ORIGINAL ARTICLE / ÖZGÜN MAKALE

Is there a relationship between Haller Index and cardiopulmonary function in children with pectus excavatum?

Pektus ekskavatumlu çocuklarda Haller İndeksi ile kardiyopulmoner fonksiyon arasında bir ilişki var mıdır?

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ABSTRACT

Background: This study aims to systematically examine the cardiopulmonary functions in children with pectus excavatum and to compare the obtained findings with the Haller Index.

Methods: Between September 2017 and June 2018, medical records of a total of 31 patients (27 males, 4 females; mean age: 14.8 ± 2.0 years; range, 9 to 18 years) with pectus excavatum were retrospectively analyzed. The patients were divided into Group 1 (<2.5), Group 2 (2.5 to 3.19), and Group 3 (>3.2) according to the Haller Index. All groups were systematically evaluated based on pulmonary function tests and echocardiography. Forced vital capacity, forced expiratory volume in 1 second, and the forced expiratory volume in 1 second/forced vital capacity ratio were calculated. Left ventricular end-diastolic diameter, ejection fraction, mitral valve prolapses, and right ventricular cavity in the apical four-chamber position were evaluated with echocardiography.

Results: Of the patients, 19.4% were in Group 1, 38.7% in Group 2, and 41.9% in Group 3. The mean Haller Index value was 3.09 ± 0.64 . According to pulmonary function test results, 16.1% of the patients had restrictive disease and 6.5% had obstructive disease. There was a negative correlation between the index and forced expiratory volume in 1 second and forced vital capacity, and there was a statistically significant decrease in these values, as the Haller Index increased (p<0.017). There was a significant difference in the ejection fraction among the groups (p<0.001) and, as the Haller Index increased, ejection fraction statistically significantly decreased.

Conclusion: Our study results show a negative correlation between the severity of pectus excavatum and pulmonary dysfunction and, as the severity increases, left ventricular function may be affected by the deformity. As a result, there seems to be a significant relationship between the severity of the deformity and cardiopulmonary functions.

Keywords: Pectus excavatum, respiratory function tests, transthoracic echocardiography.

ÖΖ

Amaç: Bu çalışmada pektus ekskavatumlu çocuklarda kardiyopulmoner fonksiyonlar sistematik olarak incelendi ve elde edilen bulgular Haller İndeksi ile karşılaştırıldı.

Çalışma planı: Eylül 2017 - Haziran 2018 tarihleri arasında pektus ekskavatumlu toplam 31 hastanın (27 erkek, 4 kız; ort. yaş: 14.8 ± 2.0 yıl; dağılım, 9-18 yıl) tıbbi kayıtları retrospektif olarak incelendi. Hastalar Haller İndeksine göre Grup 1 (<2.5), Grup 2 (2.5-3.19) ve Grup 3'e (>3.2) ayrıldı. Tüm gruplar solunum fonksiyon testleri ve ekokardiyografiye göre sistematik olarak değerlendirildi. Zorlu vital kapasite, 1. saniye zorlu ekspiratuvar volüm ve 1. saniye zorlu ekspiratuvar volüm/zorlu vital kapasite oranı hesaplandı. Sol ventrikül diyastol sonu çapı, ejeksiyon fraksiyonu, mitral kapak prolapsusu ve apikal dört boşluk pozisyonunda sağ ventrikül kavitesi ekokardiyografi ile değerlendirildi.

Bulgular: Hastaların %19.4'ü Grup 1, %38.7'si Grup 2 ve %41.9'u Grup 3'te idi. Ortalama Haller İndeks değeri 3.09 ± 0.64 idi. Solunum fonksiyon testinin sonuçlarına göre, hastaların %16.1'inde restriktif hastalık ve %6.5'inde obstrüktif hastalık izlendi. Birinci saniye zorlu ekspiratuvar volüm ve zorlu vital kapasite arasında negatif bir ilişki vardı ve Haller İndeksi arttıkça bu değerlerde istatistiksel olarak anlamlı bir düşüş görüldü (p<0.017). Gruplar arasında ejeksiyon fraksiyonu açısından anlamlı bir fark vardı (p<0.001) ve Haller İndeksi arttıkça ejeksiyon fraksiyon istatistiksel olarak anlamlı düzeyde düştü.

