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# Evaluation of Children with Chest Wall Deformities: Single Center Experience

## Göğüs Duvarı Deformitesi Olan Çocukların Değerlendirilmesi: Tek Merkez Deneyimi

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**ÖZET**

**Amaç:** Göğüs duvarı deformiteleri göğüs duvarının anormal gelişimi ile ortaya çıkabilir. Bu çalışma ile göğüs duvarı deformitesi olan çocukların demografik ve klinik özelliklerini değerlendirmek ve deformitenin fark edildiği yaş ve deformite fark edildikten sonra hastaneye başvuruya kadar geçen süre ile göğüs duvarı deformitesinin tipi arasındaki ilişkiyi araştırmak amaçlanmıştır.

**Gereçler ve Yöntem:** 2023 - 2024 yılları arasında tek bir Çocuk Göğüs Hastalıkları merkezine başvuran göğüs duvarı deformitesi olan tüm çocukların verileri retrospektif olarak hastane kayıtlarından değerlendirildi. En sık görülen iki göğüs duvarı deformitesi alt grubu demografik ve klinik bulgular açısından karşılaştırıldı. Deformitenin fark edildiği yaş ve deformite fark edildikten sonra hastaneye başvuruya kadar geçen süre arasındaki ilişki deformite tipine göre incelendi.

**Bulgular:** Çalışmaya göğüs duvarı deformitesi olan 92 çocuk dahil edildi; bunların %47,8'inde pektus karinatum ve %46,7'sinde pektus ekskavatum vardı. Ortanca yaş 6 (0,2-17) yıl idi ve göğüs duvarı deformitesinin fark edildiği ortanca yaş 4,3 (0-17) yıl idi. Deformitenin fark edildiği andan hastaneye başvuruya kadar geçen ortanca süre 6 (0-144) ay olarak bulundu. Solunum fonksiyon testi sonuçları göğüs duvarı deformitesi alt gruplarında normal sınırların içinde saptandı. Göğüs duvarı deformitesinin fark edildiği yaş ile deformite fark edildikten sonra hastaneye başvuruya kadar geçen süre arasında istatistiksel olarak anlamlı bir korelasyon bulunmadı (p=0,420).

**Sonuç:** Göğüs duvarı deformitesi olan çocuklarda akciğer kapasitesi üzerinde bir etki tespit edilmedi çünkü deformitenin fark edildiği yaş gençti ve deformite fark edildikten sonra hastaneye başvuruya kadar geçen süre göğüs duvarı deformitesinin türünden bağımsız olarak kısaydı. Bununla birlikte, göğüs duvarı deformitesi olan çocuklar zamanla akciğer sistemi etkilenebileceğinden belirli aralıklarla takip edilmelidir.

**Anahtar Kelimeler:** Çocuk, göğüs duvarı, pektus, pulmoner

**ABSTRACT**

**Aim:** Chest wall deformities may occur with abnormal development of the chest wall. It was aimed to evaluate the demographic and clinical characteristics of the children with chest wall deformity, and to investigate the relationship between the age at which the deformity was noticed and the duration until hospital admission with the type of chest wall deformity.

**Materials and Methods:** Data of all children with chest wall deformity in a single pediatric pulmonology center between 2023 - 2024 were evaluated retrospectively. The two most common chest wall deformity subgroups were compared in terms of demographic and clinical findings. The relationship between the age at which the deformity was noticed and the duration until the hospital admission was examined according to the type of deformity.

**Results:** There were 92 children with chest wall deformity; 47.8% of whom had pectus carinatum and 46.7% had pectus excavatum. The median age was 6 (0.2-17) years, the median age at which deformity was noticed was 4.3 (0-17) years and the median duration from deformity was noticed until admission was 6 (0-144) months. Pulmonary function test results were within normal limits. No statistically significant correlation was found between the age at which the deformity was noticed and the duration until admission (p=0.420).

