SERVIZIO SANITARIO REGIONALE
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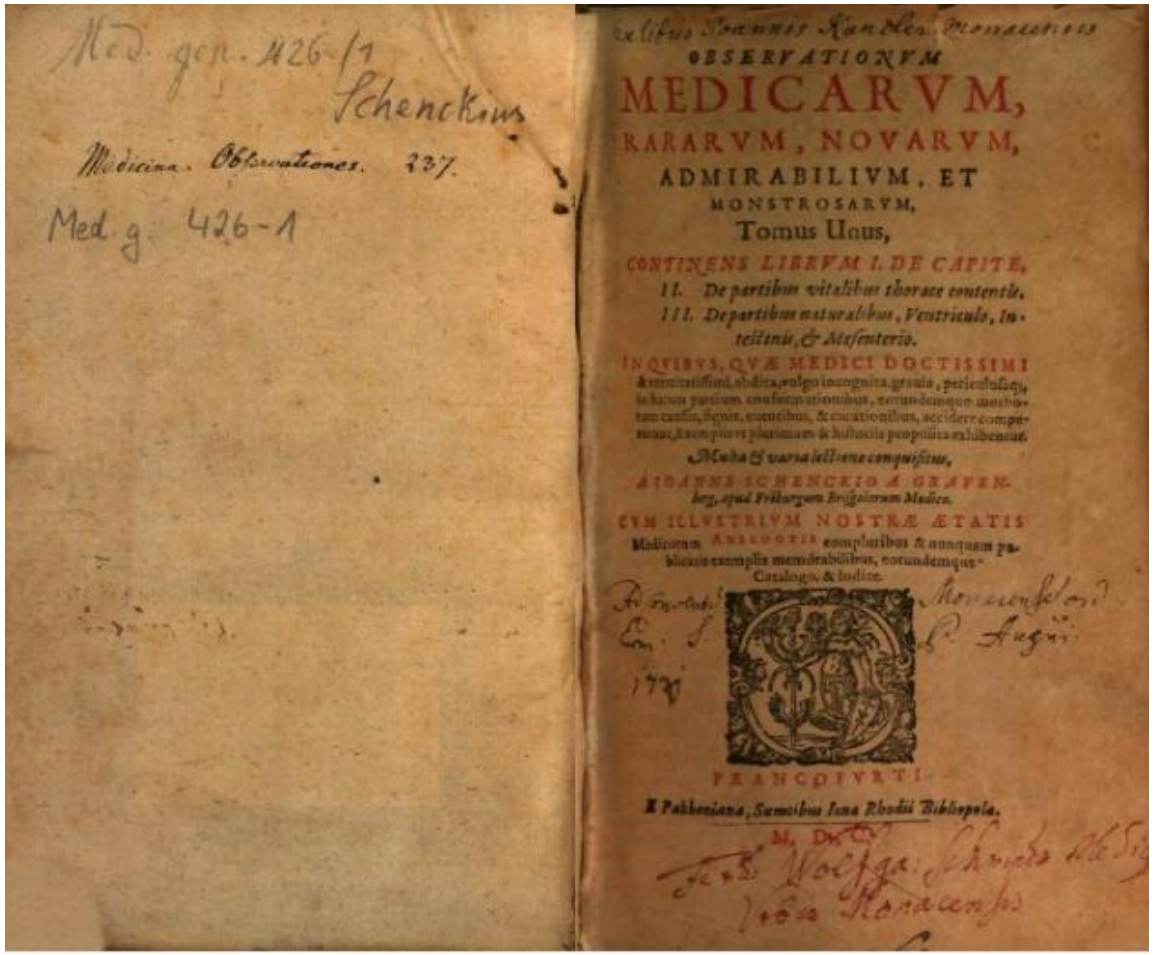
Università
degli Studi
di Ferrara

Valutazione della qualità della vita e della funzionalità respiratoria nei pazienti con pectus excavatum

Brunilda Marku

U.O Pneumologia



Described several features of the problem , including exercise intolerance in a patient from Andalusia

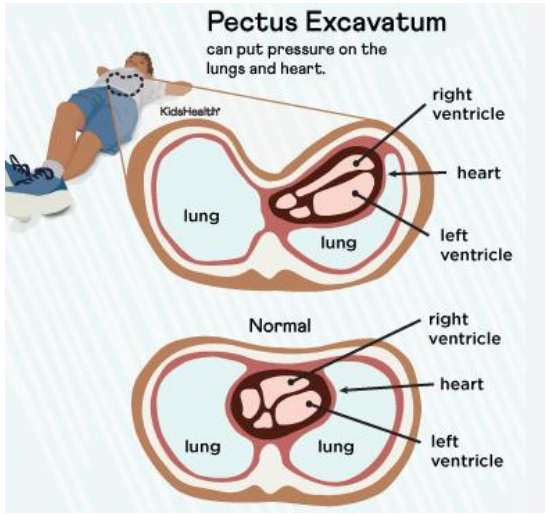
Observation 1594;264:516.

Pediatric Surgery, 4th Edition, Vol. 1 & 2. Kenneth J Welch, Judson G. Randolph, Mark M. Ravitch, James A. O'Neill, Jr., Marc I. Rowe. 1647 pp. Chicago, Year Book Medical Publishers. 1986.

