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ASSESSMENT OF CARDIAC ANOMALIES IN PEDIATRIC PATIENTS WITH PECTUS CARINATUM

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Abstract: Pectus carinatum (PC) is an anterior thoracic visible deformity manifested by protrusion of the sternum. It can be an isolated chest deformity or it can be seen together with genetic pathologies. We analyzed the cardiac findings of 40 children who were diagnosed with pectus carinatum at Kahramanmaras Sutcu Imam University's Thoracic Surgery Outpatient Clinic between 1 February 2021 and 1 February 2022 and 97 healthy controls in similar age groups. The Pectus Carinatum patients had higher rates of cardiac malposition, MVP (Mitral valve prolapse), MI (mitral insufficiency), TVP (tricuspid valve prolapse), CHD (congenital heart disease), and aortic valve pathology**.** Our study showed that the prevalence of cardiac pathologies in the pediatric PC patients was higher than that in the control group consisting of healthy children. Thus, it may be recommend the referral of pediatric PC patients to cardiology outpatient clinics for the early diagnosis of potential cardiac pathologies.

Key words: Pectus carinatum, Mitral valve prolapse, Cardiac anomaly, Mitral insufficiency

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1. Introduction

PC, which is a congenital deformity of the anterior chest wall, is a deformity that involves the protrusion of the sternum and generally results in the prominence of the 3rd-7th ribs, which also results from excessive growth of the costal cartilages relative to the ribs. PC is the second-most prevalent anterior chest wall deformity after pectus excavatum (Park et al., 2013; Martinez et al., 2019; Pink et al., 2021)

PC can be bilateral or unilateral, and the sternum is usually displaced towards the side of the deformity. It can be an isolated chest deformity, or it can often be seen together with genetic pathologies such as Marfan syndrome and Noonan syndrome (Dogan and Sert, 2023). Approximately 25% of cases have genetic predisposition in their families (Fonkalsrud and Beanes, 2000). In some cases, chest pain and shortness of breath with physical activity may be seen (Balcı and Cakmak, 2018). There is a congenital heart disease in approximately 18% of cases (Blanco et al., 2011)

We observed that valve anomaly rates are high in patients who presented to the thoracic surgery outpatient clinic due to PC. Therefore, in this study, we aimed to compare the echocardiographic findings of pediatric PC patients to healthy controls without chest deformity and discuss the results in light of the literature.

2. Materials and Methods

This study was conducted in compliance with the Declaration of Helsinki. Before starting the study, ethical approval was obtained from the Ethics Committee for Noninterventional Clinical Studies of Kahramanmaras Sutcu Imam University on January 25, 2021 (Meeting No. 2021/04, Decision No. 03). Additionally, informed consent was obtained from the families of the participants.

2.1. Design

The study was initiated by examining cardiac findings in 40 children diagnosed with PC and 97 healthy children in similar age groups as controls between 1 February 2021 and 1 February 2022, in the University's Thoracic Surgery Outpatient Clinic. Routine posteroanterior (PA) and lateral chest X-rays of all patients were taken. Each patient was examined for cardiac pathologies by a Pediatric Cardiology specialist using echocardiography (ECHO). The demographic characteristics, admission complaints, and ECHO findings of the patients were recorded.

The echocardiographic examinations of all patients were made using a Vivid 7 Pro echocardiography device (GE Healthcare, Vingmed Ultrasound AS, California, United States) with a suitable cardiac sector probe based on the age and weight of the patient when the patient was laid in the left lateral decubitus position. Offline analysis was carried out using the workstation Echopac PC'08 version

7.0.0 GE Vingmed Ultrasound (GE Healthcare, California, United States), and the measurements were consistently obtained by the same physician. The patients were examined using conventional ECHO methods according to the pediatric ECHO guidelines of the American Society of Echocardiography (Lai et al., 2006). Patients who were being followed up for congenital heart disease diagnosed before the start of the study were excluded.