**Conclusions:** In children with chest wall deformities, no effect on lung capacity was detected, likely because the deformity was noticed at a young age and the duration from detection to admission was short, regardless of the type of deformity. Nevertheless, children should be monitored because pulmonary system may be affected over time.

**Keywords:** Chest wall, pectus, pediatrics, pulmonary

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## INTRODUCTION

Chest wall deformities occur with abnormal development of the chest wall, such as the absence, shortness, fusion or bifurcation of one or more ribs or cartilages (1,2). These deformities can be seen as pectus excavatum (PE), pectus carinatum (PC), Poland syndrome, sternal defects and isolated costa-cartilage anomalies (1). The most common congenital chest wall deformity is PE, followed by PC and other abnormalities (1,3).

Pectus excavatum is the most common chest wall deformity in which the anterior chest wall appears concave due to deep sternum depression (3,4). PE occurs in 0.1–0.3% of live births and is common in men (3,5). It is usually noticed within first year of life and usually worsens at the beginning of puberty (3). Monitoring the PE depth during control examinations by measuring the distance from the deepest depression of the sternum to the top of the rib cage can show the progression of the deformity over time

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(6). PC is the second most common chest wall deformity, which is characterized by convex anterior protrusion of the sternum and costochondral joints (4,7). PC occurs in approximately 1/1000 live births (4). Lung development can be usually normal in children with PC (3). Poland syndrome is characterized by partial/total absence of pectoral muscles, pectus arcuatum (PA) is a mixed deformity that involves both excavatum and carinatum (4,8). Other chest wall deformities are sternal defects, rib and cartilage disorders (1).

Chest wall deformities can reduce respiratory system compliance by inhibiting growth or restricting movement of the thorax, and the most common effect on the lung is a restrictive lung defect (3,9). Children with chest wall deformities may be asymptomatic, and although patients may have various symptoms, these are not specific to chest wall deformities (9,10). Shortness of breath, exercise intolerance, chest pain, tachypnea and abnormal auscultatory findings may be observed (9). Although lateral and antero-posterior chest radiographs can also be helpful in follow-up, a computed tomography (CT) scan provides more detailed information (6). The Haller index, which can be measured on CT and is considered the gold standard in determining the severity of chest wall deformity, is based on the ratio of the horizontal distance between the two sides of the rib cage to the anterior-posterior diameter (6,11). Lung involvement in children with chest wall deformity can be determined by pulmonary function tests (PFT) (6). Surgical repair and follow-up of chest wall deformities are important to prevent further deterioration of lung function and rarely provide significant improvement in lung function (9). The aim of this study was to evaluate the demographic and clinical characteristics of children with chest wall deformities and to investigate the relationship between the age at which the deformity was noticed and the duration until hospital admission with the type of chest wall deformity.

## MATERIALS AND METHODS

The data of all children with chest wall deformity who applied to the pediatric pulmonology outpatient clinic between June 2023 and April 2024 were evaluated retrospectively. The age, gender, type of deformity, characteristics of the deformity (depth, asymmetrical/symmetrical), age when the deformity was noticed, duration after the deformity was noticed until admission, complaint at admission, presence of scoliosis and comorbid diseases, radiological findings (chest X-ray, thorax CT if available), PFT results, echocardiography results, thoracic surgery follow-up and reparative treatment were evaluated. PFT was performed on cooperative children aged 6 years and older in accordance with the American Thoracic Society and European Respiratory Society guidelines (12). PFT data included forced vital capacity (FVC), forced expiratory volume in one second ($FEV_1$), mid-expiratory flow between 25-75% of forced vital capacity ($MEF_{25-75}$) as the percentage of predicted, and $FEV_1$/FVC ratio. In PE, the depth of deformity was measured and recorded from the deepest point to the imaginary line passing through the areolas while the patient was in the supine position (13). Although CT can be considered the gold standard tool in the evaluation of thoracic morphology (14), the amount of radiation was kept in the foreground for the pediatric age group, and CT was not applied to every patient, and those who were requested to have thorax CT by the thoracic surgery department or those who had thorax CT for any other reason were recorded.

