Effect of Pectus Excavatum Deformity on Cardiorespiratory Fitness in Adolescent Boys

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Objective: To determine the magnitude of the effects of pectus excavatum deformity on endurance fitness and cardiorespiratory functional reserve in adolescent boys.

Design: Cross-sectional comparison of cardiac and ventilatory variables at rest and during a maximal cycle exercise test.

Setting: Pediatric exercise-testing laboratory.

Participants: Twelve boys (mean ± SD age, 14.1 ± 1.8 years; age range, 11.8-18.0 years) with moderate-to-severe pectus excavatum deformity (mean ± SD Haller index, 3.95 ± 0.88) and 20 control boys (mean ± SD age, 12.5 ± 0.4 years; age range, 12.1-13.5 years) without musculoskeletal deformity.

Main Outcome Measures: Endurance fitness (physical work capacity); respiratory rate, tidal volume, and minute ventilation; and cardiac output and stroke volume by Doppler echocardiography.

Results: Patients with pectus deformity had significantly lower endurance fitness than controls (mean ± SD physical work capacity, 2.60 ± 0.28 W · kg^{-1} vs 3.11 ± 0.45 W · kg^{-1}) and reduced mean ± SD values for maximal cardiac index (10.6 ± 1.6 L · min^{-1} vs 12.0 ± 2.2 L · min^{-1}) and peak tidal volume (3.02 ± 0.27 mL · kg^{-1} · 10^{-2} vs 3.46 ± 0.30 mL · kg^{-1} · 10^{-2}). However, considerable overlap was observed in these values between the 2 groups.

Conclusions: As a group, boys with pectus excavatum deformity have lower endurance fitness than controls, and this is associated with reduced cardiac output and tidal volume responses to exercise. However, the wide variability of these measures makes it difficult to assign pectus deformity as a cause of exercise intolerance in individual patients.

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THE EXTENT TO WHICH ANteroposterior chest compression imposed by a pectus excavatum deformity can limit exercise capacity remains controversial.[1] Moreover, the means by which such a restriction of fitness might occur—anatomic, physiologic, or psychological—is unclear. The question has particular clinical significance because indications for surgical intervention may depend on potential risks for cardiac or ventilatory dysfunction.[2] The large body of research examining this issue during the past several decades has provided no clear consensus. Early studies with few participants and anecdotal case reports suggested that a certain number of patients with moderate-to-severe pectus deformity experienced significant exercise intolerance, and in some cases this was improved by surgical intervention.[3] Some subsequent studies[4,5] demonstrated physiologic limitations at rest and during submaximal exercise, whereas other studies[2,6] did not. These cardiorespiratory deficits were highly variable and did not always correspond to the level of exercise intolerance, and improvements were not consistently seen after surgical repair.

Cardiac limitation in patients with pectus excavatum has most commonly been documented as lower resting and submaximal exercise stroke volumes in the sitting position vs supine compared with healthy subjects.[4,7] Depressed maximal breathing capacity and lung tidal volumes have been indicated as ventilatory features in some (but not all) studies of these patients as well.[5,8,9] Several researchers[1,10] have cautioned that these comparisons between patients with pectus deformity and healthy individuals have not always taken into account important confounding variables, such as degree of pectus deformity, degree of physical conditioning, and comparison with appropriate control subjects.

Previous exercise studies[4,7] have not provided information regarding cardiac variables measured during maximal exer-

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cise testing in patients with pectus excavatum deformity compared with controls. This study was designed to assess exercise endurance capacity and markers of cardiac and ventilatory functional reserve during progressive maximal exercise testing in a group of adolescent boys with moderate-to-severe pectus excavatum deformity. Findings were compared with those of healthy boys using identical testing methods.

METHODS

Eighteen adolescent boys who were being considered for surgical repair of pectus excavatum deformity underwent maximal cycle testing with measurement of cardiorespiratory variables. Of these 18 boys, 6 were considered to have failed to provide a true exhaustive exercise effort (see several paragraphs later herein), and data on the remaining 12 boys provide the basis of this study. The severity of pectus deformity was assessed using the computed tomography–derived ratio of the transverse thoracic dimension to the sternovertebral distance (the Haller index).[11] The mean±SD value was 3.95±0.88 (range, 2.93-5.37).