2.2. Statistical Analyses

The SPSS (IBM, Armonk, New York, United States, version 22) for Windows software was used for statistical analysis. The variables are presented as frequency (n) — percentage (%) and mean ± standard deviation values. The normal distribution of the variables was tested using the Kolmogorov-Smirnov test. The normally distributed parameters were analyzed using one-way analysis of variance or Student's t-test, while the Kruskal-Wallis test or the Mann-Whitney U test was used for the numeric variables that did not show normal distribution. The risk factors were evaluated with univariate and multivariate logistic regression models. The variables that were found to be significant in the univariate analyses were included in the logistic regression analysis. A P value smaller than 0.05 was considered statistically significant.

3. Results

The study included a total of 137 patients, including 39 (28.5%) female and 98 (71.5%) male patients. The mean age of the patients was 10.51 ± 4.19 (3.21–17.9) years. There was no statistically significant difference between the PC and control groups in terms of sex and age (respectively, P=0.265 and P=0.506) (Tables 1 and 2). Male patients accounted for 80% of the PC group and 68% of the control group. While 24 of the PC patients (60%) did not report a complaint at the time of presenting to the hospital, 10 (25%) reported chest pain, and 6 (15%) reported palpitations. Among the PC patients, 37 (92.5%) had chondrogladiolar-type prominence, 2 (5%) had mixed-type prominence, and 1 (2.5%) had chondromanubrial-type prominence.

In the comparison of the cardiac findings of the PC patients and the healthy control group, we determined significantly higher rates of cardiac pathologies in the PC group (P<0.001). The PC group had significantly higher rates of cardiac malposition, mitral valve prolapse (MVP), mitral insufficiency, tricuspid valve prolapse (TVP), congenital heart disease, and aortic valve pathology than the control group (respectively, P<0.001, P<0.001, P<0.001, P<0.001, P=0.041, and P=0.012) (Table 1).

Table 1. Comparison of the cardiological findings of the pectus carinatum patients and the healthy controls

		Control (97) n–%	Pectus Carinatum (40) n–%	P
Sex	Female	31–32	8–20	0.158
	Male	66–68	32–80	
Cardiac pathology		7–7.2	23–57.5	< 0.001
Cardiac malposition		0–0	5–12.5	< 0.001
MVP		5–5.2	18–45	< 0.001
Mitral regurgitation		3–3.1	13–32.5	< 0.001
TVP		0–0	9–22.5	< 0.001
CHD		1–1	3–7.5	0.041
Aortic valve pathology		2–2.1	5–12.5	0.012

MVP= mitral valve prolapse; TVP= tricuspid valve prolapse; CHD= congenital heart disease.

In the comparison of the ECHO findings, no significant difference was found between the PC and control groups in terms of their age, right ventricle end-diastole Z-score, diastolic interventricular septum diameter Z-score, end-diastolic left ventricle posterior wall thickness Z-score, ejection fraction, shortening fraction, main pulmonary artery diameter Z-score, mitral annulus Z-score, tricuspid annulus Z-score, left atrium diameter Z-score, tricuspid annular plane systolic excursion Z-score, or pulmonary blood flow velocity (respectively, P=0.151, P=0.248, P=0.098, P=0.884, P=0.120, P=0.097, P=0.628, P=0.429, P=0.807, P=0.732, P=0.118, and P=0.169). The left ventricle end-diastole internal diameter Z-score, left ventricle end-systole internal diameter Z-score, aortic sinus Z-score, and ascending aorta Z-score values of the PC group were significantly higher than those of the control group (respectively, P=0.005, P=0.001, P=0.013, and P=0.031) (Table 2).

In the results of the risk analysis that we conducted with the logistic regression analysis method for MVP and TVP development in the PC patients, we found that MVP development increased 15.05-fold, and TVP development increased 27.87-fold in the PC patients (respectively, P<0.001 and P<0.001) (Table 3).

