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The questionable benefit of pectus excavatum repair on cardiopulmonary function: a prospective study

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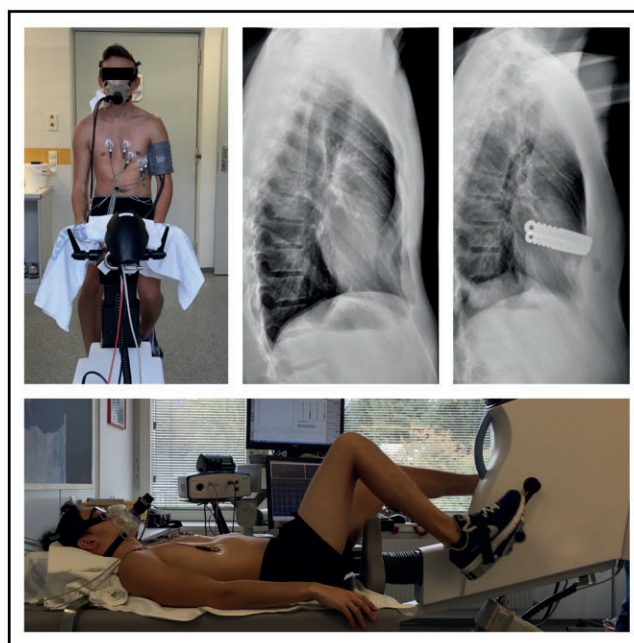
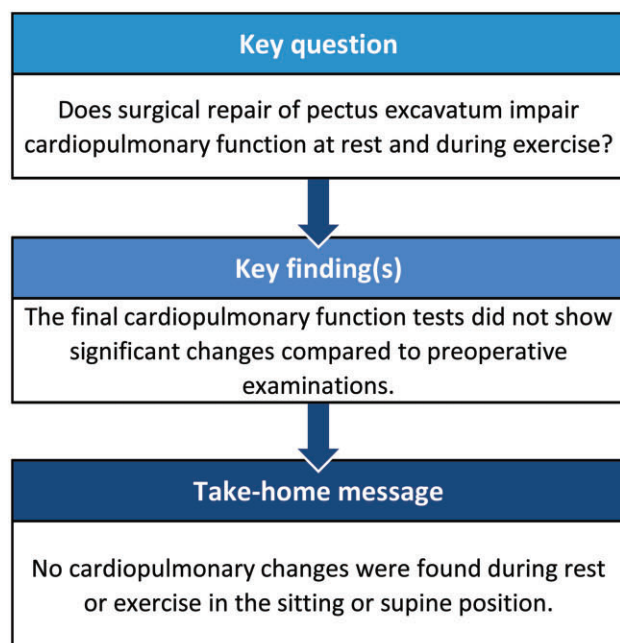
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Abstract

OBJECTIVES: Since the introduction of the minimally invasive technique for repair of pectus excavatum (MIRPE), increasing numbers of patients are presenting for surgery. However, controversy remains regarding cardiopulmonary outcomes of surgical repair. Therefore, the aim of our prospective study was to investigate cardiopulmonary function, at rest and during exercise before surgery, first after MIRPE and then after pectus bar removal.

METHODS: Forty-seven patients were enrolled in a prospective, open-label, single-arm, single-centre clinical trial (Impact of Surgical Treatments of Thoracic Deformation on Cardiopulmonary Function) [NCT02163265] between July 2013 and November 2019. All patients underwent a modified MIRPE technique for surgical correction of pectus excavatum (PE), called Minor Open Videoendoscopically Assisted Repair of Pectus Excavatum. The patients underwent pre- and postoperative chest X-ray, three-dimensional volume-rendering computer

tomography thorax imaging, cardiopulmonary function tests at rest and during stepwise cycle spiroergometry (sitting and supine position) and Doppler echocardiography. Daily physical activity questionnaires were also completed.

RESULTS: The study was completed by 19 patients (15 males, 4 females), aged 13.9–19.6 years at the time of surgery. The surgical patient follow-up was 5.7 ± 7.9 months after pectus bar removal. No significant differences in cardiopulmonary and exercise parameters were seen after placement of the intrathoracic bar, or after pectus bar removal, compared to presurgery.

CONCLUSIONS: Our findings indicate that surgical correction of PE does not impair cardiopulmonary function at rest or during exercise. Therefore, no adverse effects on exercise performance should be expected from surgical treatment of PE via the modified MIRPE technique.

