Impact of Pectus Excavatum on Cardiopulmonary Function

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Background. **Pectus excavatum is classified using the Haller Index (HI) or the Correction Index. However, no correlation between the HI and CI and cardiopulmonary impairment has been described in detail.**

Methods. **This prospective cohort study included 99 otherwise healthy patients with pectus excavatum who underwent cardiopulmonary exercise testing and magnetic resonance imaging at inspiration and expiration to correlate cardiopulmonary function with the grade of thoracic dysmorphia.**

Results. **Probands with an HI exceeding 3.25 had first an increase in heart rate at anaerobic threshold (from 148.0 ± 16.0 beats/min to 155.9 ± 15.0 beats/min, $p = 0.036$), with an HI of more than 3.6 a reduction in oxygen pulse at anaerobic threshold (from 10.7 ± 2.6 mL/beat to 9.3 ± 2.9 mL/beat, $p = 0.017$), with an HI exceeding 3.8 a reduction of maximum oxygen pulse (from 13.9 ± 3.4 mL/beat to 11.9 ± 3.7 mL/beat, $p = 0.010$), and with an HI of exceeding 4.0 a decline in maximum oxygen uptake (from 43.7 ± 6.5 mL · kg^{-1} · min^{-1} to 40.4 ± 7.4 mL · kg^{-1} · min^{-1}, $p = 0.025$). The CI of more 27% reflects cardiopulmonary changes earlier than the corresponding HI exceeding 3.25 ($p = 0.01$ for maximum oxygen pulse; $p = 0.017$ for oxygen pulse at anaerobic threshold; $p = 0.015$ for heart rate at anaerobic threshold).**

Conclusions. **The inspiratory HI and CI reflect the effect of pectus excavatum on cardiopulmonary function. The cardiopulmonary system reacts first with an increase in heart rate at anaerobic threshold, followed by a decrease in stroke volume at anaerobic threshold and maximum stroke volume. Increased severity of the deformity then leads to a decrease in cardiac output.**

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Pectus excavatum (PE) is an intrusion of the anterior chest wall into the thoracic cavity. Although the physiological and psychological consequences of PE vary, the lesion is troublesome enough for a large number of patients that they welcome a corrective surgical intervention. PE belongs to the most common chest wall deformities, with an incidence of approximately 1 of every 300 to 400 white individuals [1]. The disorder has a male predominance, with a ratio of 3 to 5 males for every female affected. Although approximately 40% of PE patients are aware of relatives with the same deformity, there is no known genetic linkage [2]. For decades, surgical correction of PE was performed primarily for cosmetic and psychological indications, without proven documentation of physiological respiratory or cardiopulmonary abnormalities or even an improvement after surgical correction [3–6].

The Haller Index (HI) and Correction Index (CI) are well established for the morphologic description of PE. An HI of more than 3.0 to 3.25 and CI exceeding 27% are considered to be indications for a corrective operation [7], but no correlation between the indices and the degree of cardiopulmonary impairment has been reported [8]. PE was radiographically first classified according to the Welch Index, which was measured from a lateral chest roentgenogram at end-inspiration with barium stripes to delineate the thoracic walls from the invaginated sternum in a standing proband [9].

The HI was first published in 1987 as the "Pectus Index," which described the severity of PE from a computed tomography (CT) scan [4]. Reports comparing the HI determined from a lateral chest roentgenogram to the HI measured from a CT image are limited when the proband is supine; therefore, whether the HI can be transferred to a different imaging modality is not clear. However, no major difference between the HI measured by roentgenogram and the CT-derived HI has been demonstrated [10]. Furthermore, whether the end-inspiration or end-expiration measured indices reflect the effect on cardiopulmonary function is not known [11]. Therefore, the literature on the effect of PE on cardiopulmonary function in relation to the

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Abbreviations and Acronyms

AUC	= area under the curve
CI	= Correction Index
CI_{exp}	= end-expiratory Correction Index
CI_{insp}	= end-inspiratory Correction Index
CPET	= cardiopulmonary exercise testing
ECG	= electrocardiogram
HI	= Haller Index
HI_{exp}	= end-expiratory Haller Index
HI_{insp}	= end-inspiratory Haller Index
HR_{AT}	= heart rate at anaerobic threshold
HR_{max}	= maximum heart rate
HR_{max}%	= percentage of predicted heart rate
MRI	= magnetic resonance imaging
O_2-$pulse_{AT}$	= oxygen pulse at anaerobic threshold
O_2-$pulse_{max}$	= maximum oxygen pulse
PE	= pectus excavatum
RBB	= right bundle branch block
ROC	= receiver operating characteristics
V_{CO_2}	= exhaled CO_2
$V_{O_{2AT}}$	= oxygen uptake at anaerobic threshold
VO_{2max}	= maximum oxygen uptake
VO_{2max}%	= percentage of predicted oxygen uptake
$Watt_{max}$	= maximum physical work capacity (Watt/kg)

degree of deformity is not consistent. In addition, most of the reports could not correlate impaired cardiopulmonary function with the HI or CI [12–17], which may be related to respiration being at end-inspiration, end-expiration, or somewhere in between, giving completely different measurements and estimates of the degree of dysmorphia [11].

