# Exercise improvement after pectus excavatum repair is not related to chest wall function<sup>+</sup>

Johanna Acosta<sup>a</sup>, Amy Bradley<sup>b</sup>, Vinesh Raja<sup>a</sup>, Andrea Aliverti<sup>c</sup>, Saurin Badiyani<sup>a</sup>, Andrea Motta<sup>c</sup>, Stefano Moriconi<sup>c</sup>, Kim Parker<sup>b</sup>, Pala Rajesh<sup>b</sup> and Babu Naidu<sup>d,\*</sup>

<sup>a</sup> International Digital Laboratory, University of Warwick, Coventry, UK

<sup>b</sup> Academic Department of Anaesthesia, Critical Care, Pain and Resuscitation, Heart of England NSH Foundation Trust, Birmingham, UK

<sup>c</sup> Dipartimento di Elettronica, Informazione e Bioingegneria, Politecnico di Milano, Milan, Italy

<sup>d</sup> Birmingham Medical School, University of Birmingham, Birmingham, UK

\* Corresponding author. International Digital Laboratory, University of Warwick, Coventry CV4 7AL, UK. Tel: +44-24-76523523; fax: +44-121-4242200; e-mail: b.naidu@bham.ac.uk (B. Naidu).

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## INTRODUCTION

Pectus excavatum is a congenital deformity of the chest wall with an incidence of one in 300–400 Caucasian male births [1] and is more common than Down's syndrome [2]. Patients presenting with pectus excavatum may seek surgical intervention for a variety of reasons, including dyspnoea on mild exertion, chest discomfort, tachycardia, poor body image and its resulting psychological effect [3].

Recent meta-analyses [4, 5] conclude that there is a global improvement in cardiopulmonary function after pectus repair. The mechanism of improvement may involve both cardiac and pulmonary effects. However, the effects of pectus correction on dynamic chest wall function during exercise are unknown. This is of particular relevance as procedures of perceived low clinical value are being rationed both in the UK and globally because of

the financial climate. Thereby proving benefit from surgery and mechanism may be important in deciding who will benefit from surgery and thus preserve this practice which has benefited thousands of young men and women in the past.

Therefore, we study whether dynamic chest wall function improves after Nuss surgical correction in pectus excavatum patients.

## MATERIALS AND METHODS

#### **Participants**

Optoelectronic plethysmography (OEP; BTS, Milan, Italy), a validated method of measuring total and regional chest wall volumes [6–9], was used in 7 male patients over the age of 16 years (mean 19.7  $\pm$  2.7 years) with severe pectus excavatum (Haller index from 3.4 to 8) who were scheduled for a surgical correction called the Nuss procedure at a regional thoracic surgery unit. The Haller index is the ratio between the transverse thoracic diameter and

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the lowest distance from the sternum to the vertebra obtained from the same level [10]. Computed tomography of the chest was used to calculate the Haller index for each patient. None of the participants had previously undergone chest correction surgery. Surgery was performed under general anaesthesia and patients returned to a dedicated thoracic surgery ward postoperatively. Postoperative pain control was achieved by continuous thoracic epidural analgesia, or intrathecal morphine, followed by systemic opioids (oral or intravenous patient-controlled administration). The choice of analgesic technique was made by the anaesthetist. All patients had a daily physiotherapy programme from the first postoperative day, which included deep breathing exercises, supported coughing and mobilization. There is no formal rehabilitation programme for these patients, but for the first 3 months following surgery, they are discouraged from heavy exercise and for 6 months from contact sports.

#### Study design

A prospective observational study was undertaken in a regional thoracic centre on young male patients. The research study was approved by the Local Research Ethics Committee. Informed consent was obtained from all the participants involved.

Chest wall motion was measured in participants preoperatively, postoperatively as an inpatient, then at 4–6 weeks and at 6 months. Data collected pre- and postoperatively included participants age, height, weight, smoking status, pain score and any relevant past medical history. Spirometry was performed (Carefusion Microlab) prior to capture of chest wall motion measurements according to the ATS/ERS guidelines [11].

Acquisition of chest wall motion was performed using the SMART suite software (BTS). OEP cameras were calibrated each day prior to the tests. The acquisition/procedure required 79 hemispherical and 10 spherical reflective markers to be placed onto the participants' chest wall and back using a bioadhesive hypoallergenic tape [6, 12]. Standard placement of markers was done according to Cala *et al.* [6].