The mean±SD age of the patients was 14.1±1.8 years (age range, 11.8-18.0 years). All the patients were in good health and had no evidence of cardiac or respiratory disease; none was taking medications that would affect cardiorespiratory fitness. Nine patients (75%) had participated on organized sports teams (basketball, hockey, or football) in the preceding 3 months, but none was involved in an aerobic training program. By questionnaire, the parents scored their child's level of habitual physical activity on a 5-point scale from 1 (inactive—watches television, reads, or does homework after school; no extracurricular sports) to 5 (very active—participates regularly in extracurricular sports; dislikes sedentary activities). On this scale, the average score was 3.7.

Measured cardiovascular variables were compared with those of a control group of 20 healthy boys (mean±SD age, 12.5±0.4 years; age range, 12.1-13.5 years) without musculoskeletal deformity who were participants in an earlier study regarding cardiovascular fitness and run performance in children.[12] The controls were selected to provide a broad range of fitness, based on school 1-mile run times. Fourteen controls (70%) had recently participated on community sports teams. On the activity questionnaire, the mean score was 3.8.

Anthropometric techniques, exercise protocol, and measurement of cardiorespiratory variables were identical in patients with pectus excavatum and controls. Height and weight were recorded using a stadiometer and a calibrated balance beam scale, respectively. Average triceps and scapular skinfold thicknesses were determined from triplicate measurements on the right side of the body to the nearest 0.1 mm using standard techniques. Mean values were summed to create a skinfold score.

Exercise was performed in an air-conditioned laboratory (20°C-22°C with moderate humidity). Participants cycled in the upright position to exhaustion, with a progressive protocol of 3-minute work stages and 25- or 50-W increments (depending on body size). Pedaling cadence was 50 rpm. A true exhaustive effort was considered to have been achieved if the boys demonstrated (1) a peak heart rate exceeding 183 beats per minute (bpm) (95% of predicted) and (2) subjective evidence of fatigue (hyperpnea, sweating, or facial flushing).[13] Endurance performance was assessed using physical work capacity (PWC)—the highest workload achieved—prorated for incomplete work stages and related to body mass. Ventilatory measures (tidal volume, respiratory rate, and calculated minute ventilation) were determined using a Q-Plex Cardiopulmonary System (Quinton Instruments, Seattle, Wash), which uses a pneumotachometer in the expiratory line.

Heart rate was measured using electrocardiography. Stroke volume at rest and during exercise was estimated using standard Doppler echocardiographic techniques.[14] The validity and reliability of this method have previously been reported.[15,16] Ascending aortic blood flow velocity was recorded using a 2.0-MHz transducer positioned in the suprasternal notch. Area under the velocity curve across time for each beat was integrated to obtain the velocity-time integral. The mean velocity-time integral was calculated from the 3 to 10 highest and most distinct curves measured at rest, in the final minute of each work stage, and immediately before termination of exercise. This value was multiplied by the cross-sectional area of the aortic root to obtain the stroke volume. The aortic area was calculated from the greatest systolic diameter measured at the sinotubular junction as viewed from the parasternal long axis on a 2-dimensional echocardiogram with the subject seated on the treadmill just before exercise testing. Cardiac output was determined as the product of stroke volume and heart rate.

Stroke volume and cardiac output were related to body surface area (stroke index and cardiac index) based on studies by Armstrong and Welsman[17] and Rowland et al.[18] Minute ventilation was adjusted for height, given reports that exercise minute volume (expired) related to stature is independent of size in adolescent boys,[19] and tidal volume was expressed relative to body mass, a relationship which is constant during childhood.[20]

Comparisons between patients with pectus excavatum and controls were made at rest, at a given absolute submaximal workload (50 W), and during maximal exercise. Mean differences were assessed using independent *t* tests. Relationships among severity of pectus deformity (Haller index), endurance fitness (PWC per kilogram), and resting and maximal values of tidal volume, minute ventilation, stroke index, and cardiac index were assessed using Pearson product moment correlation coefficients. Statistical significance was defined as $P<.05$. This study was reviewed and approved by the institutional review board of the Baystate Medical Center. Informed consent and assent were obtained from parents and participants, respectively. Data are given as mean±SD.

RESULTS

The 12 patients with pectus excavatum were heavier (57.6±13.4 kg [range, 47.0-71.5 kg] vs 47.2±10.9 kg [range, 32.6-71.9 kg]; $P=.02$) and taller (168±12 cm [range, 146-188 cm] vs 157±8 cm [range, 139-176 cm]; $P=.01$) than the 20 control subjects. No difference was observed between the 2 groups in body composition, with a skinfold thickness sum of 19.7±7.6 mm in patients and 19.2±7.0 mm in controls. As noted previously herein, the 2 groups were similar in estimated level of habitual activity and in participation in organized sports.