Mark Ravitch commented that patients could fool around playing basketball, but not play a whole game, or a few games of tennis, but not a set

Physiological concerns

- Affects heart and lungs
- Physical experiences



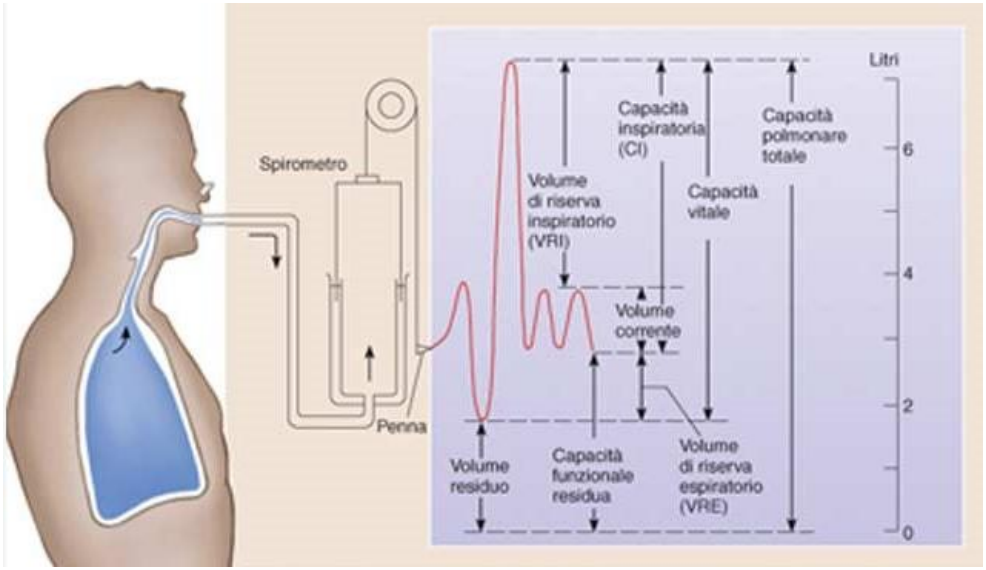
Psychosocial concerns

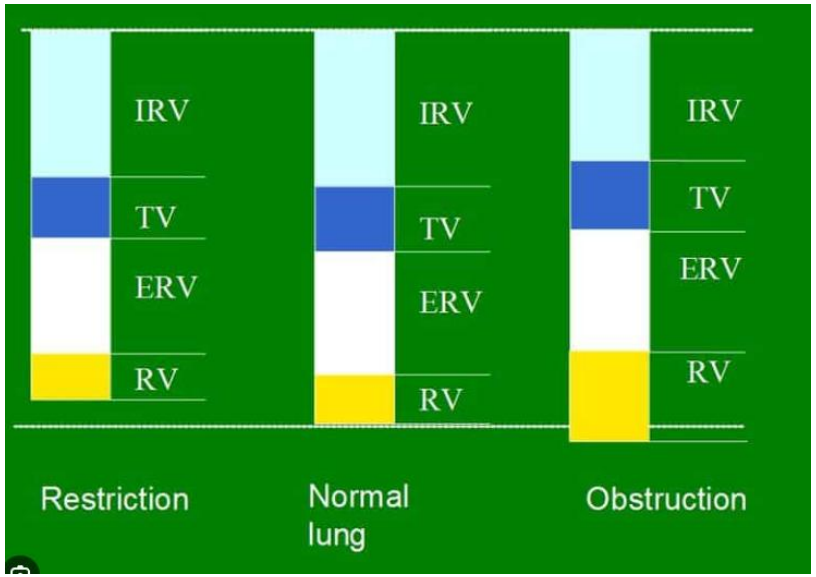
- Embarrasment
- Psycosocial concerns
- Inability to partecipate in activities

Lung function test in PE

It is estimated that PFTs are abnormal in about 20–25% of pectus patients

- Prevalent restrictive pattern with reduction of VC, RV and TLC –more severe if scoliosis is present
- Obstructive defect when other comorbidities are present (asthma)





Lawson ML 2011, Kelly 2010, Sesia SB 2016

The Prevalence and Effects of Pectus Excavatum and Pectus Carinatum on the Respiratory Function in Children between 7–14 Years Old