Table 2. Comparison of echocardiographic findings of the pectus carinatum patients and the healthy control group

	Control (97) χ ± SD	Pectus carinatum (40) χ ± SD	P
AGE	0.18 ± 4.21	11.32 ± 4.07	0.151
RVDD ZS	0.958 ± 0.385	1.055 ± 0.431	0.248
IVSd ZS	0.390 ± 0.440	0.535 ± 0.5 15	0.098
LVIDd ZS	-0.318 ± 0.535	0.039 ± 0.711	0.005
LVPWd ZS	0.314 ± 0.499	0.330 ± 0.599	0.884
LVIDS ZS	-0 .590 ± 0 .574	-0.241 ± 0 .585	0.001
EF	72.50 ± 3 .73	71.55 ± 3 .24	0.120
FS	41.41 ± 3.27	40.42 ± 2.80	0.097
Ao rt ZS	-0 .588 ± 0.734	0.033 ± 1.352	0.013
MPA ZS	-0 .142 ± 0.583	-0.207 ± 0 .715	0.528
Mannulus ZS	-0 .475 ± 0 .454	-0.374 ± 0.745	0.429
Tannulus ZS	-0 .489 ± 0.484	-0.455 ± 0. 544	0.807
LA ZS	0.554 ± 0.584	0.704 ± 0.509	0.732
TAPSE ZS	-0.298 ± 0.821	-0.537 ± 0.750	0.118
Ascending Aorta ZS	-0 .395 ± 0 .814	0.023 ± 1.119	0.031
Pulmonary flow	1.050 ± 0.055	1.032 ± 0.071	0.159

RVDD ZS= end-diastole right ventricle diameter Z-Score; IVSd ZS= diastolic interventricular septum diameter Z-Score; LVIDd ZS= end-diastole left ventricle diameter Z-Score; LVPWd ZS= left ventricular posterior wall thickness Z score; LVIDS ZS= end-systole left ventricle diameter Z-Score; EF= ejection fraction; KF= shortening fraction; Aort ZS= aortic sinus Z-Score; MPA ZS= main pulmonary artery diameter Z-Score; LA ZS= left atrium diameter Z-Score; TAPSE ZS= tricuspid annular plane systolic excursion Z-Score.

Table 3. Risk analysis for mitral and tricuspid valve prolapse in the pectus carinatum patients

	OR	95% CI	P
MVP	15.05	5.039–44. 981	< 0.001
TVP	27.87	3.395–228 .805	< 0.001

MVP= mitral valve prolapse; TVP= tricuspid valve prolapse, OR= odds ratio CI= confidence interval.

4. Discussion

PC, which has one-fifth of the prevalence of pectus excavatum, is the second most prevalent chest wall deformity (Yuksel et al., 2018). The prevalence of PC among all thoracic deformities was reported as approximately 16%. It is seen 4 times more frequently in male patients (Martinez et al., 2019). In this study, similar to the cited previous study, 80% of the pediatric PC patients consisted of males.

Although there is no clear information about its etiology, it is known that the excessive growth of the ribs is responsible for the pathogenetic mechanism of PC (Haje et al., 1999). Furthermore, the chondrogladiolar form of PC is also accompanied by the dislocation of the anterior of the sternal corpus and usually that of the lower costal cartilages. In the mixed type, both pectus excavatum and PC are seen together. The characteristic feature of the chondromanubrial type is that the protrusion of the manubrium second and third costal cartilage and the sternal corpus are relatively compressed (Fonkalsrud and Beanes, 2000).

The chondrogladiolar type is the most frequently encountered form of PC. In this study, the result was compatible with the results of previous studies, and 92.5% (n = 37) of the patients with PC were found to have the chondrogladiolar type.

In contrast to pectus excavatum, it is very rare for PC to emerge right after birth. It is generally seen in later periods of life, in preadolescence or during puberty, but it may sometimes be seen in infants and children. The incidence of PC has a tendency to increase in the period where growth occurs fast (Colombani, 2009).