Maximal heart rate was 191±8 bpm in patients with pectus excavatum and 195±6 bpm in controls ($P=.17$), indicating an equivalent maximal exercise effort in the 2 groups. Endurance fitness as indicated by PWC per kilogram was significantly lower in the pectus excavatum group (2.60±0.28 $W \cdot kg^{-1}$) compared with controls (3.11±0.45 $W \cdot kg^{-1}$; $P=.001$) (**Figure 1**). No correlation was observed between the degree of pectus deformity using the Haller index and PWC per kilogram ($r=0.18$; $P=.37$).

Accurate estimates of stroke volume using the Doppler echocardiographic technique are contingent on the

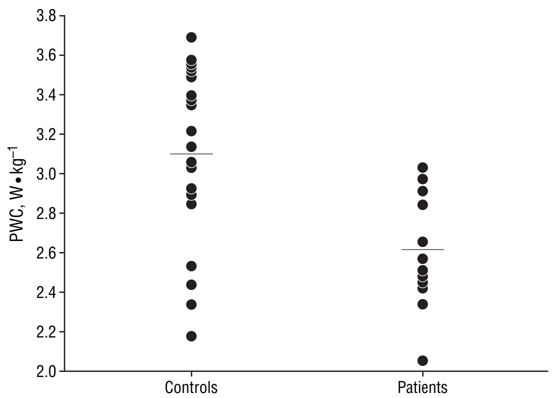

Figure 1. Cycle endurance fitness as defined by physical work capacity (PWC) relative to body mass in 12 patients with pectus excavatum deformity and 20 control subjects. Horizontal bars indicate means.

assumption of a circular aortic cross section. The aortic root diameter (related to the square root of body surface area[21]) was similar in patients and controls (1.79±0.12 cm · m^{-2} and 1.76±0.11 cm · m^{-2}, respectively), suggesting no differences in aortic root cross-sectional shape (ie, no anteroposterior compression by the pectus deformity).

At rest, few differences were observed between patients with pectus excavatum and controls (**Table**). The stroke index was 13% higher in controls (P=.07). Resting tidal volume per kilogram was significantly diminished in patients with pectus excavatum (1.14±0.22 mL · kg^{-1} · 10^{-2}) compared with controls (1.35±0.30 mL · kg^{-1} · 10^{-2}). A moderate correlation was observed between the Haller index and resting tidal volume per kilogram (r=0.51; P=.08). There were no group mean differences in the cardiac index or height-adjusted minute ventilation.

At a common absolute workload of 50 W, similar findings were observed (Table). The only statistically significant difference between groups was a lower tidal volume per kilogram in patients with pectus excavatum (1.75±0.34 mL · kg^{-1} · 10^{-2} vs 2.06±0.31 mL · kg^{-1} · 10^{-2}; P=.02), but this did not affect ventilatory response because mean values of minute volume (expired) related to height were similar. The stroke index was again slightly lower in boys with pectus deformity, and their mean cardiac index was 9% lower (P=.09).

At maximal exercise, the trend for a lower stroke index in patients with pectus excavatum persisted, but the 11% difference was not significant (P=.11) (Table). Maximal stroke index (r=0.64; P=.001) and cardiac index (r=0.67; P=.001) correlated positively with PWC per kilogram. (The maximal exercise cardiac index was higher in control subjects than in patients with pectus deformity (12.0±2.2 L · min^{-1} · m^{-2} vs 10.6±1.6 L · min^{-1} · m^{-2}; P=.05) (**Figure 2**). Tidal volume at peak exercise continued to be significantly greater in control subjects than in patients with pectus excavatum (3.46±0.43 mL · kg^{-1} · 10^{-2} and 3.02±0.44 mL · kg^{-1} · 10^{-2}, respectively; P=.01), but no group differences were seen in maximal minute ventilation. No significant relationships were observed between the Haller index and any of the physiologic variables at peak exercise.

Table. Cardiorespiratory Variables at Rest, During Submaximal Exercise, and During Maximal Exercise in Patients With Pectus Excavatum Deformity and Control Subjects*