Zafer Kutay Coskun · Hasan Basri Turgut · Sadik Demirsoy · Ali Cansu

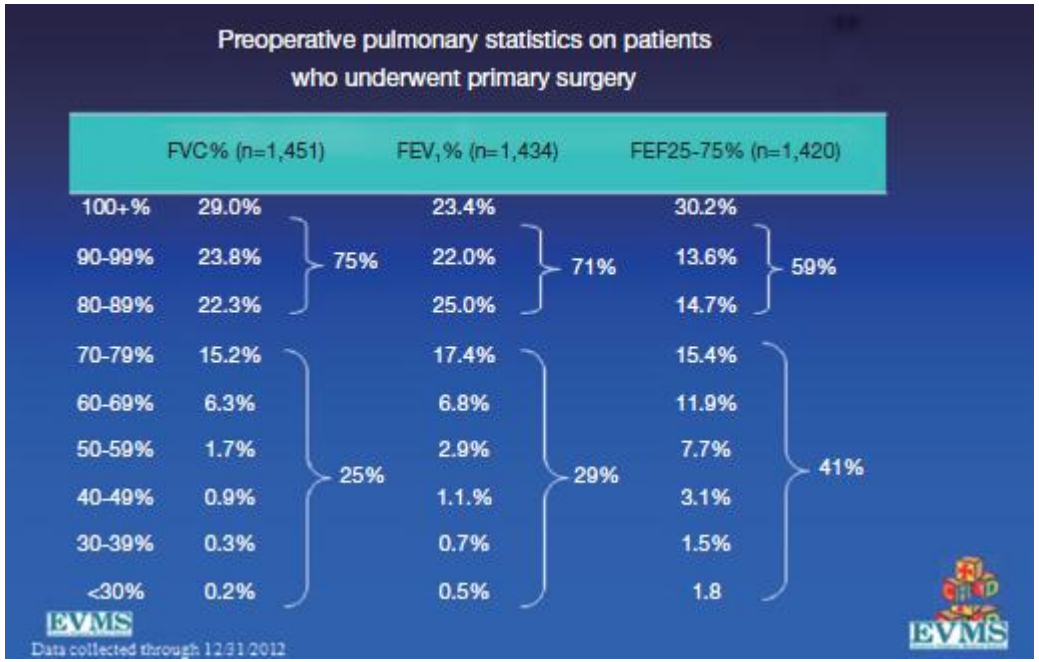
Abstract The study involved 1342 primary school students aged 7–14 years who applied to Ankara, a primary care center for general health check-up between 2006 and 2007. Forty-three students, 35 of whom had PE and 8 of whom had PC, were subjected to thorax measurement. All 43 students underwent pulmonary function tests (PFT).The prevalence rate of PC was 0.6%, and of PE, 2.6%. The thorax widths of the groups were similar (P=0.273). The thorax circumference and depth of PE group were lower than those of the controls (P<0.05). The probability rate of abnormality in PFT scores of PE group was statistically significantly higher than that of the controls (P=0.022) whereas absence of normal PFT scores the difference between PC group and the controls was not statistically significant (p=0.095). The results indicate that more than half of the individuals with pectus deformity do not have any physical complaints and do not have statistically significant differences in their PFT parameters.

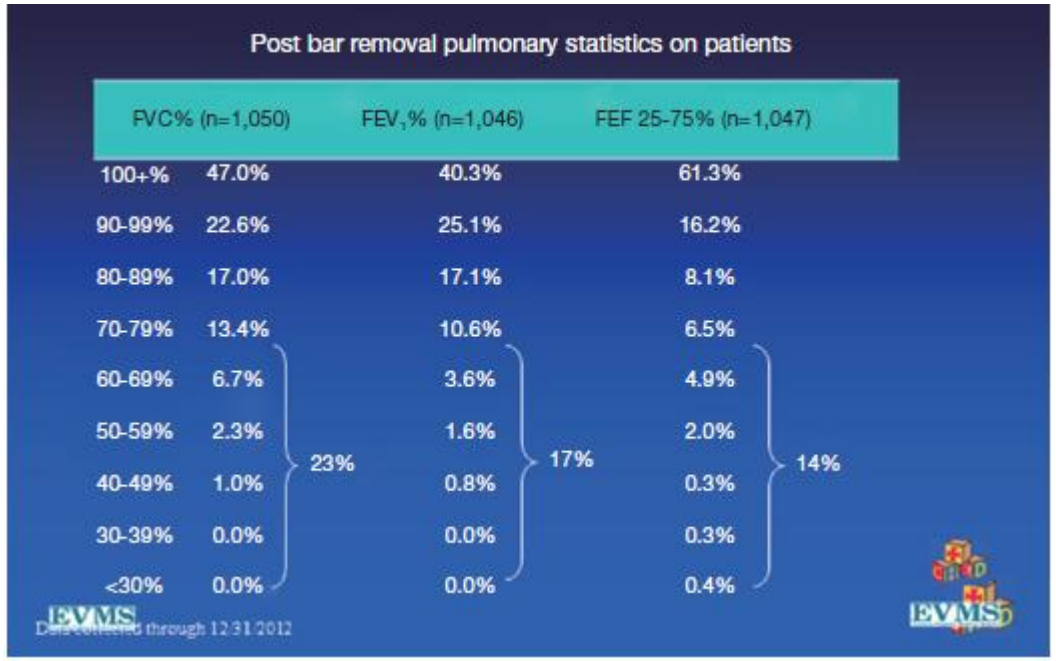
	Control		PC		PE		Total
	Frequency	%	Frequency	%	Frequency	%	Frequency
Normal	51	87.9	5	62.5	24	68.6	79.2
Abnormal	7	12.1	3	37.5	11	31.4	20.8
Mild restriction	5	8.6	2	25.0	9	25.7	15.8
Moderate obstruction	1	1.7	0	0.0	0	0.0	1.0
Severe restriction	1	1.7	0	0.0	0	0.0	1.0
Mild obstruction	0	0.0	1	12.5	2	5.7	3.0
Total	58	100.0	8	100.0	35	101	100

