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***Original Article** / Özgün Makale*

Determination of severity of deformity with rib length to costal cartilage length ratio in thorax deformities

Toraks deformitelerinde kaburga kemiği uzunluğunun kostal kıkırdak uzunluğuna oranı ile deformite şiddetinin belirlenmesi

Ali Karakılıç[1], Volkan Karaçam[2], Hasan Ersöz[3], İsmail Ağababaoğlu[4], Fatma İlknur Ulugün[2], Aydın Şanlı[2]

Institution where the research was done:
Medical Faculty of Dokuz Eylül University, İzmir, Turkey

Author Affiliations:
[1]Department of Thoracic Surgery, Menemen State Hospital, İzmir, Turkey
[2]Department of Thoracic Surgery, Medical Faculty of Dokuz Eylül University, İzmir, Turkey
[3]Department of Thoracic Surgery, İzmir Katip Çelebi University, İzmir, Turkey
[4]Department of Thoracic Surgery, Yenimahalle Training and Reserach Hospital, Ankara, Turkey

ABSTRACT

Background: This study aims to investigate whether rib length to costal cartilage length ratio is effective in determining the severity of chest wall deformity.

Methods: The study included 72 patients (64 males, 8 females; mean age 18.5±6 years; range, 6 to 40 years) who were operated because of chest wall deformity and 38 control subjects (22 males, 16 females; mean age 14.6±4.2 years; range, 6 to 25 years). Of the patients, pectus excavatum was detected in 69 and pectus carinatum in three. All participants' rib length-costal cartilage length ratio index, Haller index, correction index and computed tomography depression index were measured and compared.

Results: In patient group, there was a mild-level significant negative relationship between computed tomography depression index and rib length-costal cartilage length ratio index ($p<0.05$). Except for the computed tomography depression index, there was no significant relationship between rib length-costal cartilage length ratio index and other indexes and control group indexes in patient group. Rib length was higher than costal cartilage length in patient group compared to control group. There was no statistically significant difference between patient and control groups in terms of costal cartilage length ($p>0.05$).

Conclusion: Contrary to what would be expected, there was no significant difference between patient and control groups in terms of costal cartilage length. Therefore, studies with larger series are required to demonstrate if costal cartilage length is effective in determining the severity of chest wall deformities.

Keywords: Chest wall deformities; Haller index; costal indexes.

ÖZ

Amaç: Bu çalışmada kaburga kemiği uzunluğunun kostal kıkırdak uzunluğuna oranının göğüs duvarı deformitesi şiddetinin belirlenmesinde etkili olup olmadığı araştırıldı.

Çalışma planı: Çalışmaya göğüs duvarı deformitesi nedeniyle ameliyat edilen 72 hasta (64 erkek, 8 kadın; ort. yaş 18.5±6 yıl; dağılım, 6-40 yıl) ve 38 kontrol deneği (22 erkek, 16 kadın; ort. yaş 14.6±4.2 yıl; dağılım, 6-25 yıl) dahil edildi. Hastaların 69'unda pektus ekskavatum ve üçünde pektus karinatum saptandı. Tüm katılımcıların kaburga kemiği uzunluğu-kostal kıkırdak uzunluğu oranı indeksi, Haller indeksi, düzeltme indeksi ve bilgisayarlı tomografi depresyon indeksi ölçüldü ve karşılaştırıldı.

Bulgular: Hasta grubunda bilgisayarlı tomografi depresyon indeksi ve kaburga kemiği uzunluğu-kostal kıkırdak uzunluğu oranı indeksi arasında hafif düzeyde anlamlı negatif bir ilişki vardı ($p<0.05$). Bilgisayarlı tomografi depresyon indeksi dışında, hasta grubunda kaburga kemiği uzunluğu-kostal kıkırdak uzunluğu oranı indeksi ve diğer indeksler ve kontrol grubu indeksleri arasında anlamlı bir ilişki yoktu. Kontrol grubuna kıyasla hasta grubunda kaburga kemiği uzunluğu kostal kıkırdak uzunluğundan daha yüksekti. Hasta ve kontrol grupları arasında kostal kıkırdak uzunluğu açısından istatistiksel olarak anlamlı farklılık yoktu ($p>0.05$).