	Patients (n = 12)	Controls (n = 20)
Rest		
Heart rate, bpm	92 ± 15	90 ± 16
Stroke index, mL · m^{-2}	40 ± 7	45 ± 9
Cardiac index, L · min^{-1} · m^{-2}	3.62 ± 0.60	4.04 ± 0.88
Minute ventilation, L · min^{-1} · m^{-1}	7.5 ± 1.8	7.5 ± 2.0
Tidal volume, mL · kg^{-1} · 10^{-2}	1.14 ± 0.22	1.35 ± 0.30†
Respiratory rate, bpm	22 ± 5	20 ± 5
Submaximal exercise, 50 W		
Heart rate, bpm	123 ± 11	128 ± 13
Stroke index, mL · m^{-2}	53 ± 9	57 ± 11
Cardiac index, L · min^{-1} · m^{-2}	6.49 ± 0.99	7.23 ± 1.40
Minute ventilation, L · min^{-1} · m^{-1}	16.8 ± 2.7	17.1 ± 1.7
Tidal volume, mL · kg^{-1} · 10^{-2}	1.75 ± 0.34	2.06 ± 0.31†
Respiratory rate, bpm	31 ± 8	30 ± 6
Maximal exercise		
Heart rate, bpm	191 ± 8	195 ± 6
Stroke index, mL · m^{-2}	56 ± 9	62 ± 11
Cardiac index, L · min^{-1} · m^{-2}	10.61 ± 1.62	12.00 ± 2.20†
Minute ventilation, L · min^{-1} · m^{-1}	52.7 ± 12.4	53.6 ± 8.8
Tidal volume, mL · kg^{-1} · 10^{-2}	3.02 ± 0.44	3.46 ± 0.43†
Respiratory rate, bpm	53 ± 8	56 ± 9

Abbreviation: bpm, beats per minute.
*Data are given as mean ± SD.
†P<.05.

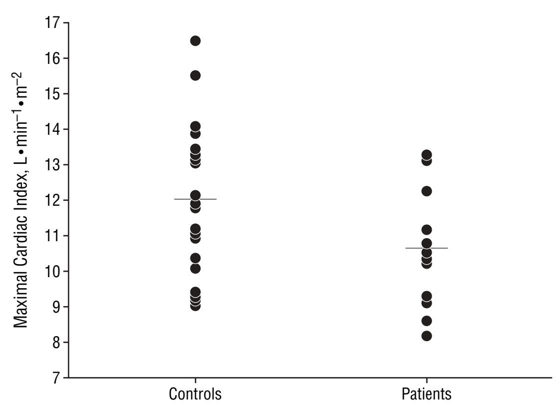

Figure 2. Maximal exercise cardiac index estimated using Doppler echocardiography in 12 patients with pectus excavatum deformity and 20 control subjects. Horizontal bars indicate means.

COMMENT

The findings from this study are generally consistent with those of previous studies: as a group, patients with a significant pectus excavatum deformity of the chest demonstrate lower endurance fitness than controls. Even in this study of a relatively small cohort of 12 patients with pectus excavatum, mean PWC (relative to body mass) on a progressive cycling test was 16.4% lower than that of control subjects. At the same time, although statistically significant, the difference in mean values of endurance times of patients with pectus deformity and control subjects in such tests is not large, with wide intersubject variability. In fact, many individuals with pectus excavatum deformity have not only normal but even above-normal fitness levels compared with controls (Figure 1). From a clinical standpoint, concluding that any complaint of exercise intolerance in a given patient is related to their pectus deformity is problematic.

In this study, use of the Haller index as a marker of the severity of pectus deformity was not predictive of either endurance fitness or maximal cardiorespiratory variables. This finding is consistent with previous studies of the failure of severity of pectus deformity to correspond to symptoms of exercise intolerance,[22] cardiorespiratory variables,[4,23] or measured fitness.[4,8] However, Malek et al[5] found that patients with pectus excavatum deformity with a Haller index greater than 4.0 were 8 times more likely to demonstrate a low "metabolic threshold" (oxygen uptake above which a sustained increase in blood lactate concentration is observed) during exercise than those with an index of 4.0 or less.

Depressed endurance fitness in young people with pectus excavatum deformity could reflect lower levels of habitual activity or sports participation, cardiac or respiratory limitations, or the influence of some other factor independent of cardiorespiratory function. This study provides some insights into this issue.

DEPRESSED AEROBIC FITNESS FROM SEDENTARY LIFESTYLE

Patients with pectus deformity might be more reticent to engage in physical activities than their peers for psychological reasons. Moreover, the pattern of diminished stroke index and cardiac index observed in this study is consistent with that observed in healthy youths with low fitness.[24] However, involvement in sports teams and physical activity level by questionnaire were similar among patients with pectus deformity and control boys in this study. Eighteen of the 21 patients with pectus excavatum studied by Malek et al[5] had been performing regular aerobic activity 30 minutes to 2 hours daily an average of 3 times a week. Despite this, their average maximum oxygen consumption was 75% of predicted, and their oxygen pulse (an indicator of stroke volume) was significantly lower than reference values. This information suggests that depressed endurance fitness in youths with pectus deformity cannot be explained by lower habitual physical levels.