In a large study that looked at respiratory function in 1342 primary school students, 35 of which had pectus excavatum, PFT results were normal in 87.9% of the controls versus 68.6% in those with pectus excavatum

Diminished pulmonary function in pectus excavatum: from denying the problem to finding the mechanism

Robert E. Kelly Jr, Robert J. Obermeyer, Donald Nuss





Ann Cardiothorac Surg 2016;5(5):466-475

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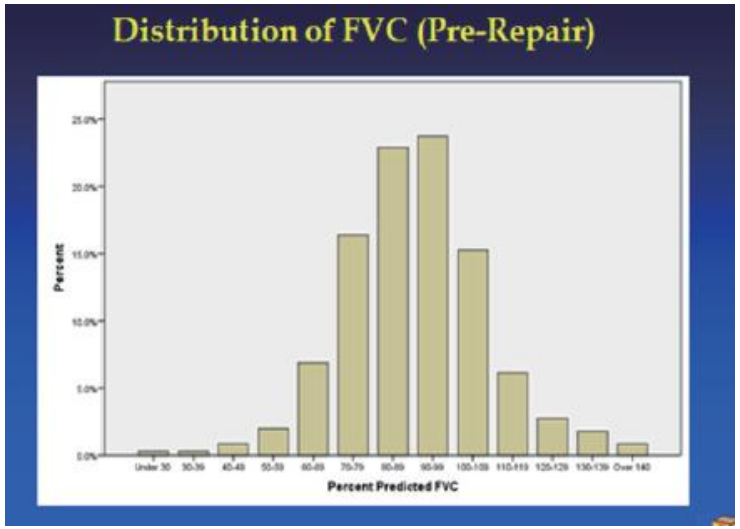

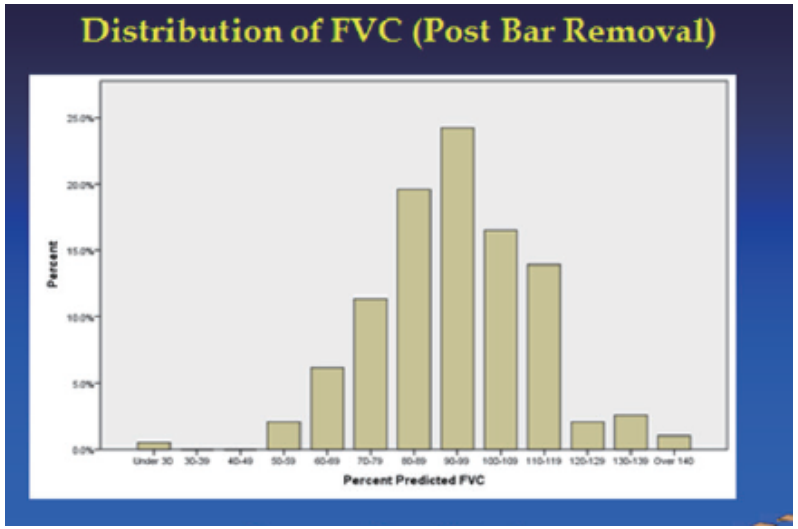

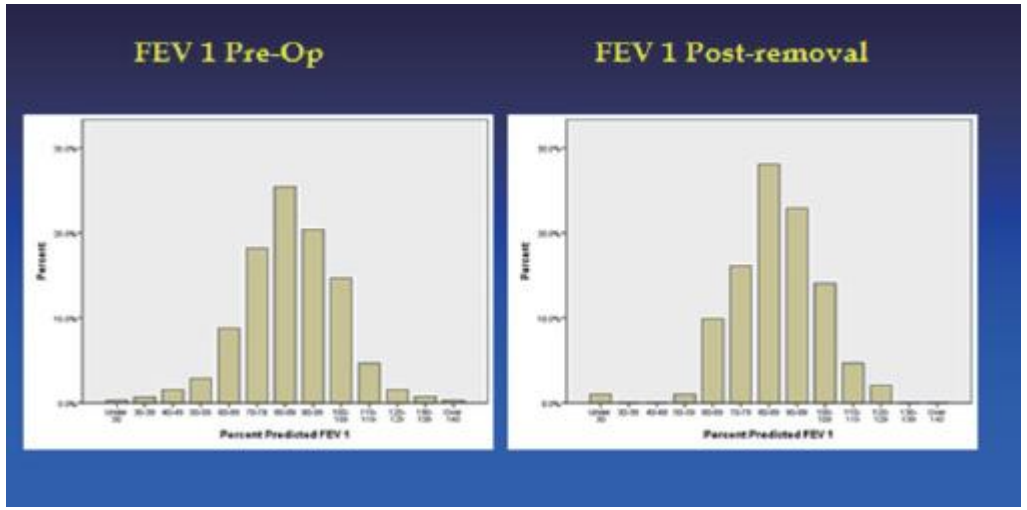

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ORIGINAL ARTICLE

(F)utility of preoperative pulmonary function testing in pectus excavatum to assess severity

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Abstract

Purpose The utility of pulmonary function testing (PFT) in pectus excavatum (PE) has been subject to debate. Although some evidence shows improvement from preoperative to postoperative values, the clinical significance is uncertain. A high failure-to-completion rate for operative PFT (48%) was identified in our large institutional cohort. With such a high non-completion rate, we questioned the overall utility of PFT in the preoperative assessment of PE and sought to evaluate if other measures of PE severity or cardiopulmonary function could explain this finding.