Clinical trial registration number: clinicaltrials.gov [ClinicalTrials.gov number, NCT02163265].

Keywords: Pectus excavatum • Thoracoplasty • Cardiopulmonary function • Spiroergometry • Echocardiography • Exercise

ABBREVIATIONS

CT	Computed tomography
FEV1	Forced expiratory volume in 1 s
FVC	Forced vital capacity
HI	Haller index
IPAQ	International Physical Activity Questionnaire
MIRPE	Minimally invasive technique for repair of pectus excavatum
MOVARPE	Minor Open Videoendoscopically Assisted Repair of Pectus Excavatum
PE	Pectus excavatum
PFT	Pulmonary function test
% pred	Percentage of predicted values
VO ₂	Oxygen consumption
VO _{2max}	Maximal oxygen uptake

INTRODUCTION

Pectus excavatum (PE) is the most common congenital anterior chest wall deformity, with a reported incidence of 0.1–0.3%. Males are 3–5 times more likely to be affected than females [1, 2]. PE is caused by excessive growth of the costal cartilage, resulting in a concave anterior chest wall [3]. The deformities frequently present not only as an aesthetic disturbance but also in association with mild limitation of physical activity, obstructive pulmonary mechanics, slight dyspnoea and abnormal cardiac physiology [4]. For decades, surgical correction of PE was performed primarily for cosmetic and psychological reasons, without proven postsurgical benefits on cardiopulmonary deficits [1, 5]. Surgical repair of PE using a retrosternal metal bar (Nuss procedure) is a widely accepted operative procedure for the correction of PE [5, 6].

Restrictions in pulmonary functions at rest, such as low lung volume, lower airway obstruction, elevated residual volume, and a decrease in spirometry parameters, were reported [7]. However, no differences in dynamic lung function parameters compared to healthy controls were described [8–10]. Depending on the mechanical compression, diverse cardiac abnormalities may manifest [11]. Regarding impairments in submaximal and maximal exercise performance, there is controversy in the literature, which is mainly attributable to heterogeneity in test protocols, exercise parameters and PE severity, as well as small, statistically underpowered samples [12].

Studies on cardiopulmonary function after surgical repair also show conflicting results regarding improvements (or not) in exercise performance after surgery [12]. Regarding the first stage of minimally invasive technique for repair of pectus excavatum (MIRPE) (insertion of the metal bar), exercise data are inconsistent, with increases in maximal oxygen uptake (VO_{2max}) [13], no changes [14], and even reductions all being reported [15].

Given the lack of evidence of effects of PE surgery (with a retrosternally implanted pectus bar) on cardiopulmonary outcome variables, the goals of this study were to: (i) investigate cardiopulmonary function at rest, before surgical correction of PE and after pectus bar removal and (ii) investigate submaximal and peak exercise parameters in the sitting and supine positions, before and after surgical correction. Based on the above evidence, we hypothesized that there would be an improvement in cardiopulmonary function and that physical well-being and perceived exercise ability would improve, at the various follow-up exams. To the best of our knowledge, no prospective study of only PE patients has performed a pre- and postoperative comparison of cardiopulmonary function and exercise.

PATIENTS AND METHODS

Study design

All patients with PE deformity presenting at the Department of Plastic, Reconstructive and Aesthetic Surgery of the Medical University Innsbruck were invited to participate in this prospective, observational, single-arm, single-centre, non-randomized clinical study. Those with subjective impairments (cardiopulmonary, aesthetic or psychological/aesthetic) were screened using a standardized protocol, which was approved by the Institutional Review Board of Medical University Innsbruck (Approval Number AN4741; date of approval: 27 July 2012). The study was registered at clinicaltrials.gov [NCT02163265] [16]. Inclusion criteria were: (i) aged 10–50 years; (ii) male or female with PE deformities; and (iii) minor-to-severe PE. The age limits for the patients in our study were set according to the recommendations of the Ethics Committee. In addition, our patient selection criteria were based on the previously made clinical decision to operate only on PE patients aged at least 13 years (due to body maturation), but not older than 20 years (due to public health insurance restrictions).

The exclusion criteria were: (i) Poland's syndrome; (ii) previous repair of PE using any technique; (iii) previous thoracic surgery;



Figure 1: Clinical presentation of a male patient (aged 16 years) who had undergone Minor Open Videoendoscopically Assisted Repair of Pectus Excavatum surgery and pectus bar removal: presurgery (left picture) and at 8 months post-surgery (right picture).