Demographic, clinical and radiological characteristics of children with PE and PC, the two most common chest wall deformity subgroups, were compared.

Ethics committee approval was obtained for this study, and all procedures in the study were carried out in accordance with ethical rules and the principles of the Declaration of Helsinki. The study was designed retrospectively, and informed consent was not obtained because the data obtained during routine follow-up of the participants were used and no additional examinations, interventions, etc. were performed for the study.

### *Statistical Analysis*

The IBM SPSS Statistics version 22.0 for Windows (IBM, Armonk, NY, USA) was used for statistical analysis. The conformity of variables to the normal distribution was examined using visual (histogram and probability graphs) and analytical methods (Kolmogorov-Smirnov Test). For descriptive statistics, categorical variables were expressed

**Table 1.** The demographic and clinical characteristics of all children with chest wall deformity

| | |
|---|---|
| **Gender** | |
| Female (n / %) | 22 / 23.9 |
| Male (n / %) | 70 / 76.1 |
| **Current age** (years) [median (min-max)] | 6 (0.2-17) |
| **Age at which deformity was noticed** (years) [median (min-max)] | 4.3 (0-17) |
| **Type of deformity (n / %)** | |
| Pectus excavatum | 43 / 46.7 |
| Pectus carinatum | 44 / 47.8 |
| Other | |
| Pectus arcuatum | 1 / 1.1 |
| Costal arch dislocation | 2 / 2.2 |
| Costal collapse | 1 / 1.1 |
| Poland syndrome | 1 / 1.1 |
| **Complaint at admission** (n / %) | (n=18) |
| Dyspnea | 8 / 44.4 |
| Cough | 6 / 33.3 |
| Chest pain | 3 / 16.7 |
| Wheezing | 1 / 5.6 |
| **Chronic diseases** (n / %) | (n=30) |
| Respiratory* | 11 / 36.7 |
| Cardiovascular** | 5 / 16.7 |
| Neurological & Neuromuscular*** | 4 / 13.3 |
| Diaphragm related problems**** | 3 / 10.0 |
| Other***** | 7 / 23.3 |

*Asthma, cystic fibrosis, primary ciliary dyskinesia
** Aortic dilatation, congenital heart disease, dextrocardia
*** Epilepsy, muscular dystrophies, cerebral palsy
**** Operated diaphragmatic hernia, diaphragm eventration
***** Autism, immunodeficiency, Down syndrome, metabolic storage disease, diabetes mellitus, operated esophageal atresia

as absolute numbers and percentages, and continuous variables were expressed as means ± standard deviations or medians (minimum-maximum). For comparisons between two independent variables, Mann-Whitney U test was used for data not normally distributed, and independent samples t test was used for normally distributed data. For correlations, Spearman's correlation test was used for data not normally distributed, and Pearson's correlation test was used for normally distributed data. Chi-square tests were used for comparisons of categorical variables between independent groups. The p value <0.05 was considered statistically significant.

## RESULTS

A total of 92 children with chest wall deformity were included in the study, 70 (76.1%) of them were male. The median age was 6 (0.2-17) years. The median age at which deformity was noticed was 4.3 (0-17) years and the median duration after deformity was noticed until admission was 6 (0-144) months. 18 (19.6%) children had complaint at admission, 8 (44.4%) of them had dyspnea. Scoliosis was found in 12 (13.0%) children. Other chronic diseases was found in 30 (32.6%) children. The demographic and clinical characteristics of all children were presented in Table 1.

All of children had chest X-ray; 49 (53.3%) were normal, 36 (39.1%) had interstitial thickness and 7 (7.6%) had other findings such as linear atelectasis, elevated diaphragm, increase in aeration, chronic findings due to comorbid diseases. 11 (12.0%) had thorax CT and 5 (45.5%) were normal, in others peribronchial thickening, bilateral reticular densities and nodules were observed.