The purpose of this prospective study was to evaluate the influence of the severity of dysmorphia on cardiopulmonary function by following a standardized protocol and to estimate which standardized magnetic resonance imaging (MRI)–derived respiratory status and index reflects the cardiopulmonary impairment most accurately.

Material and Methods

Patients

Between September 2012 and July 2016, 108 consecutive patients with PE underwent cardiopulmonary exercise testing (CPET) and thoracic MRI in hold end-inspiration and end-expiration. CPET and MRI were performed within 1 week. The analyses excluded 9 patients with additional known cardiac, pulmonary, and neurologic diseases leading to impaired CPET results. All adult patients or legal guardians gave written informed consent. The University of Mainz Ethical Board waived the need for committee approval of studies concerning publication of results from routine examinations of patients.

MRI Protocol

All participants underwent MRI scans on a 1.5-Tesla (T) scanner (MAGNETOM Avanto, Siemens AG, Munich, Germany) or a 3-T scanner (MAGNETOM Skyra, Siemens AG), both with a gradient strength of 45 mT/m and a 32-channel body coil for signal detection. A standard T2-weighted half-Fourier acquisition single-shot turbo spin echo sequence in hold end-inspiration, and end-expiration (transverse) was used to calculate the HI and CI. Technical sequence details were described previously. Additional sequences were obtained for further evaluations not published here [18].

CPET Procedure

CPET was performed on an H/P/cosmos treadmill using CareFusion MS-CPX 5.70 software (CareFusion, Höchberg, Germany) combined with GE-electrocardiogram (ECG) Cardiosoft 6.7 (GE Healthcare, Chicago, IL). The exercise protocol was an altered, improved treadmill protocol recommended by the German Society of Pediatric Cardiology [19]. According to this protocol, the test started with a speed of 2 km/h on a flat treadmill, increasing stepwise in increments of 0.5 km/h and 3% inclination every 90 seconds to a maximum incline of 21%. Beyond this inclination, only the speed continued to increase in increments of 0.5 km/h. The testing was stopped when the calculated target heart rate (ie, 220 – age in years) was reached or when complete exhaustion occurred, as indicated by the patient giving a hand sign. The end of the exercise test protocol consisted of a recovery phase on a flat treadmill at a speed of 2 km/h.

Maximum oxygen uptake ($V_{O_{2max}}$ [mL · kg^{-1} · min^{-1}]), percentage of predicted oxygen uptake ($V_{O_{2max}}$% [%]), oxygen uptake at anaerobic threshold ($V_{O_{2AT}}$ [mL · kg^{-1} · min^{-1}]), maximum oxygen pulse (O_2-$pulse_{max}$ [mL/beat]), percentage of predicted oxygen pulse (O_2-$pulse_{max}$% [%]), oxygen pulse at anaerobic threshold (O_2-$pulse_{AT}$ [mL/beat]), maximum physical work capacity (Watt/kg), maximum heart rate (HR_{max} [beats/min]), percentage of predicted heart rate (HR_{max}% [%]), and heart rate at anaerobic threshold (HR_{AT} [beats/min]), were evaluated and compared against calculated reference values based on age, sex, height, and weight [20]. The anaerobic threshold was calculated by the V-slope method in which exhaled CO_2 (V_{CO_2}) is plotted against oxygen uptake (V_{O_2}) on *x*–*y* coordinates of equal scale. Cardiopulmonary impairment in CPET was defined as a reduction in $V_{O_{2max}}$% of less than 85% of the predicted value [21]. Level of training was estimated by calculation of $V_{O_{2AT}}$/predicted$V_{O_{2max}}$. A level of more than 40% was considered to be normal.

Baseline electrocardiograms were analyzed for signs of right atrial enlargement, defined as a p-wave exceeding 0.3 mV in II; conduction disturbances, such as incomplete (defined as rsr′ or rsR′ with a QRS duration < 120 ms), or pathologic (defined as Rsr′ with a QRS duration > 120 ms) right bundle branch block (RBBB); and right ventricular hypertrophy (defined as S > 0.5 mV in V_6) [22, 23]. An experienced cardiologist performed all CPETs using the same standardized protocol.

Statistical Analysis

Statistical analyses were performed using SPSS 24 software (IBM, Armonk, NY). The Mann-Whitney *U* test was