(iv) congenital heart disease; (v) history of major anaesthetic risk factors (e.g. malignant hyperthermia or pregnancy); and (vi) contraindication for cycle spirometry.

Patients or their parents gave written consent after receiving oral and written information. The study period was from July 2013 to November 2019. Baseline demographic data were collected, including medical history, physical examination results, a standardized photo-series, thorax calliper measurement of the transversal and sagittal chest diameters (MedXpert Company GmbH, Heitersheim, Germany) and chest X-ray. Three-dimensional volume-rendering computed tomography was performed to determine the Haller index (HI), which is the most commonly used index of PE severity [17]. Pulmonary function tests (PFT), electrocardiography and transthoracic Doppler-echocardiography at rest followed by cycle spirometry (in both the sitting and supine positions) until exhaustion were performed at the Institute of Sports Medicine, Alpine Medicine and Health Tourism (ISAG) prior to surgery. Patients completed standardized questionnaires on the quality of life and physical activity in daily life.

The Minor Open Videoendoscopically Assisted Repair of Pectus Excavatum (MOVARPE) technique (a modification of the MIRPE technique) for PE repair was carried out as previously described by the authors [18–20]. Patients indicated for invasive thoracoplasty underwent surgical repair using this semi-open procedure, performed by the same surgeon (Anton H. Schwabegger) in all cases.

Six months after surgery (retrosternally placed pectus bar), the clinical examination, thorax calliper measurements, PFTs, cycle spirometry until exhaustion (sitting and supine position), transthoracic Doppler echocardiography and questionnaires were repeated. The pectus bar was removed after 26.5 ± 6.6 months during a second-stage operation. At least 3 months

after pectus bar removal, the final study visit was scheduled and included all preoperative measurements and tests except for chest X-ray (Fig. 1).

Forty-seven patients (38 males, 9 females) met the inclusion criteria and provided informed consent. Out of these, 3 patients were aged between 10 and 14 years, 26 were aged between 14 and 18 years and 18 were aged 18 years or older at the time of inclusion in this study. All 47 patients underwent initial clinical evaluations; 28 patients were subsequently excluded (23 males, 5 females) for different reasons, as described in Table 4.

Surgical technique

The initial surgical steps for the MOVARPE technique are the same as for the MIRPE technique, as described by Nuss, with bilateral incisions made at each of the anterior midaxillary lines [5]. An additional parasternal incision is then made, allowing horizontal sternum osteotomy and relaxing chondrotomies to be performed [20]. Under video-assisted thoracoscopy, a round tunnelizer creates a substernal tunnel at the deepest point of the PE [21]. In cases of severe PE deformity, and in patients with rigid habitus of the thorax, multiple paramedian chondrotomies are performed via transcutaneous stab incisions, to improve elasticity of the thorax. After the pectus bar (Biomet Microfixation Pectus Bar; Biomet Microfixation, Jacksonville, FL, USA) has been introduced and rotated, the convexity of the bar lifts the depressed chest wall. The bar wings are fixed with circumcostal double-armed 0 polydioxanone sutures to prevent bar displacement [19]. Then, the wound is closed. Thoracic peridural analgesia utilizing bupivacaine is applied for postoperative pain management for 2–4 days in almost all cases. Non-steroidal anti-inflammatory drugs are also used if necessary. Postoperative X-rays are taken in the

postanaesthesia care unit to rule out pneumothorax and verify that the bar is implanted in the correct location. Bars are left in place for ~1–2 years for the MOVARPE technique. Bars are subsequently removed in a second-stage operation under general anaesthesia.

Testing equipment and procedure

Pulmonary function test. To assess pulmonary function, patients fully inhaled and fully exhaled in a spirometer (GE Medical Systems Inc., Milwaukee, WI, USA) as quickly as possible. The forced vital capacity (FVC; liters), forced expiratory volume in 1 s (FEV1; L) and Tiffeneau index (FEV1/FVC; %) were used for further analysis. To exclude any possible influence of growth in the adolescent patients, these parameters were also calculated as percentages relative to of an age-adjusted reference population [data given as % of predicted values (% pred) according to Quanjer *et al.* [22]]. Spirometry data (FVC, FEV1 and FEV1/FVC) are provided as a percentage of the predicted values; values above and below 100% indicate better and worse pulmonary function, respectively, compared to the respective standard cohort.

Echocardiography. Transthoracic Doppler echocardiography (two-dimensional, M-mode) was performed to exclude mitral valve regurgitation and other possible structural abnormalities, as well as to evaluate fractional shortening, left ventricular systolic and diastolic diameter, ejection fraction and right ventricular diastolic diameter (ACUSON SC2000; Siemens, Munich, Germany).