The OEP acquisition protocol included that observations of heart rate, oxygen saturations and blood pressure were recorded after 1 min quiet breathing and after the patient reached 80% of the maximum heart rate (220 - age × 80/100). During the OEP acquisition, rib cage and abdominal volumes were recorded by eight infrared cameras operating at 60 Hz. The OEP system tracked the displacement of the markers during vital capacity, quiet breathing and cycle ergometry up to 80% of the maximum heart rate with increasing load each minute maintaining 60 revolutions per minute. From total and compartmental (rib cage and abdominal) chest wall volumes acquired by OEP, the following parameters were obtained: tidal volume (VT), as the total chest wall volume variation, total and compartmental volumes at end of expiration (Vee) and end of inspiration (Vei), respiratory rate (RR) and minute ventilation (VT × RR). These parameters were analysed at quiet breathing, 100% maximum exercise (time when 80% of the maximum heart rate is reached, also called exercising time) and 50% maximum exercise [(exercise starting time - exercising time)/2].

#### Statistical analysis

Data are presented as mean [standard deviation (SD)] or median (95% confidence intervals) unless otherwise stated. Paired or

unpaired Student's *t*-tests were used as appropriate to compare data pre- and postoperatively for each patient for normally distributed data and Wilcoxon rank-sum test for non-normally distributed continuous data. All statistical analyses were performed using the SPSS version 19 statistical software (IBM, NY, USA).

A statistical significance of P < 0.05 was used for all analyses, with appropriate corrections for multiple comparisons.

## RESULTS

Table 1 summarizes demographic data and breathing pattern for the preoperative pectus population involved in the study. They were a young male group with severe pectus excavatum (PE) deformities who underwent surgical correction for cosmetic reasons. No patient reported limitation in exercise capacity or any other significant respiratory symptoms. During OEP measurements, all patients achieved the target of 80% of the maximum heart rate during cycle ergometry. However, they had lower than predicted spirometric values in terms of both forced expiratory volume in 1 s (FEV<sub>1</sub>) and forced vital capacity (FVC; Table 1).

Decrease in respiratory function postoperatively is evidenced by changes in spirometry as shown in Fig. 1. Although there is considerable recovery in FEV<sub>1</sub> and FVC postoperatively, these measurements are still lower than baseline by  $4.2 \pm 13$  and  $7.8 \pm 4\%$ , respectively, at 6 months postoperatively.

According to data obtained from OEP, VT decreased significantly immediately post-operation and partially recovered after 6 months. This is most marked during maximum exercise. VT at 50% of maximal exercise was significantly reduced from  $1.8 \pm 0.7$  preoperatively to  $1 \pm 0.2 \mid (P = 0.013)$  postoperatively (4–6 days). This immediate postoperative reduction in VT recovered from  $1 \pm 0.2$  to  $1.6 \pm 0.4 \mid (P = 0.001)$  6 months pre- and postoperatively. However, VT did not recover to preoperative measurements 6 months after the operation at any stage of the test (quiet breathing and exercise).

VT at rest does not significantly differ pre- and postoperatively, but during exercise VT initially drops postoperatively in all compartments (Fig. 2A) and persists in the rib cage compartment at 6 months. Sub-compartment analysis reflects a decrease in the contribution of the combined upper and lower rib cage immediately postoperatively (Fig. 2B), while the abdominal contribution to VT is preserved (Fig. 2C).

**Table 1:** Baseline patient and breathing pattern (N = 7)

	Mean	SD
Age	19.7	2.7
Weight (kg)	71.8	8.9
Height (cm)	184.4	6.8
BMI	21.2	3.1
FEV <sub>1</sub> (I)	4.1	0.7
%FEV <sub>1</sub> predicted	87.5	12.2
FVC (I)	4.7	0.9
%FVC predicted	83.4	15
FEV <sub>1</sub> /FVC (%)	88.2	6.9
Haller index	5.4	1.8

BMI: body mass index; FEV<sub>1</sub>: forced expiratory volume in 1 s; FVC: forced vital capacity; SD: standard deviation.