Methods Demographics, clinical findings, and results from cardiac MRI, PFT (spirometry and plethysmography), and cardiopulmonary exercise tests (CPET) were reviewed in 270 patients with PE evaluated preoperatively between 2015 and 2018. Regression modeling was used to measure associations between PFT completion and cardiopulmonary function.

Results There were no differences in demographics, symptoms, connective tissue disorders, or multiple indices of pectus severity and cardiac deformation in PFT completers versus non-completers. While regression analysis revealed higher RVEF, LVEF, and LVEF-Z scores, lower RV-ESV/BSA, LV-ESV/BSA, and LV-ESV/BSA-Z scores, and abnormal breathing reserve in PFT completers vs. non-completers, these findings were not consistent across continuous and binary analyses.

Conclusions We found that PFT completers were not significantly different from non-completers in most structural and functional measures of pectus deformity and cardiopulmonary function. Inability to complete PFT is not an indicator of pectus severity.

	Fully Acceptable & Repeatable (PFT Completers)	Partially Interpretable (PFT Non-completers)	p-value
	147 (54.4%)	123 (45.6%)	
Age (years), mean (SD)	15.1 (3.50)	15.5 (3.32)	0.38
Age group (years), % (n)			0.59
< 11	2.7% (4)	2.5% (3)	
11-17	84.3% (123)	80.3% (98)	
18+	13.0% (19)	17.2% (21)	
Male sex, % (n)	84.9% (124)	76.2% (93)	0.07
White race, % (n)	98.6% (144)	97.5% (119)	0.66

	Fully Acceptable & Repeatable	Partially Interpretable	p-value
	147 (54.4%)	123 (45.6%)	
Symptoms, % (n)	71.9% (105)	77.2% (96)	0.23
Chest pain	41.0% (60)	40.2% (49)	0.88
Shortness of breath	62.3% (91)	62.3% (76)	0.99
Heart palpitations	11.6% (17)	13.9% (17)	0.57
Exercise intolerance	63.7% (93)	63.1% (77)	0.92
Connective tissue disorder, % (n)	9.6% (14)	8.2% (10)	0.69
Hypermobility syndrome	6.9% (10)	4.1% (5)	0.33
Ehlers-Danlos syndrome	2.1% (3)	4.1% (5)	0.48
Marfans	0.7% (1)	0.8% (1)	1.00
POTS	0.7% (1)	0% (0)	1.00
Haller Index, median (Q1,Q3)	4.8 (4.0, 5.8)	4.8 (4.0, 6.0)	0.94
Depression Index, mean (SD)	0.61 (0.27)	0.65 (0.31)	0.24
Correction Index, mean (SD)	33.9 (14.00)	34.0 (15.22)	0.93

Ventilatory limitations are not associated with dyspnea on exertion or reduced aerobic fitness in pectus excavatum

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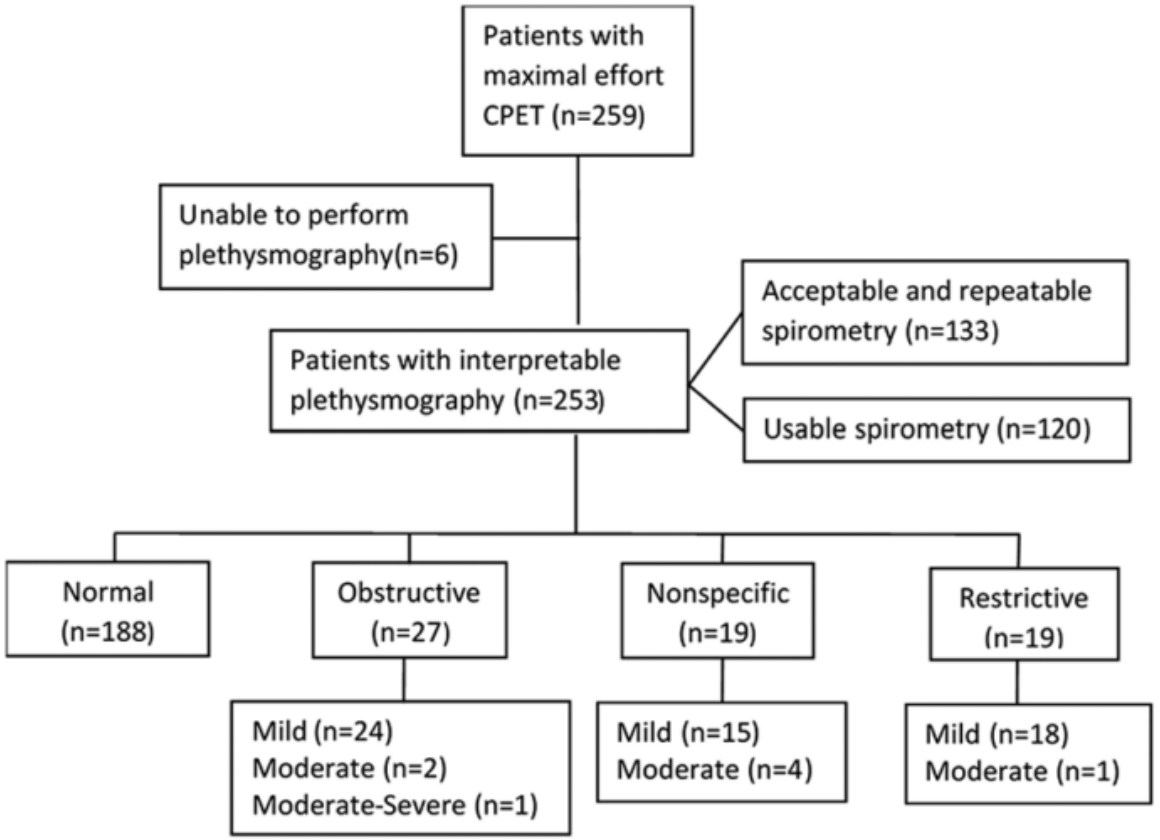