Sonuç: Çalışma sonuçlarımız pektus ekskavatumun şiddeti ve pulmoner fonksiyon bozukluğu arasında negatif bir ilişki olduğunu ve şiddet arttıkça sol ventrikül fonksiyonunun deformiteden etkilenebileceğini göstermektedir. Sonuç olarak deformitenin şiddeti ile kardiyopulmoner fonksiyonlar arasında anlamlı düzeyde bir ilişki olduğu görülmektedir.

Anahtar sözcükler: Pektus ekskavatum, solunum fonksiyon testleri, transtorasik ekokardiyografi.

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Cite this article as: Katrancioglu O, Ozgel M, Inceoglu F, Katrancioglu N, Sahin E. Is there a relationship between Haller Index and cardiopulmonary function in children with pectus excavatum?. Turk Gogus Kalp Dama Pectus excavatum (PE) is the most common anterior chest wall deformity.^[1] It is caused by dorsal deviation of the sternum and underlying costal cartilages and has an estimated prevalence of 1.2%.^[2] Although, for many years, these deformities were considered only as an aesthetic problem, there has been increasing evidence of functional limitation recently. Numerous studies have shown that airway obstruction increases with age and also has varying degrees of restrictive patterns.^[3,4] Additionally, in some clinical studies, it has been demonstrated that patients with PE deformity have limited exercise tolerance due to both decreased thoracic volume and cardiac compression.^[5,6]

Although there are studies examining the relationship between Haller Index (HI) and the severity of the deformity in the literature, there are few studies evaluating the relationship between the HI and cardiopulmonary functions (CPFs) in children.^[7] In the present study, we, therefore aimed to systematically examine CPFs in pediatric patients with PE and to compare the results obtained with the HI.

PATIENTS AND METHODS

This single-center, retrospective study was conducted at Turgut Özal University Faculty of Medicine, Department of Thoracic Surgery between September 2017 and June 2018. Medical records of a total of 31 patients (27 males, 4 females; mean age: 14.8±2.0 years; range, 9 to 18 years) were reviewed. All patients were systematically evaluated based on pulmonary function test (PFT) and cardiac echocardiography (ECHO). Patients with isolated congenital PE who did not receive conservative or surgical treatment previously were included in the study. Those with pulmonary or cardiovascular abnormalities and missing data were excluded from the study.

The HI was calculated by dividing the transverse diameter of the chest by the distance between the posterior aspect of the sternum at the deepest point of the sternum and the anterior aspect of the vertebrae by thoracic computed tomography (CT) in all patients.^[7]

Pulmonary function tests examination

The PFTs at rest were performed in all patients using a spirometer device (Cosmed, Quark PFT 1, Rome, Italy). In this study, the forced vital capacity (FVC), forced expiratory volume in 1 second (FEV1), and FEV1/FVC ratio were calculated. According to PFT results, those without airway disease had a FVC of >80%, FEV1 of >80%, and FEV1/FVC of >70%. Those with decreased FVC, FEV1, and FEV1/FVC ratios were classified as obstructive. Those with decreased FEV1 and FVC values with normal or increased FEV1/FVC ratios were classified as restrictive airway disease.^[7]

Echocardiography examination

Transthoracic M-mode and two-dimensional ECHO (Vivid 5, GE Ultrasound, USA) was used to evaluate left ventricular end-diastolic diameter (LVEDV), left ventricular ejection fraction (LVEF), and mitral valve prolapse (MVP) in the parasternal long-axis position, and the right ventricular cavity was evaluated in the apical four-chamber position. For the diagnosis of MVP, systolic displacement of a leaflet into the left atrium below the plane of the mitral annulus was used as a criterion in the ECHO examination performed by a pediatric cardiologist. A LVEF of <60% was considered as LV systolic dysfunction.

