cardiac output is likely a component of the observed increase in O_2 pulse and VO_{2max} seen in our patients. A limitation of our study is that we did not control for, or document the degree of physical activity of the patients. However, the VO_{2max}/kg, which is a measure of physical conditioning (Table 3) [21], did not change, and so sustained conditioning from an increase in exercise following surgical correction is not likely the explanation for the improved exercise capacity. Overall, the improvement in subjective ability to exercise following correction preceded any significant improvement in ventilatory capacity, or VO_{2max}, but was associated with the early improvement in cardiac output, and index [10, 19].

Interestingly, following pectus repair, despite the increased capacity for oxygen utilization, there was a reduction in minute ventilation at exercise (Table 3). This suggests that with correction of the pectus defect, there is an increase in the efficiency of the chest wall. At exercise a significant proportion of energy is used to provide respiratory air exchange, with the majority of this energy being used by the diaphragm [22]. An improvement in efficiency of the chest wall/diaphragm unit would free up oxygen and energy available for general musculoskeletal use. The increase in the *r*/*q* ratio, at maximal loading, may also be explained by a switch to oxygen utilization by the peripheral muscle mass, depending on the energy substrates used [20, 22] (Table 4).

The long term effects of pectus correction on cardiac function were somewhat surprising (Table 3). Although there was an increase in the absolute stroke volume, and cardiac output, this could not be separated from the general increase due to the growth of the patients. We suspect these findings may different from our previous observation due to a wider range of pectus severity in the present cohort, including more patients with moderate pectus. Our earlier findings were perhaps somewhat biased by our initial operative candidates who tended to have a more severe pectus index. As well, the findings reflect the inadequacies of static testing of cardiac function. To truly determine the effects of pectus repair on cardiac output at exercise requires a direct measurement of cardiac function. O_2 pulse, as a surrogate measure of stroke volume at exercise, did increase (6, 7).

In summary the results of these studies show that following the closed repair of pectus excavatum in moderate to severe defects that there is a significant improvement in pulmonary function, and aerobic exercise tolerance. In this cohort of typical pectus patients, we suggest that the observed benefits in cardiopulmonary function are due to a combination of improved chest wall efficiency and cardiac output at exercise. The importance of this, in the present age of awareness of fitness in general, is self evident. These results certainly support the evolution of practice that is being observed, with increased use of the closed repair for pectus correction in moderate to severe cases. However, they specifically do not apply to more limited cases, and do not completely define the effects on cardiac function; further, more direct study is required.

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