Figure 1: Pre- and postoperative spirometric measures. \*\*P < 0.01. Box representing first and third quartiles, band inside the median and whiskers the range.



Figure 2: Compartmental VT at maximum exercise. \*P < 0.05, \*\*P < 0.01, \*\*\*P < 0.001.

Figure 3 shows that the postoperative reduction in VT is due to the reduction in end-inspiratory volume of the total chest wall that progressively tends to come back to the preoperative values. This reduction is totally due to the rib cage compartment whose end-inspiratory volumes are significantly reduced (P = 0.026), indicating a decreased mobility of the rib cage 6 months postoperatively. Conversely, the abdominal compartment maintains a similar behaviour pre- and at all phases postoperatively.

Surgery has an effect also on absolute total and compartmental volumes. Overall, there is a trend towards increase in thoracic volumes at rest and during exercise postoperatively. End-expiratory absolute volume of the rib cage at maximum exercise increased from  $16 \pm 2.5$  preoperatively to  $17.2 \pm 3.4$  l (P = 0.020) postoperatively. Conversely, the end-expiratory volume of abdominal compartment tends to decrease (from  $6.1 \pm 1.9$  to  $5.1 \pm 1.7$  l, P = 0.046 at 50% maximum exercise).

After pectus surgery, there is an increase in the RR at quiet breathing and during exercise (Table 2). This is particularly marked few days after operation at 50% maximum exercise  $(23.5 \pm 9.3 - 31.4 \pm 11.9, P = 0.023)$ . After 6 months, the RR has decreased, but it is still higher than baseline. Elevated breathing is a mechanism to compensate for the drop in VT by reducing its impact on minute ventilation, especially during exercise (Table 2).

A significant difference in exercise tolerance for all the patients was found 6 months after surgical correction. As seen in Fig. 4, the exercising time to reach 80% of the maximum heart rate increased

from  $288 \pm 89$  preoperatively to  $415 \pm 85$  s 6 months postoperatively (*P* = 0.009), which represents a rise of 44%.

#### DISCUSSION

Our study shows that lung function as measured by OEP and spirometry is reduced early postoperatively in PE patients, which is in keeping with other published results. Sigalet *et al.* [13] have reported a significant reduction 3 months after pectus correction (FVC:  $92 \pm 22-72 \pm 15\%$ , P < 0.05; FEV<sub>1</sub>:  $79 \pm 22-68 \pm 19\%$  and VO<sub>2</sub>:  $69 \pm 24-56 \pm 25\%$ , P < 0.05). Similarly, Borowitz *et al.* [14] have reported a decrease in predicted values of FVC, FEV<sub>1</sub> and total lung capacity, as a part of a study evaluating patients 6 months and 1 year postoperatively, when the bar is still in place.

Interestingly, improvements in lung function, cardiac output and exercise tolerance have been reported several years after correction by the same authors. Sigalet *et al.* [15] reported improvements after bar removal (FEV<sub>1</sub> 78 ± 16–84 ± 18%, P < 0.05). This finding was confirmed by Lawson *et al.* [16] who also showed small improvements in spirometric measurements specifically in patients above the age of 11 years after 3 years from bar removal.

However, not all studies have reported benefits for patients late after PE correction. Castellani *et al.* [17] concluded from a study involving 59 patients that pectus repair is not followed by improvements in lung function or exercise performance (FVC went from 91 ± 14 to  $88 \pm 13\%$  and VO<sub>2</sub> remained around  $49 \pm 7$  ml/kg pre- and postoperative). Similarly, Bawazir *et al.* [18] state that pulmonary function remained under baseline values after bar removal after evaluation of 11 patients. Unlike Castellani *et al.* [17], Bawazir *et al.* [18] did report improvement in exercise tolerance 21 months after repair, which are in agreement with our findings.