Statistical analysis

Statistical analysis was performed using the IBM SPSS version 25.0 software (IBM Corp., Armonk, NY, USA). The Shapiro-Wilk test was used to check whether the data included in the study fit the normal distribution.^[8] Since the variables did not have a normal distribution, the analysis was continued with non-parametric test methods. For the comparisons in independent dual groups, since the assumption of normality was not provided, the Mann-Whitney U test was used. The Kruskal-Wallis test analysis was performed for comparisons in independent multiple groups. Since the p value would increase depending on the increase in the number of comparisons in the variables with a difference, the Bonferroni corrected p value was used.^[9] The Spearman rank correlation coefficient was used.^[10] When one of the variables was categorical and the other was continuous, the point double series correlation value was calculated. At this value, the η (Eta) statistic was calculated. A p value of <0.05 was considered statistically significant.

RESULTS

Patient characteristics

The most common complaints were chest pain and shortness of breath. According to HI, the patients were divided into three groups: Group 1 (<2.5), Group 2 (between 2.5 and 3.19), and Group 3 (>3.2). Groups 1, 2, and 3 had 19.4%, 38.7%, and 41.9% of the patients, respectively. The reason why most of the patients were in Groups 2 and 3 was that more operable patients were referred to the surgical outpatient clinic. A statistically significant difference

		Female		Male			Total			
Variables	n	%	Mean±SD	n	%	Mean±SD	n	%	р	
Age (year)			12.5±2.7			15.2±1.7	31	100	0.036*	
Body mass index			0.17±0.0			0.19±0.0	31	100	0.175	
Airway disease									0.397	
Restrictive	1	100		4	66.7		5	71.4		
Obstructive	-	-		2	33.3		2	28.6		
Family history									0.015*	
Yes	3	75.0		4	14.8					
No	1	25.0		23	85.2					
Haller Index									0.752	
Group 1	1	25.0		5	18.5					
Group 2	2	50.0		10	37.1					
Group 3	1	25.0		12	44.4					

Table 1. Demographic characteristics of patients

SD: Standard deviation; Mann-Whitney U test p value; statistical significance, * p<0.05; There is a statistically significant difference between the groups.

was found between boys and girls according to age (p=0.036) and family history (p=0.015) variables. There was no statistically significant difference between boys and girls in terms of airway disease (p=0.397), HI (p=0.752), and body mass index (BMI) (p=0.175). The demographic characteristics of the patients are summarized in Table 1.

Pulmonary functions

According to PFT results, 16.1% of the patients had restrictive disease, 6.5% had obstructive disease, and 77.4% had no pulmonary dysfunction. It was evaluated whether there was a significant difference among the groups according to the FVC, FEV1, and

FEV1/FVC variables and the results are given in Table 2.

A statistically significant difference was found among the groups in FVC (p=0.008) and FEV1 values (p<0.001). According to statistical analysis, in the FVC and FEV1 values, there was a statistically significant difference between Groups 1 and 3 (p<0.017). A statistically significant difference was also found between Groups 2 and 3 (p<0.017). There was a negative correlation between the index and FEV1 and FVC (Figure 1). As the HI increased, there was a statistically significant decrease in FEV1 and FVC values (Table 2).

Measurement	Mean±SD	Median	Min-Max	Test	р	Difference		
FVC				9.718	0.008*	1-3.2-3		
Group 1	102±11.5	98	88-117					
Group 2	95.4±10.0	92	80-119					
Group 3	84.2±13.3	82	65-119					
FEV1				17.157	<0.001*	1-3.2-3		
Group 1	107.3±12.9	102	95-128					
Group 2	102.2±9.3	102.5	86-122					
Group 3	83±9.6	85	70-102					
FEV1/FVC				1.928	0.381	-		
Group 1	104 ± 4.9	102.5	98-112					
Group 2	105.1±5.1	104	99-118					
Group 3	95.6±15.2	102	65-112					

Table 2.	Comparison	of respirator	v functions	according to	aroups
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SD: Standard deviation; FVC: Forced vital capacity; FEV1: Forced expiratory volume in 1 second. Kruskal-Wallis test; * p<0.05.



Figure 1. FEV1 and HI were found to be strongly negative correlated, and FVC and HI were found to be moderately significantly correlated (p<0.05).