Cycle spiroergometry. Patients performed stepwise cycle ergometry in sitting (Lode B.V., Groningen, Netherlands) and supine positions (ebike L; Ergoline GmbH, Bitz, Germany) until exhaustion, or until meeting objective criteria for exercise termination [23]. Oxygen consumption (VO_2) was measured via gas analyses (Vyntus CPX; Care Fusion, Hoechst, Germany). The test started at 25 W, with the workload increasing by 25 W every 120 s. The same protocol was used for both spiroergometries. For lactate analysis, capillary blood (20 μl) was collected from the hyperaemic ear lobe every 120 s at the end of each workload step (Biosen S-Line Lab+; EKF Diagnostics, Barleben, Germany). From the relation between blood lactate at the end of each step and the corresponding power output (W/kg) the relative mean power at fixed blood lactate concentrations of 2 and 4 mmol/l was calculated. These parameters can be used as a measure of submaximal exercise performance. The parameters of interest were the relative mean power at 2 and 4 mmol/l lactate (W/kg), peak power (absolute and relative), heart rate (peak), VO_2 peak (ml/kg/min) and peak blood lactate concentration. Due to the moderate exercise capacity of the patients, we did not determine individual lactate thresholds for the evaluation of submaximal exercise performance.

Computed tomography examination

Computed tomography (CT) scans were obtained before and after chest correction to compare objective measurements. Images were acquired as described previously by our group, with radiation doses (CT dose index) of 3.5–6.5 mSv [16]. The HI was calculated from measurements on transverse images ($\text{HI} = \text{A/B}$)

[17]. Then, 3D volume-rendered rotating images of the rib cage were generated, to visualize individual anatomic characteristics and plan cartilage incisions.

Questionnaires

International physical activity questionnaire. The International Physical Activity Questionnaire (IPAQ) is a reliable and valid tool to assess health-related physical activity in populations aged 15–69 years. This questionnaire consists of 27 questions and measures the frequency, duration and intensity of physical activity during the previous week in 4 physical activity domains (work-related physical activity, transport-related physical activity, domestic and gardening activities and leisure-time physical activity) [24].

Physical activity behaviour of adolescents. This questionnaire consists of 13 questions and assesses the health-related physical activity level during the previous week in populations younger than 15 years. The subscales (School Walking, Outdoor Activity Group, Leisure Activity Group, Leisure Activity Alone) are summed to yield a Total Physical Score, reflecting the frequency and duration of activity during 1 week.

Statistical analysis

Data are presented as mean \pm standard deviation for continuous variables, and as percentages or frequencies for categorical variables. Repeated-measures ANOVA, including Bonferroni *post hoc* tests (if applicable), was performed to compare mean pre-, post-surgery 1 and post-surgery 2 values for the PFTs, spiroergometry data (sitting and supine positions), and questionnaire data. Partial eta square was used to quantify effect sizes. Statistical significance was set at $P < 0.05$. All calculations were performed using IBM SPSS statistical software (version 24.0; IBM Corporation, Armonk, NY, USA).

RESULTS

Nineteen patients (15 males and 4 females) completed the full evaluation after bar removal and remained in the study. The baseline characteristics of patients at the cardiopulmonary exercise testing time points are presented in Table 1.

During MOVARPE or bar removal surgery, no intraoperative complications occurred. Early postoperative complications (within the first 6 postoperative weeks) were noted in 3 patients: in 1 patient, a pectus bar dislocation occurred within 48 h after surgery; surgical revision and additional stabilizer implantation were required. Another patient had pneumothorax with a collapsed lung and required placement of a chest tube. Finally, 1 patient was treated with a compression brace for 8 weeks due to reactive sternal protrusion after bar placement. During pectus bar removal surgery, minor aesthetic corrections were performed in 3 patients. No late complications occurred.

Patient age and demographic data are provided in Table 1. The pectus bar was removed 26.5 ± 6.6 months (range: 20–45.6 months) after insertion (Fig. 1). The mean final clinical follow-up time after pectus bar removal was 5.7 ± 7.9 months (range: 2.2–35.15 months).