Several other studies using the same technology (OEP) have looked at dynamic chest wall function. When PE patients were



Figure 3: Change in end-inspiratory volume (Vei) (open symbols) and end-expiratory volume (Vee) (closed symbols) at quiet breathing (QB) and exercise, pre- and postoperatively.

compared with normal controls with deep breathing manoeuvres, PE patients had a significant increase in the volume within the abdominal rib cage compartment (PE: 0.8 I, C: 0.6 I, P < 0.01) during maximal respiration, though total chest volumes were the same in both groups [10]. Six months following pectus repair, there is a modest increase in all three chest compartment volumes and improvement in respiratory excursion of the deepest part of the sternum [19]. In another study [20] looking at differences in 19 PE vs controls during maximal voluntary ventilation (MVV), PE subjects exhibited a mild restrictive lung defect, avoiding dynamic hyperinflation by setting operational volumes at values lower than controls. The same authors reported on the group of PE patients who underwent pectus repair and have shown that at rest there is a modest increase in functional residual capacity (FRC) as determined by body plethysmography, which is associated with an increase mainly in the volume of rib cage pulmonary compartment [21]. We did not measure FRC in our study, but we did not show an increase in the volumes of any compartment. But importantly, by looking at the temporal change in values, we have better described the postoperative and early outcomes following surgery. By studying patients during maximum exercise, we have also contributed to a better understanding of the dynamic changes in chest wall function and this is a meaningful clinical relevance.

These two groups [20, 21] did not find any correlation with the Haller index of severity and any differences in chest wall kinematics or lung function. The Haller index is limited in its definition of severity, because it inadequately describes different morphologies.

We conclude from our data that early improvements in exercise capacity after Nuss repair are not due to improvement in dynamic chest wall function. After 6 months of correction, we found that during exercise VT, end-expiratory volume, minute ventilation and RR have not returned to baseline preoperative measurements. Additionally, we have shown a significant decrease which is being partly compensated for by higher contribution of the abdomen in expiratory motion. In spite of this, our study has shown a 44% improvement in exercise capacity. Therefore, this early improvement

	QB		Exercise			
			50%		100%	
	Mean ± SD	P-value	Mean ± SD	P-value	Mean ± SD	P-value
Respiratory rate						
Preoperative	16.1 ± 3.3		23.5 ± 9.3		30.8 ± 7.2	
Postoperative inpatient	20.0 ± 6.4	NS	31.4 ± 11.9	0.023	35.1 ± 14.5	NS
4–6 weeks	16.3 ± 6.0	NS	27.5 ± 10.9	NS	35.2 ± 8.7	0.043
6 months	17.8 ± 7.2	NS	26.5 ± 4.5	NS	32.3 ± 5.3	NS
Tidal volume						
Preoperative	0.9 ± 0.4		1.8 ± 0.7		2.4 ± 0.6	
Postoperative inpatient	0.7 ± 0.2	NS	1.0 ± 0.2	0.012	1.6 ± 0.4	< 0.001
4–6 weeks	0.8 ± 0.2	NS	1.6 ± 0.7	NS	2.0 ± 0.6	NS
6 months	0.8 ± 0.4	NS	1.6 ± 0.4	NS	1.9 ± 0.4	0.045
Minute ventilation						
Preoperative	13.9 ± 5.7		37.5 ± 14.4		73.6 ± 21.7	
Postoperative inpatient	13.4 ± 3.8	NS	29.7 ± 11.1	NS	51.9 ± 18.5	0.004
4–6 weeks	12.5 ± 4.1	NS	42.5 ± 23.6	NS	69.4 ± 20.6	NS
6 months	12.3 ± 6.0	NS	43.0 ± 13.2	NS	61.8 ± 15.9	NS

Table 2: Mean and SD of RR (breaths/min), VT (I) and minute ventilation (I) during quiet breathing (QB) and exercise, pre- and postoperatively



Figure 4: Total time of exercising until reaching 80% of the maximum heart rate. \*\*P < 0.01.

is likely to be related to changes in cardiac function. There is strong evidence to suggest that there is improvement in cardiac function early after pectus repair [5, 22, 23].

There is a need to determine the physiological benefits and mechanisms of pectus repair to avoid such procedures being tagged 'of low clinical value'. Further research is warranted in assessing dynamic lung function in a larger number of pectus patients evaluating the longer term sequel after bar removal. It will also be important to relate surrogate markers of function to patient reported outcomes such as breathlessness. Non-invasive dynamic chest wall motion analysis has inherent benefits in the future, and it may identify subgroups of patients who respond differently to surgery. Regardless, this study has important clinical implications in patients' counselling as a part of the process of surgical recovery by warning patients about potential restriction in rib cage motion and early postoperative loss of function.

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