Pulmonary function test could be attempted in 40 (43.5%) children, 8 (20.0%) of whom could not cooperate and perform the test. The mean $FEV_1$, FVC, $FEV_1$/FVC and $MEF_{25-75}$ were 108.1±18.5 %, 98.6±16.1 %, 115.5±14.9 and 124.2±29.0 %, respectively.

Echocardiography was found in 27 (29.3%) children; 17 (63.0%) of them were normal, 2 (7.4%) of them had mild pulmonary hypertension and were being followed without medication.

Although patients were directed to follow-up at the thoracic surgery clinic, there were 12 (13%) children who applied to the thoracic surgery clinic and were followed up at the time the study was conducted. Vacuum was recommended for 4 (66.7%) of the 6 children with PE who were followed up in the thoracic surgery department, and a chest belt was recommended for 1 (20.0%) of the 5 children with PC. There were no children who had a surgical plan.

Pectus excavatum was in 43 (46.7%), PC was in 44 (47.8%), PA was in 1 (1.1%) and other deformities (such as costa arch dislocation, costal collapse, Poland syndrome) was in 4 (4.3%) children. The deformity was symmetrical in 28 (62.2%) children with PC. The median depth of deformity in children with PE

**Table 2.** The demographic, clinical and radiological characteristics of children with pectus excavatum and pectus carinatum

| | Pectus excavatum | Pectus carinatum |
|---|---|---|
| **Gender** | | |
| Female (n / %) | 9 / 20.9 | 11 / 25 |
| Male (n / %) | 34 / 79.1 | 33 / 75 |
| **Current age** | | |
| (years) [median (min-max)] | 7 (0.2-17.0) | 6 (0.5-17.0) |
| **Age at which deformity was noticed** | | |
| (years) [median (min-max)] | 5 (0-17.0) | 4 (0-14.0) |
| **Duration after deformity was noticed until admission** | | |
| (months) [median (min-max)] | 6 (0-72) | 6 (0-144) |
| **Complaint at admission** (n / %) | (n=10) | (n=6) |
| Dyspnea | 3 / 30 | 4 / 66.7 |
| Cough | 5 / 50 | 1 / 16.7 |
| Chest pain | 1 / 10 | 1 / 16.7 |
| Wheezing | 1 / 10 | - |
| **Echocardiography** (n / %) | (n=15) | (n=10) |
| Normal | 8 / 53.3 | 8 / 80 |
| Abnormal / other findings | 7 / 46.7 | 2 / 20 |
| **Radiological examination** | | |
| Chest X-ray (n / %) | (n=43) | (n=44) |
| Normal | 22 / 51.2 | 23 / 52.3 |
| Interstitial thickness | 18 / 41.9 | 17 / 38.6 |
| Other findings* | 3 / 7.0 | 4 / 9.1 |
| Thorax CT (n / %) | (n=4) | (n=5) |
| Normal | 2 / 50.0 | 2 / 40 |
| Other findings# | 2 / 50.0 | 3 / 60 |

* linear atelectasis, elevated diaphragm, increase in aeration, chronic findings due to comorbid disease
# peribronchial thickening, bilateral reticular densities, nodules

**Table 3.** The comparison of pulmonary function tests of children with pectus excavatum and pectus carinatum

| | Pectus excavatum (n=14) | Pectus carinatum (n=17) | p |
|---|---|---|---|
| $FEV_1$[#] (%) | | | |
| median (min-max) | 98.5 (75-119) | 115 (82-148) | 0.024* |
| FVC[+] (%) | | | |
| median (min-max) | 89 (67-111) | 101 (71-130) | 0.009* |
| $FEV_1$/FVC | | | |
| median (min-max) | 118 (101-188) | 116 (93-118) | 0.532 |
| $MEF_{25-75}$[β] (%) | | | |
| median (min-max) | 112 (76-154) | 129 (80-191) | 0.164 |

* statistically significant
[#] forced expiratory volume in one second
[+] forced vital capacity
[β] mid-expiratory flow between 25-75% of forced vital capacity

was 10 (5-31) mm. The demographic, clinical and radiological characteristics of children with PE and PC are shown in Table 2. There were 14 children with PE and 17 children with PC who could perform PFT. Although $FEV_1$ and FVC values in the PFTs were statistically significantly lower in children with PE compared to those with PC, median PFT values in both groups were within the normal range. The comparison of PFTs of children with PE and PC is shown in Table 3.