Table 1: Characteristics of the patients ($n = 19$) at the pre- and post-surgery 1 and 2 cardiopulmonary testing time points and at the time of surgery

Variables	Pre (M \pm SD)	MOVARPE (M \pm SD)	Post 1 (M \pm SD)	Post 2 (M \pm SD)
Age (years)	15.6 \pm 1.6	16.6 \pm 1.7	16.7 \pm 1.7	18.6 \pm 1.9
Body weight (kg)	56.3 \pm 7.5	56.5 \pm 7.2	59.9 \pm 8.5	62.8 \pm 8.6
Body height (cm)	176.3 \pm 7.5	176.6 \pm 7.2	178.1 \pm 7.0	179.6 \pm 7.3

M \pm SD: mean \pm standard deviation; MOVARPE: Minor Open Videoendoscopically Assisted Repair of Pectus Excavatum; Post 1: 7.6 \pm 2.2 months after surgery; Post 2: 5.7 \pm 7.9 months after pectus bar removal; Pre: baseline (before surgical intervention).

Table 2: Data from the pre- and post-surgery 1 and 2 pulmonary function tests ($n = 19$)

Variables	Pre (M \pm SD)	Post 1 (M \pm SD)	Post 2 (M \pm SD)	P-value (Eta Square)
FVC (l)	3.8 \pm 0.9	3.8 \pm 0.7	4.3 \pm 0.7***	<0.001 (0.44)
FVC (%pred) ^a	78.7 \pm 13.4	75.4 \pm 9.8	80.9 \pm 9.9	0.076 (0.32)
FEV1 (l)	3.3 \pm 0.9	3.5 \pm 0.7	3.7 \pm 0.8*	0.001 (0.31)
FEV1 (%pred)	79.3 \pm 16.5	80.1 \pm 11.7	81.7 \pm 15.2	0.63 (0.03)
FEV1/FVC (%) ^a	85.8 \pm 9.0	90.8 \pm 7.4*	85.7 \pm 11.6**	<0.001 (0.31)
FEV1/FVC (%pred) ^a	99.8 \pm 10.6	105.6 \pm 8.8*	99.6 \pm 13.4**	0.001 (0.31)

P-values <0.05 were considered statistically significant.

FEV1: forced expiratory volume in 1 s; FVC: forced vital capacity; M \pm SD: mean \pm standard deviation; % pred: percentage of predictive value according to Quanjer et al. [22]; Post 1: 7.6 \pm 2.2 months after surgery; Post 2: 5.7 \pm 7.9 months after pectus bar removal; Pre: baseline (before surgical intervention);

^aFriedman test.

*Statistically significant compared to 'Pre'.

**Statistically significant compared to 'Post 1'.

Pulmonary function tests

FVC (absolute) and FEV1 (absolute) significantly increased from pre- to post-surgery 2 ($P < 0.001$ and $P = 0.001$, respectively). Both, FEV1/FVC (%) and FEV1/FVC (% pred) changed significantly over time ($P < 0.001$ and $P = 0.001$, respectively), with a peak at post-surgery 2). Further details are presented in Table 2.

Spiroergometry

Our results from both cycle spiroergometry tests (sitting and supine position) are shown in Table 3. In the sitting position, we found a significant reduction in mean power (W/kg) at lactate 4 mmol/l ($P = 0.01$), an improvement in peak power (W) ($P = 0.03$) and an increase in peak lactate ($P = 0.03$). In the supine position, there were a significant change in peak power (W/kg; $P = 0.04$) and an increase in peak power (W; $P = 0.005$) (Table 3).

Echocardiography

The baseline right ventricle end-diastolic diameter and left ventricle end-diastolic diameter were 32.0 \pm 2.57 and 45.0 \pm 4.67 mm, respectively. Neither right ventricle end-diastolic diameter nor left ventricle end-diastolic diameter changed significantly over the study period.

Computed tomography examination and thorax calliper measurements

The mean HI before surgery was 4.2 \pm 1.0 (range: 3.1–6.3) and that after surgery was 2.9 \pm 0.8 (range: 2.1–5.2,

$P < 0.0001$). Operative correction significantly reduces the HI and markedly improves the shape of the entire chest (Figs. 1 and 2).

Sagittal chest diameter, measured by thorax calliper, was 12.6 \pm 1.6 cm (range: 10–15 cm) at initial diagnosis and 17.7 \pm 1.6 cm (range: 15.5–20.5 cm) following thoracoplasty ($P < 0.0001$). The sagittal chest diameter showed no significant change between the post-MOVARPE time point and the final follow-up after pectus bar removal (17.3 \pm 1.8 cm; range: 14–21 cm) ($P = 0.12$). Before surgical intervention, transverse chest diameter was 25.5 \pm 2 cm (range: 22–28 cm) and increased significantly to 26.5 \pm 1.9 cm (range: 23–30 cm) after pectus bar removal ($P = 0.012$).