	Normal	Obstructive	NSVL	Restrictive	*p* Value
Total number subjects	188	27	19	19	
Age (years), mean (*SD*)	15.7 (3.3)	16.3 (1.6)	17.7 (4.3)	15.1 (1.1)	*.041*
Body mass index, mean (*SD*)	18.9 (3)	19.9 (2.9)	19.1 (2.4)	18.9 (2.1)	.40
Haller index, median (Q1, Q3)	4.8 (4, 5.9)	4.5 (4, 5.3)	5.0 (4.3, 6.2)	4.8 (4, 5.4)	.60
Correction index, mean (*SD*)	34.4 (14.1)	31.2 (16.8)	35.0 (14.8)	31.2 (10.4)	.59
Dyspnea with exercise, % (*n*)	64.4% (121)	63.0% (17)	68.4% (13)	42.1% (8)	.27
Chest Pain, % (*n*)	42.6% (80)	48.2% (13)	42.1% (8)	10.5% (2)	*.045*
Peak VO_2, mean (*SD*)	89.9 (16.5)	87.3 (16.1)	85.9 (15.9)	84.5 (18.7)	.42
Peak O_2 pulse, mean (*SD*)	92.0 (17.6)	89.4 (17.6)	86.3 (17.2)	85.7 (18.7)	.28
Breathing reserve, mean (*SD*)	0.27 (12.8)	0.33 (11.1)	0.32 (13.4)	0.26 (15.1)	.08
Vt/IC, mean (*SD*)	0.67 (0.11)	0.66 (0.12)	0.69 (0.16)	0.69 (0.11)	.73
Respiratory rate, mean (*SD*)	52.5 (8.9)	50.1 (7.8)	49.8 (10.3)	52.7 (9.0)	.35

Cardiopulmonary exercise test

- The most reliable and sensitive test to investigate exercise intolerance
- Can investigate accurately a patient's exercise intolerance when it is most troublesome, which is during maximal exertion in a vertical position.
- It is also able to give an accurate estimation as to how abnormal the patient is compared to an age- and gendermatched control and may be repeated postoperatively to assess the results of the operation.
- All centres seeing patients should have access to CPET services
- Abnormal VO2 max < 85%

Cardiopulmonary Outcomes After the Nuss Procedure in Pectus Excavatum

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Variable	Preoperative CPET	Postoperative CPET	*P* value (95% CI for the difference)
Demographics			
Age, y	32.4±10.0	35.5±14.2	
Weight, kg	72.8±15.4	75.3±15.4	<0.001 (1.3 to 3.5)
Body mass index, kg/m^2	22.5±3.6	23.0±3.5	0.002 (0.2 to 0.9)
Maximum workload			
Estimated METS	9.4±1.5	9.7±1.7	0.032 (0.02 to 0.50)
Actual METS	7.4±1.7	8.1±2.0	<0.001 (0.5 to 1.0)
Work, W	174.5±44.8	185.2±44.1	<0.001 (5.9 to 15.5)
Work (W per kg body weight)	2.4±0.5	2.5±0.5	0.040 (0.0 to 0.1)
Heart rate at rest, bpm	90.8±13.9	83.4±15.3	<0.001 (4.8 to 10.0)
Maximum heart rate, bpm	161.6±15.4	164.7±14.3	0.013 (0.6 to 5.4)
RER	1.22±0.1	1.24±0.1	0.029 (0.0 to 0.1)
DBP at rest, mm Hg	82.1±10.7	78.9±8.8	0.003 (1.1 to 5.4)
DBP at peak exercise, mm Hg	84.4±11.9	83.8±11.7	0.600 (−2.8 to 1.7)
SBP at rest, mm Hg	124.9±14.7	123.6±13.6	0.376 (−4.4 to 1.6)
SBP at peak exercise, mm Hg	164.5±26.2	180.6±26.7	<0.001 (11.7 to 20.3)
VE/CO_2 slope	27.0±5.2	26.1±3.5	0.075 (−1.9 to 0.1)
VO_2 max			
Relative VO_2 max, mL/kg per min	25.9±6.0	28.5±7.0	<0.001 (1.6 to 3.5)
Relative VO_2 max/predicted (%)	72.8±15.4	84.2±20.6	<0.001 (8.6 to 14.1)
Absolute VO_2 max, L/min	1.9±0.6	2.1±0.6	<0.001 (0.2 to 0.3)
Normal VO_2 max values (n)	30.8% (40)	58.5% (76)	<0.001
O_2 pulse			
O_2 pulse, mL/beat	11.7±3.6	12.9±3.7	<0.001 (0.8 to 1.6)
O_2 pulse/predicted, %	84.5±16.9	94.3±21.4	<0.001 (6.9 to 12.6)
Anaerobic threshold			
VO_2 at anaerobic threshold, mL/kg per min	14.6±4.3	16.9±6.4	<0.001 (1.2 to 3.3)
Maximal ventilation			
VE BTPS, L/min	67.5±18.8	73.3±17.7	<0.001 (3.1 to 8.4)
VE BTPS/predicted, %	39.2±9.8	48.7±12.4	<0.001 (7.6 to 11.5)