There were no significant differences between PE and PC in terms of current age, age at which deformity was noticed and duration after deformity was noticed until admission (p=0.554, p=0.541 and p=0.795, respectively).

There was no statistically significant correlation between the age at which deformity was noticed and the duration after deformity was noticed until admission (p=0.420). There was no statistically significant correlation between the age at which deformity was noticed and PFT results (p>0.005).

## DISCUSSION

This study stated that although the age at which chest wall deformity was noticed was young and the duration between detection of deformity and hospital admission of children was short, these did not differ according to the type of deformity.

In a study conducted with 15,862 children aged 12-19, 1.6% of whom were found to have chest wall deformities, it was concluded that 30% of those with deformities were aware of their deformities and that awareness was higher in those with severe deformities (15). In this study, children were only asked whether they were aware or not of their chest wall deformities, and no questions were asked about the age at which they became aware (15). In our study, it was determined that the age at which chest wall deformity was noticed did not differ according to the type of deformity. This suggested that parents' awareness was high because the age at which the deformity was noticed was found to be young in our study.

Studies have shown male predominance in PE and PC (2-4). In our study, both PE and PC were detected more frequently in males, similar to the literature. In chest wall deformities, especially pectus excavatum, the deformity usually becomes clearer between the ages of 7-9 (1). However, although it can be easily noticed in childhood, it can sometimes be ignored (4). In our study, the age at which the deformity was noticed was relatively younger, and there was no statistical difference between the types of deformity.

As reported in some studies, children with chest wall deformities may be asymptomatic and may also have some signs and symptoms, although they are not specific to these deformities (9,10). Most of the children with chest wall deformity in our study were asymptomatic. Children with symptoms had symptoms such as dyspnea and cough, consistent with the literature. However, the symptoms of these children were not evaluated as being due to chest wall deformity but rather as being related to their additional chronic diseases.

Chest wall deformities may affect lung capacity due to the inability of the rib cage to fully expand. This effect may develop gradually and a decrease in total lung capacity may occur over time (9). In our study, it was observed that chest wall deformities did not impair PFTs in children who were able to perform the test. When PE and PC, the two major subgroups of chest wall deformity, were compared, although the $FEV_1$ and FVC values in PFT were significantly lower in those with PE compared to those with PC, the median was within the normal range in both groups. Children did not require further testing such as diffusing capacity of the lungs for carbon monoxide (DLCO); because the first step, PFT, was found to be not impaired.

In a study by Lawson ML et al. (16), it was observed that the preoperative $FEV_1$ and $FEF_{25-75}$ of patients aged 11 years and older were at the lower limit of the normal range of predicted percentages, but were not considered impaired because they were still within normal limits. In the same study, it was discussed that other studies have also reported decreases in lung function in patients with chest wall deformities, but the results are often within the normal range (16). In an adult study, PFT values were found to be lower than predicted but were still interpreted as within the normal range (17). The authors noted in another study that patients with more severe deformities were much more likely to demonstrate a restrictive lung pattern on PFT (18). Since there were no children with severe deformities in our study, PFT values may have been found to be in the normal range. Patients should remain under follow-

up as their lung capacity may be affected by the progression of their chest deformities

The limitations of the study are that it is a single center and retrospective study. However, it was considered an important issue to draw attention to since chest wall deformity can be overlooked compared to other diseases.

## CONCLUSION

In conclusion, in this study, regardless of the type of deformity, the age at which the deformity was noticed was found to be young and the time from detection to hospital admission was short. In addition, it was observed that there was no effect on lung capacity in children with chest wall deformity, but since lung capacity may be affected over time in children with chest wall deformity, it is important to follow up the child in all types of chest wall deformity, even if the child is asymptomatic.

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