Physical activity questionnaires

Physical activity, assessed with the IPAQ (and physical activity behaviour of adolescents in the younger population), did not show significant changes during the whole study.

DISCUSSION

This prospective study evaluated PE, and the effect of pectus repair with MOVARPE, before and after bar removal, on cardiopulmonary function at rest and during exercise. Our findings indicate that pectus repair leads to significant improvement of FVC (absolute) and FEV1 (absolute) after bar removal, but no significant changes in relevant parameters were seen during spiroergometry, in either the sitting or supine position. Echocardiographic data and self-reported physical activity levels also remained unchanged.

Table 3: Cycle spirometry results obtained in the sitting and supine positions (n = 19)

Variables	Sitting position			Supine position				
	Pre, M ± SD (range)	Post 1, M ± SD (range)	Post 2, M ± SD (range)	P-value (eta square)	Pre, M ± SD (range)	Post, M ± SD (range)	Post 2, M ± SD (range)	P-value (eta square)
Mean power 2 mmol (W/kg) ^a	1.5 ± 0.4 (0.5; 2.3)	1.3 ± 0.3 (0.8; 2.1)	1.3 ± 0.3 (0.9; 1.8)	0.25 (0.12)	1.4 ± 0.3 (0.6; 1.9)	1.4 ± 0.3 (0.9; 1.9)	1.4 ± 0.3 (0.9; 2.0)	0.95 (0.003)
Mean power 4 mmol (W/kg) ^{a,b}	2.2 ± 0.4 (1.4; 2.8)	2.1 ± 0.3 (1.4; 2.8)	2.0 ± 0.3* (1.4; 2.4)	0.01 (0.21)	2.1 ± 0.3 (1.6; 2.5)	1.9 ± 0.2 (1.5; 2.3)	2.0 ± 0.5 (1.6; 4.0)	0.33 (0.05)
Peak power (W/kg)	3.1 ± 0.5 (2.0; 4.1)	3.0 ± 0.4 (2.0; 3.7)	3.0 ± 0.3 (2.4; 3.5)	0.26 (0.07)	2.5 ± 0.5 (1.6; 3.6)	2.3 ± 0.5* (1.4; 3.4)	2.4 ± 0.5 (1.7; 3.3)	0.04 (0.11)
Peak power (W)	173.9 ± 37.2 (100.0; 238.0)	179.2 ± 36.2 (104.0; 250.0)	187.6 ± 33.9* (132.0; 275.0)	0.03 (0.18)	139.8 ± 34.3 (87.0; 220.0)	141.5 ± 34.7 (81.0; 225.0)	153.3 ± 30.2** (92.0; 200.0)	0.005 (0.25)
VO ₂ peak (ml/kg/min)	41.0 ± 6.2 (26.4; 52.5)	40.9 ± 6.2 (27.6; 54.3)	40.4 ± 4.0 (32.7; 48.9)	0.86 (0.01)	36.2 ± 6.4 (26.5; 49.3)	34.5 ± 6.1 (22.6; 47.3)	34.4 ± 5.6 (24.3; 45.4)	0.6 (0.1)
HR _{peak} (bpm)	192.5 ± 7.81 (171.0; 206.0)	192.2 ± 8.5 (164.0; 203.0)	188.5 ± 9.8 (173.0; 206.0)	0.06 (0.14)	169.1 ± 18.6 (129.0; 193.0)	165.3 ± 19.6 (129.0; 196.0)	169.4 ± 18.9 (141; 196)	0.77 (0.05)
Lactate _{peak} (mmol/l)	8.7 ± 2.5 (4.3; 13.7)	9.6 ± 2.7* (4.0; 13.9)	9.8 ± 2.1 (6.3; 13.5)	0.03 (0.17)	6.6 ± 3.0 (2.2; 12.5)	6.7 ± 2.9 (2.3; 12.4)	7.4 ± 2.8 (3.8; 13.1)	0.24 (0.08)

P-values <0.05 were considered statistically significant.

HR_{peak}: heart rate (peak); M ± SD: mean ± standard deviation; Pre: baseline (before surgical intervention), Post 1: 7.6 ± 2.2 months after surgery; Post 2: 5.7 ± 7.9 months after pectus bar removal; VO₂: oxygen consumption.