FEV1: Forced expiratory volume in 1 second; HI: Haller Index; FVC: Forced vital capacity.

Although it was not seen in Groups 1 and 2, 53.8% of the patients in Group 3 had pulmonary dysfunction. There was a statistically significant difference among the groups in terms of the incidence of pulmonary dysfunction (p<0.05).

No statistically significant difference was found between those with restrictive and obstructive diseases according to the HI (p>0.017). However, the index was higher in patients with restrictive airway disease. When the groups with restrictive airway disease and those with normal respiratory functions were compared according to the HI, a statistically significant difference was found (p<0.017). When the groups with obstructive airway disease and those with normal respiratory functions were compared according to the HI, no statistically significant difference was found (p>0.017), but the index was found to be higher in those with obstructive airway disease compared to the normal group. If the HI value increased by one point, the risk of developing restrictive and obstructive airway disease would increase 0.646 times (p=0.001). Increases in the HI were found to be associated with airway diseases (Table 3).

Cardiac functions

According to the ECHO findings, the mean LVEF was $72.3\pm4.1\%$ in Group 1, $65.6\pm2.5\%$ in Group 2, and $59.5\pm4.2\%$ in Group 3 (Table 4). There was a significant difference among the groups in terms of LVEF (p<0.001) and, as the HI increased, the LVEF statistically significantly decreased (Figure 2). However, no statistically significant difference was

Measurement	Mean±SD	Median	Min-Max	Test	р	Eta (η)
Haller Index				13.737	0.001*	0.646
Restrictive airway disease	4.2±0.2	4.2	4-4.4	(1-2 p=0.095,		
Obstructive airway disease	3.3±0.1	3.25	3.2-3.3	1-3 p=0.001*,		
Respiratory functions normal	2.8±0.4	2.8	2.2-3.7	2-3 p=0.185)		

SD: Standard deviation; Mann-Whitney U test; * p<0.05.

Table 4. Comparison of ejection fraction according to gre	oups
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Measurement	Mean±SD	Median	Min-Max	Test	р	Difference
Ejection fraction				18.978	< 0.001*	1-2, 1-3, 2-3
Group 1	72.3±4.1	74.5	65-75			
Group 2	65.6±2.5	65	60-70			
Group 3	59.5±4.2	60	55-70			

SD: Standard deviation; Kruskal-Wallis test; * p<0.05.



Figure 2. A statistically significant negative correlation was found between ejection fraction and Haller Index (p<0.05).

found in the LVEDV. Mitral valve prolapse was detected by ECHO in only four (12.9%) of the patients.

DISCUSSION

Anterior chest wall deformities are the result of abnormal development of costal cartilage due to genetic predisposition or idiopathic mechanisms.^[11] It is the most common PE and, although there is no known genetic predisposition, approximately 40% of patients have relatives with the same deformity.^[6] There are studies in the literature that are associated with family history at varying rates (30 to 65%).^[12,13] In our study, only seven children (22.5%) had a positive family history. This rate was lower compared to the literature, suggesting that it may be due to family members hiding their deformities.

Studies in the literature have shown that PE is more common in males.^[3,7] In our study, it was more common in males (87.1%), consistent with the literature. No statistically significant relationship was found between sex and HI.

Although PE is primarily considered an aesthetic problem, it also has effects on CPF. The deformity may result in heart compression and a reduction in thoracic volume, thereby worsening CPF. Early childhood has a lower prevalence of symptoms, which increases with age.^[7,14] These patients can be seen with chest pain, dyspnea, palpitations, and frequent upper respiratory tract infections.^[7] In a large-volume study in the literature, 62% reported dyspnea and 32% had chest pain at rest.^[15] In a recent study by Ramadan et al.,^[16] 13.3% of patients reported dyspnea while exercising and 20% reported chest pain at rest. The most common complaints in our study were chest pain and shortness of breath, particularly with

exercise. In contrast to the literature, the HI and complaints did not increase with age.