The pectus care guidelines: best practice consensus guidelines from the joint specialist societies SCTS/MF/CWIG/BOA/BAPS for the treatment of patients with pectus abnormalities

Joel Dunning [a,*] (UK), Clare Burdett [a] (UK), Anne Child [b] (UK), Carl Davies [c] (UK), Deborah Eastwood [d] (UK), Tim Goodacre [e] (UK), Frank-Martin Haecker [f,g] (Switzerland), Simon Kendall [h] (UK), Shyam Kolvekar [i] (UK), Lisa MacMahon [j,k] (USA), Sean Marven [l] (UK), Sarah Murray [m] (UK), Babu Naidu [n] (UK), Bejal Pandya [i] (UK), Karen Redmond [o] (UK) and Aman Coonar [p] (UK)

Recommendation: Patients with suspected exercise intolerance

We recommend that all patients undergo cross-sectional imaging of the thorax to determine the Haller index and a review of the anatomical compression of the right ventricle.
(Class I, Level of Evidence B)

All patients should undergo echocardiography to exclude other causes of shortness of breath and to measure the size of the aortic root. This test is not sensitive in excluding patients from having cardiac compression.
(Class IIa, Level of Evidence C)

All patients should undergo lung function testing to exclude other causes of shortness of breath. A normal test result does not exclude the patient from having cardiac compression.
(Class IIa, Level of Evidence C)

We recommend that all patients with severe pectus excavatum and exercise intolerance undergo a cardiopulmonary exercise test because it is the best test to identify exercise intolerance that is caused by pectus excavatum. A ventilatory update of oxygen (VO_2) max below 85% of predicted is regarded as an abnormality.
(Class I, Level of Evidence B)

Cardiac MRI scanning may be performed, and it may show abnormalities that recover after an operation or following the application of a vacuum bell, but the test is not reliable enough to exclude a patient from having exercise intolerance as a result of severe pectus excavatum.
(Class IIb, Level of evidence B)

Quality of life, psychological states, and personality traits in patients with pectus excavatum

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ABSTRACT

Objective: The quality of life (QOL) and psychological states of patients with pectus excavatum (PE) have yet to be well understood. This study aimed to evaluate the health-related QOL (HRQOL), psychological states, and personality traits of patients with PE, alongside the associations of these factors with the severity of PE.

Methods: A cross-sectional evaluation was prospectively performed in patients scheduled to undergo PE repair surgery between July 2019 and April 2021. The primary outcome was the patients' HRQOL, and the secondary outcomes were depression, social anxiety, self-efficacy, and personality traits.

Results: In total, 129 patients were subjected to analyses. Patients' HRQOL had a lower role component summary score (mean ± standard deviation: 41.8 ± 12.8, $P < .001$) than the general population controls. Patients' HRQOL had a significantly better physical component summary (54.0 ± 10.4, $P < .001$) and mental component summary (53.3 ± 8.8, $P < .001$) than that of the general population. Fourteen patients' (10.9%) and 56 patients' (43.4%) scores indicated the presence of depression and social anxiety disorder, respectively. Patients' self-efficacy (46.1 ± 11.4, P, .001) and level of extraversion (46.5 ± 11.8, $P < .001$) were lower than those of the general population. No significant correlation was found between the severity of PE and these scores.