^aFriedman test sitting position.

^bn = 14.

*Statistically significant compared to 'Pre'.

**Statistically significant compared to 'Post 1'.

Surgery and pulmonary function tests

Although not all patients with PE have subnormal PFTs, deficits in PE patients have been reported, albeit to varying degrees depending on factors like age and the severity of PE deformities. For example, Kelly *et al.* [25] reported lower FVC, FEV1 and forced expiratory flow between 25% and 75% of vital capacity values for in >1500 adult and paediatric patients with severe PE. Changes after surgical repair are inconsistent and may depend on the surgical method itself [12]. Focusing on the MIRPE method, studies either performed PFT after bar insertion and bar removal, or only after bar removal. In small cohorts, Borowitz *et al.* [14] and Udholm *et al.* [10] observed no changes before and after replacement of the intrathoracic bar. After a temporary decrease, FVC returned to pre-operative values following bar removal [8]. In contrast, others showed improvements in a variety of PFT parameters [13, 26–28].

In contrast to the literature on MIRPE, we found a significant increase in FEV1/FVC (% and % pred) in our patients with the bar implanted, and a return to presurgery values after bar removal. Thus, we conclude that bar insertion does not worsen and may even improve spirometric parameters. In addition, at least in our setting, temporary, transient pulmonary restriction by the metal bar and negative PFT outcomes can be ruled out.

Surgery and cardiopulmonary exercise tests

Since parameters at peak exercise are influenced by motivation and, as during daily life, maximum exercise performance plays a subordinate role, we recorded the mean power at the fixed thresholds of 2 and 4 mmol/l blood lactate concentration for further interpretation of the surgical outcomes in relation to cardio-respiratory performance. We observed no changes in any cycle spirometry parameters, except the relative mean power at 4 mmol/l and absolute peak power (small reduction) in the sitting position, and the absolute peak power in the supine position (small increase). No statistically significant exercise parameter was of clinical relevance since the absolute changes were only minor.

A similar lack of effect on VO_{2max} with the pectus bar in situ was described by Borowitz *et al.* [14] and Castellani *et al.* [8]. In contrast, in children with severe PE, maximum exercise capacity increased after bar placement [29], while Sigalet *et al.* [15] reported an increase in VO_{2max} after bar replacement. However, 3 years after surgery and bar removal, the lower exercise performance (compared to controls) had disappeared [26].

Training deficiency may be the factor most responsible for the lower exercise capacity of PE patients. Since many patients perceive their thoracic deformity to be a substantial physical deficit, leading to psychological problems due to aesthetic concerns, low motivation may be an additional barrier to regular sports participation. It has been proposed that better exercise performance following PE correction surgery could be purely due to greater motivation for regular sports and improved stamina [12]. However, our IPAQ and KAGAS questionnaire data indicated no changes in habitual daily physical activity (frequency or duration). Studies documenting daily activity before and after surgery also showed no increase in physical activity throughout the study period [26, 30]. Therefore, the lack of change in cardiopulmonary performance during the study cannot be attributed to any changes in sports participation in daily life.