Most patients do not have clinical symptoms or discomfort. However, aesthetic concerns are among main complaints. Additionally, some patients have respiratory symptoms, palpitations, and skeletal complaints (Hebra et al., 2000). In our study, most patients (60%, 24 patients) consisted of those who were asymptomatic, while among the ones who had symptoms, the most frequently reported symptoms were chest pain in 10 patients (25%) and palpitations in 6 patients (15%).

PC can be seen bilaterally or unilaterally, and the sternum is usually displaced towards the side of the deformity. PC may be an isolated deformity or may be accompanied by cardiovascular diseases (Wang et al., 2022). To examine the structure and function of the heart, pediatric cardiologists routinely utilize ECHO (Sigalet et al., 2003). Congenital heart diseases or other malformations can be encountered in addition to these deformities. Echocardiographic anomalies are frequent,

and these anomalies develop due to the abnormal formation of the chest wall and the displacement of the heart towards the left chest wall. Studies have shown that the incidence of congenital heart disease is 20% in children with PC, and an atrial septal defect is the most frequently encountered (18%) cardiac anomaly in these patients (Frick, 2000; Ercapan and Sisli, 2020). In our study, congenital heart diseases were found in three patients (bicuspid aortic valve in two patients and secundum atrial septal defect in one). The rate of cardiac pathologies that was 7.2% in the healthy control group was 57.5% in the PC group. In terms of the rates of aortic valve pathologies, in comparison to the healthy control group (2.1%), the rate in the PC group was approximately seven times as high (12.5%). There was also malposition in the standard ECHO examination of five patients (12.5%).

In agreement with the literature, in the PC group, the cardiac pathology, malposition, aortic valve pathology, and congenital heart disease rates were higher than those in the control group. Accordingly, we believe that it is highly important to examine PC patients in a multidisciplinary manner at thoracic surgery and pediatric cardiology outpatient clinics during their diagnoses and follow-ups so that congenital heart diseases can be detected, and their treatment can be started in the early period.

The prevalence of mitral valve prolapse in PC patients can be higher than that in the general population (Huang et al., 2023). According to the results of the risk analysis in our study that was conducted with the logistic regression analysis method, in comparison to the control group, the risk of MVP development increased 15.05-fold, and the risk of TVP development increased 27.87-fold in the PC group.

As our hospital is a tertiary referral hospital, there might have been a higher rate of patients with severe chest deformities included in the sample rather than mild PC deformities. Moreover, the fact that our study was conducted in a relatively short period of 12 months did not allow us to investigate the effects of chest deformities in long-term follow-ups. In addition to these issues, a limitation of this study was that the genotypic characteristics of the PC patients were not studied. Despite all these factors, this article is valuable in that it is one of the rare studies conducted for the cardiac assessment of pediatric PC patients in the literature.

5. Conclusion

Consistently with previous literature findings, our study revealed higher rates of cardiac pathologies in the pediatric PC patients compared to the healthy control group. Hence, we suggest, it is important to refer pediatric PC patients presenting to outpatient clinics to pediatric cardiology outpatient clinics for the early diagnosis of potential cardiac pathologies in this patient group.

Author Contributions

The percentage of the author(s) contributions is presented below. All authors reviewed and approved the final version of the manuscript.

	A.A.	U.U.G.	Ş.G.
C	40	30	30
D	40	30	30
S	40	30	30
DCP	40	30	30
DAI	40	30	30
L	40	30	30
W	40	30	30
CR	40	30	30
SR	40	30	30
PM	40	30	30
FA	40	30	30

C=Concept, D= design, S= supervision, DCP= data collection and/or processing, DAI= data analysis and/or interpretation, L= literature search, W= writing, CR= critical review, SR= submission and revision, PM= project management, FA= funding acquisition.

Conflict of Interest

The authors declared that there is no conflict of interest.

Ethical Approval/Informed Consent

The study is approved by Kahramanmaras Sutcu Imam University Clinical Research Local Ethics Committee (approval date: January 25, 2021, protocol code: 03). The research was conducted in accordance with the Principles of the Declaration of Helsinki.

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