IMPAIRED CARDIAC FUNCTION

Previous investigations have provided a solid basis for the concept that lower cardiac stroke volume (presumably from smaller, compressed ventricles) in patients with pectus deformity is responsible for depression in maximal cardiac output, and this in turn accounts for their lower maximum oxygen consumption and endurance fitness. The present study, which measured cardiac output and stroke volume at peak exercise, supports this idea. The maximal cardiac index was significantly lower in patients with pectus excavatum, and stroke index and cardiac index values at peak exercise correlated well with endurance performance (PWC per kilogram). Thus, the diminished cardiac index was a reflection of a consistently lower stroke index during rest and exercise in these patients (although the stroke index differences from controls did not reach statistical significance).

Cardiac findings in the present study mimic those in the study by Zhao et al.[4] These investigators compared submaximal stroke values (using Doppler echocardiography) at moderate exercise intensity in 13 patients with pectus excavatum (aged 10-31 years) with those in a height- and weight-matched control group. Mean stroke volumes at rest were 46 mL in patients and 50 mL in controls. During upright exercise, these values increased to 55 mL in the pectus deformity group and 65 mL in controls. During supine exercise, there were no differences in stroke volume between the 2 groups, suggesting that "upright exercise capacity in this disease is affected by reduced filling of the heart in the nonsupine position."[4(p163)]

Beiser et al[7] demonstrated the same postural-dependent response of stroke volume and cardiac output to submaximal exercise in 6 adults with pectus deformity during cardiac catheterization. Wynn et al[10] measured cardiac output using the acetylene rebreathing technique during maximal exercise testing in 12 children with pectus excavatum deformity. Comparison of maximal cardiac values with normative data was not reported. The rate of increase in cardiac output and stroke volume with respect to oxygen consumption during exercise was similar in controls, but values were in the low range of predicted.

VENTILATORY LIMITATIONS

Previous studies have provided evidence of a variety of ventilatory deficiencies in patients with pectus deformities that might prove limiting to exercise performance. In the present study, tidal volume (adjusted for body mass) was significantly reduced at rest, during submaximal exercise, and during maximal exercise in patients with pectus deformity compared with controls. However, this did not impair minute ventilation, which was similar to that of controls at all measurements, including peak exercise.

These results are consistent with those of Haller and Loughlin,[2] who found no differences in respiratory rate or minute ventilation at maximal exercise in 36 teenagers with pectus deformity. In that study, 58% of the patients had subjective complaints of exercise intolerance, but treadmill endurance time and maximum oxygen consumption were no different than those of controls. Similarly, the patients with pectus deformity studied by Malek et al[5] had normal maximal exercise values for minute ventilation, respiratory rate, and tidal volume despite a maximum oxygen consumption of 75% of predicted.

Other researchers, however, have described findings that suggest a possible ventilatory limitation of exercise capacity in these patients. Borowitz et al[25] found that minute ventilation at maximal exercise was only 60% of predicted in their study of 10 boys with pectus deformity. Minute ventilation was 59 ± 14 L · min^{-1} at maximal upright exercise in the patients reported by Zhao et al[4] compared with 76 ± 23 L · min^{-1} in controls.

SOMETHING ELSE

As noted previously herein, cardiorespiratory physiologic measures do not always correspond to descriptions of exercise intolerance in patients with pectus excavatum deformity. This raises the possibility that other influences (peripheral skeletal muscle changes or altered psychological perceptions of exercise discomfort) might be involved. In the present study, for example, is it possible that the reduced exercise capacity and lower cardiac output are not cause-and-effect but rather expressions of another undefined factor that limits exercise performance? Although the question cannot be answered definitively, the observation that stroke volume and cardiac output were lower in boys with pectus excavatum at rest and during submaximal exercise as well as during peak exercise supports a primary role of circulatory limitations on exercise capacity in these patients.

In summary, the findings from this study add to a generally consistent picture of the effect of a significant pectus excavatum deformity on cardiorespiratory fitness in youths. As a group, patients with pectus excavatum deformity have lower endurance fitness than healthy youths, but such differences are small. There is wide variability of exercise capacity in children and adolescents with a pectus deformity, and implicating a pectus defect as being responsible for claims of exercise intolerance in a given patient is difficult. It is likely that diminished circulatory responses to exercise as a result of depressed cardiac stroke volume are the major physiologic feature that limits exercise capacity in these patients.

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