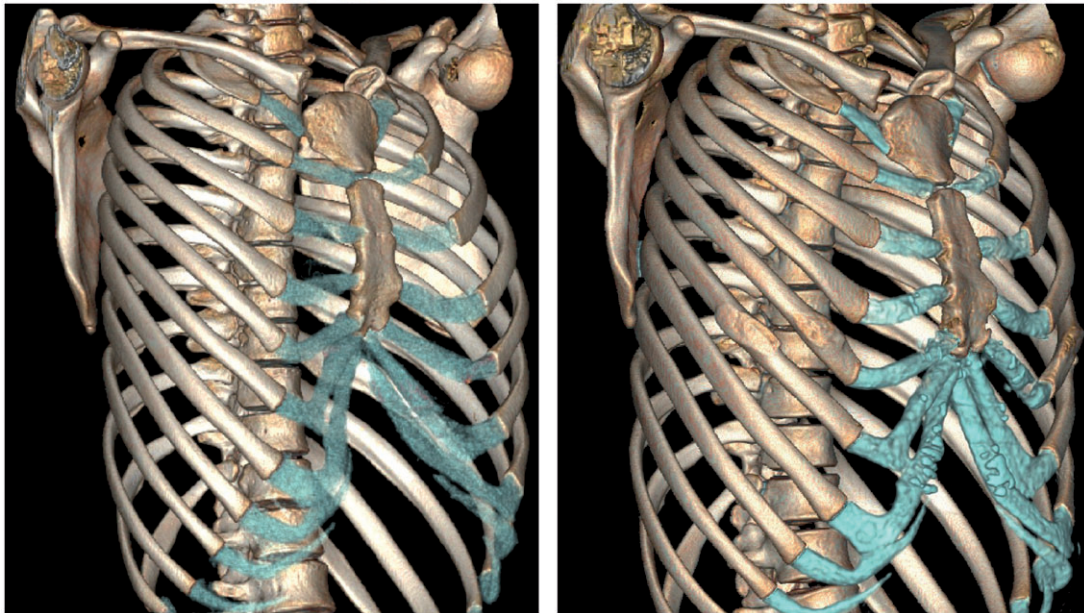


Figure 2: Three-dimensional volume-rendered computed tomography of the thorax of the same patient shown in Fig. 1: pre-surgery (left) and at 6 months after pectus bar removal (right).

Table 4: Reasons for patient exclusion from this study

Number of patients excluded (males/females)	Reason
4 (2 males/2 females)	Failed to appear for first study visit after giving consent to be included
5 (4 males/1 female)	Had no objective pathology at their preoperative screening and thus no desire for surgery
7 (6 males/1 female)	Had no objective indications for surgery even though correction of deformity was desired
6 (6 males)	Had objective indications for surgical correction other than MOVARPE, e.g. lipofilling, custom-made silicone implant
4 (4 males)	Could not be reached for final follow-up after MOVARPE
2 (1 male/1 female)	Had cardiological findings (long QT syndrome, $n = 1$; supraventricular tachycardia, $n = 1$) and excessive risk of complications according to PFTs

MOVARPE: Minor Open Videoendoscopically Assisted Repair of Pectus Excavatum; PFT: pulmonary function test.

Limitations

There were some limitations to our investigation. One limitation was the small final sample size due to the rarity of PE, and low motivation, to complete the study. The small final sample size was due to multiple factors, as listed in Table 4, and may have increased the risk of type II error. In addition, due to the multiple outcome measures, there may also have been an increased risk of type I error. We tried to minimize this issue by providing all effect sizes, thus enabling the reader to adequately assess the outcomes. Because PE occurs more often in males than females, most patients enrolled in the study were male. Furthermore, due to the long study period and multiple test designs, only

motivated patients completed all postoperative examinations, including the CT scan and cardiopulmonary function tests. This was especially challenging in the adolescent patients in our study. A further limitation was that the IPAQ and physical activity behaviour of adolescents evaluate exercise only during the last week prior to the test days; thus, no data about exercise over a longer period (e.g. the last month) were available.

CONCLUSION

Our findings indicate that surgical correction of PE does not impair cardiopulmonary function, at rest or during exercise. Therefore, no adverse effects on exercise performance should be expected from surgical treatment of PE utilizing the MOVARPE technique. However, without increasing regular physical activity after surgery, an improvement of cardiopulmonary function cannot be expected. Despite the improved aesthetic appearance, and thus potentially elevated self-esteem, the patients did not perform more daily exercise compared to before surgery. Finally, based on our findings, the intention and furthermore the medical indication to improve cardiopulmonary function via surgical thoracic wall elevation still remain an unsteady, but prevailing aesthetic and psychological indication. This new insight must be considered when dealing with PE patients, in terms of the selection of treatment and obtaining informed consent. In addition, our findings emphasize the high importance of standardized preoperative physical and psychological evaluations.

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Author contributions

Barbara Del Frari: Conceptualization; Funding acquisition; Investigation; Methodology; Project administration; Resources; Supervision; Validation; Writing—original draft; Writing—review & editing. **Cornelia Blank:** Data curation; Formal analysis; Methodology; Validation; Writing—original draft; Writing—review & editing. **Stephan Sigl:** Conceptualization; Data curation; Formal analysis; Investigation; Methodology; Project administration; Validation; Visualization; Writing—original draft; Writing—review & editing. **Anton H. Schwabegger:** Conceptualization; Methodology; Project administration; Supervision; Validation; Writing—original draft; Writing—review & editing. **Eva Gassner:** Data curation; Formal analysis; Investigation; Methodology; Validation; Visualization; Writing—original draft. **David Morawetz:** Data curation; Formal analysis; Methodology; Validation; Visualization; Writing—original draft. **Wolfgang Schoberberger:** Investigation; Methodology; Project administration; Resources; Supervision; Validation; Writing—original draft; Writing—review & editing.

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