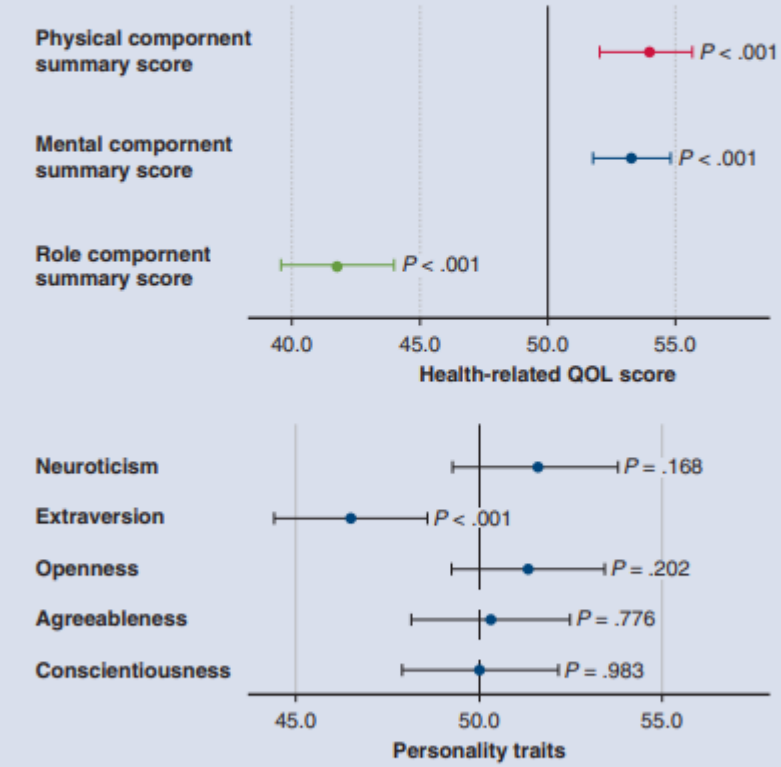
Conclusions: Our study revealed that patients with PE had decreased social-role QOL, depressive tendencies, increased social anxiety, lower self-efficacy, and introversion. No correlation between the severity of PE and the patients' psychological outcomes leads us to conclude that surgical implications of PE should not be decided solely by a physical index. (JTCVS Open 2024;19:355-69)

QOL and psychological status in patients with pectus excavatum

Methods:

- A cross-sectional evaluation was performed in patients with PE (n = 129).
- July 2019 - April 2021
- Using questionnaires to measure the mental health status before repair surgery for PE.
  1. Health-related QOL
  2. Psychological distress (depression, social anxiety, lower self-efficacy)
  3. Personality traits

Results:



Implications:

- The current study demonstrated that patients with PE had decreased social-role QOL, depressive tendencies, increased social anxiety, lower self-efficacy, and introverted personalities.
- Further prospective studies are needed to verify whether surgical repair can improve the psychosocial aspects of patients with PE.

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Study	Population	Intervention and comparison	Outcomes reported
Kelly *et al.*, 2020, prospective study [38] USA	$n = 996$ children and adults with PE Haller index: 5.46 ± 8.26 No subgroups reported	**Intervention** Nuss procedure **Comparison** No comparator	**Important outcomes** Postoperative cosmetic satisfaction Safety: early (< 30 day) and late (timescale not stated) postoperative complications
Pawlak *et al.*, 2018, retrospective study [41] Poland	$n = 1006$ with excavatum, including $n = 44$ with recurrence after previous surgery. Age: 18.6 ± 5.7 years (range 7–62 years) Haller index: 3.7 ± 1.4 (2.6–17.3) No subgroups reported	**Intervention** Nuss procedure using video-assisted thoracoscopy **Comparison** No comparator	**Important outcomes** Early (up to 30-day) thoracoscopy-dependent postoperative complications
Pawlak *et al.*, 2016, retrospective study [42] Poland	$n = 680$ with PE who had surgery. The authors confirm that this is a subset of their more recent paper [39].	**Intervention** Nuss procedure **Comparison** No Comparator	**Critical outcomes** Cardiopulmonary outcomes—imaging
Luo *et al.*, 2017, prospect-	$n = 266$ with PE	**Intervention** the Nuss or Ravitch procedure	**Critical outcomes** Psychological outcomes:
O'Keefe *et al.*, 2013, prospective study [18] Canada	$n = 67$ with PE Age: 13.9 ± 2.3 years Haller index: 4.4 ± 1.3 No subgroups reported	**Intervention** Nuss procedure with bar removal when the patients had reached skeletal maturity or after 3 years, whichever came first **Comparison** No comparator	**Critical outcomes** Cardiopulmonary outcomes—exercise testing preoperatively and 3 to 6 months after bar removal Cardiopulmonary outcomes—imaging preoperatively and 3 to 6 months after bar removal **Important outcomes** Lung function tests preoperatively and 3 to 6 months after bar removal Postoperative cosmetic satisfaction
Neviere *et al.*, 2011, Prospective study [17] France	$n = 70$ with PE Age: 27 ± 11 years (range 18–62 years) Haller index: 4.5 ± 1.1. No subgroups reported	**Intervention** Simplified Ravitch-type repair with removal of strut 6 months after initial procedure **Comparison** No comparator	**Critical outcomes** Cardiopulmonary outcomes—exercise testing preoperatively and 1 year postoperatively Cardiopulmonary outcomes—imaging pre- and postoperatively: Haller index **Important outcomes** Lung function tests

Recommendation

There is good evidence that patients who are psychologically impacted by their pectus abnormality benefit from surgery in terms of improved quality of life, reduced depression and anxiety scores and that the operation has good patient satisfaction.
(Class IIa, Level of Evidence B)

The assessment of the psychological impact of a pectus abnormality on a patient should be assessed by a psychologist prior to the operation for psychological benefit.
(Class IIa, Level of Evidence C)

The NUSS questionnaire or the NUSS questionnaire modified for adults (also known as the Pectus Excavatum Evaluation Questionnaire–PEEQ) should be used to demonstrate an improvement in quality of life in patients undergoing pectus surgery.
(Class IIa, Level of Evidence B)

Conclusions

- Data on lung function test are controversial
- CPET can accuretely investigate patient’s exercise intolerance
- Important psychosocial impact
- Benefits of surgery are an important issue for life style improvement