The HI is one of the most widely used radiological methods to rate the severity of PE. It is calculated by dividing the transverse diameter of the chest by the anterior-posterior diameter at the deepest depression level in CT images.^[17] In our study, the patients were divided into three groups using this index. Group 1 patients had a mild deformity, whereas Group 3 patients had a severe deformity.

Lung expansion is limited in patients with PE due to the change in thoracic volume during inspiration. Therefore, the PFT must be performed in patients during static and/or exercise to identify the effects of deformity on respiration. Although static PFT is less sensitive than dynamic PFT in detecting restrictive airway disease, it is extremely useful in detecting a significant decrease in FVC and maximum ventilation volume.^[7] Most patients with PE have decreased static pulmonary function.^[18] In a study, less than 2% of patients diagnosed with PE had obstructive (FEV1/FVC <67%) and 14.5% had restrictive (FEV1 <80%, FVC <80%, FEV1/FVC >80%) airway disease.^[19] In another study, low lung volumes were found in 23% of PE patients.^[16] Consistent with the literature, in our patient group, the FVC and FEV1 values were found to be below normal in Groups 2 and 3. In our study, the values of FEV1 and FVC statistically significantly decreased, as the HI increased. Similar to the literature. restrictive airway disease was seen in 16.1% of the patients, and all patients were in Group 3. These results can be explained by the decrease in mechanical compliance and the effect of respiratory muscles in PE patients. In addition, increased chest wall rigidity may have caused these results.

Although deformities in the anterior chest wall lead to difficulties in the evaluation of the heart with ECHO, it is usually preferred to evaluate cardiac functions with ECHO.^[20] In our study, the cardiac functions of the patients were evaluated with ECHO. The prevalence of MVP in patients with isolated PE is reported to be between 15 and 55% in the literature.^[21,22] In our study, MVP was found at a lower rate compared to the literature. No significant relationship was found between the HI and presence of MVP.

The percentage of LVEF is very important in the evaluation of cardiac functions and a LVEF below 55% indicates low cardiac function in this age group.^[16] Right atrial and ventricular compression can be seen in patients with PE, particularly in those

with severe deformities, and there may be changes in the shape and function of the right ventricle. This situation may affect not only the right ventricle, but also the LV function.^[23] Since the right and LVs share a common septum and are in the same pericardial cavity, they are anatomically integrated with each other. Compression of the right side of the heart in PE patients may also cause functional changes in the LV, resulting in cardiac dysfunction in patients with PE.^[23,24] In our study, although LEVF values remained within the normal limits in patients with severe PE (Group 3), a significant decrease was observed compared to patients with Group 1 and 2 deformities. According to our results, there was a statistically significant correlation between the HI and LVEF, indicating that a rise in the HI value can affect the heart, as well as the respiratory system.

Nonetheless, there are some limitations to this study. First, it has a single-center, retrospective design with a relatively small sample size and without a control group. Second, the evaluation was able to be performed at rest only. The same evaluations during exercise may help to better understand the anatomical role of pectus pathophysiology. Third, since the postoperative CPF were unable to be assessed, the inability to evaluate the relationship between preoperative HI severity and physiological recovery after repair is another limitation.

In conclusion, our study results show that pectus excavatum primarily affects right heart functions, and progressive Haller Index also affects left heart and respiratory functions. We believe that further multicenter, large-scale studies on this subject may lead to a recovery of cardiopulmonary functions, if pectus excavatum correction is performed in patients with lower Haller Index.

Ethics Committee Approval: The study protocol was approved by the Malatya Turgut Ozal University Faculty of Medicine Ethics Committee (date: 09.08.2022, no: 2022/132). The study was conducted in accordance with the principles of the Declaration of Helsinki.

Patient Consent for Publication: A written informed consent was obtained from each patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Idea/concept: O.K., N.K., E.S.; Design: O.K.; Control/supervision: E.S., N.K.; Data collection and/or processing: O.K., M.O., F.I.; Analysis and/or interpretation: E.S., N.K.; Literature review: O.K., N.K., F.I.; Writing the article: O.K., N.K.; Critical review: N.K., E.S.; References: M.O.; Materials: M.O. **Conflict of Interest:** The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

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