Sonuç: Beklenenin aksine, hasta ve kontrol grupları arasında kostal kıkırdak uzunluğu açısından anlamlı farklılık yoktu. Dolayısıyla, göğüs duvarı deformitelerinin şiddetinin belirlenmesinde kostal kıkırdak uzunluğunun etkili olup olmadığını ortaya koymak için daha geniş serili çalışmalar gereklidir.

Anahtar sözcükler: Göğüs duvarı deformiteleri; Haller indeksi; kostal indeksler.

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Correspondence: Ali Karakılıç, MD. Menemen Devlet Hastanesi, Göğüs Cerrahisi Kliniği, 35660 Menemen, İzmir, Turkey
Tel: +90 544 - 728 36 82 e-mail: dralikarakilic@hotmail.com

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Congenital chest wall deformities are the most common congenital anomalies that may be seen in various forms of deformities.[1] Pectus excavatum (PE) is the most common chest wall deformity, with a prevalence of one in 300-400 to 1000 live births. On the other hand, the second most common chest wall deformity is pectus carinatum (PC). Compared to excavatum type deformity, PC is seen 10 times less frequently.[2,3] It usually occurs at birth or in the first year of life, resulting from posterior collapse of the sternum together with abnormal growth cartilage, or protrusion to the front. In males, it is encountered three to four times more.[4] Cardiac and respiratory problems are more indicative of surgical treatment due to social behavior and aesthetic concerns that restrict self-confidence.[5] At present, it has been reported that treatment of PE with Nuss procedure has a positive effect on physical and psychological state and cosmetic concerns of young adults apart from increasing the quality of life despite the pain and surgical injuries in the early period.[6] Surgical correction is performed using different techniques to maintain the normal position of the sternum and the normal appearance of the chest wall.[7]

Indexing of chest wall deformities may be necessary to ensure the standardization of the treatment plan and the timing of operation. For this purpose, many indexes have been determined. Haller index (HI) is the most commonly used index today to determine the severity of chest wall deformities.[8] Haller index is derived from dividing the transverse diameter of the thorax on computed tomography (CT) to the distance between vertebra anterior surface and the posterior surface at the deepest point of the sternum. The HI is 2.6±0.4 for healthy people. Once above 3.25, there is indication of surgery. When the index is lower than 2.5, it is considered as mild, when between 2.5 and 3.2, it is considered as moderate, and if it is greater than 3.2, it is considered as severe deformity. The index does not correlate with age, operative time, postoperative bar infection or duration of hospitalization.[9]

In this study, we measured the rib length-costal cartilage length ratio index of participants on the basis of the recommendations of Onen, who has many studies on chest wall deformity surgery and is an accepted author on this subject in our country with work on hundreds of cases.[6] In our study, since our observations on the cases we operated revealed excessive cartilage length, we assumed that proportioning rib length with that of costal cartilage length might shed light on determining the severity of deformity in PE and PC cases. Therefore, in this study, we aimed to investigate whether rib length to costal cartilage length ratio is effective in determining the severity of chest wall deformity.

PATIENTS AND METHODS

This retrospective study included 72 patients (64 males, 8 females; mean age 18.5±6 years; range, 6 to 40 years) operated due to chest wall deformities at Dokuz Eylul University Medical Faculty Hospital Thoracic Surgery Clinic between June 2009 and October 2011 and 38 controls (22 males, 16 females; mean age 14.6±4.2 years; range, 6 to 25 years) who applied to our clinic due to other clinical symptoms except for chest wall deformities during the same period and who were performed thorax CT. The study protocol was approved by Ethics Committee of Non-Interventional Research in Dokuz Eylul University dated 17.09.2015 and numbered 2015/22-05. A written informed consent was obtained from each participant. The study was conducted in accordance with the principles of the Declaration of Helsinki.

In our study, patients operated due to chest wall deformity was named as PEC group (PE and PC), while controls without chest wall deformities were defined as N (normal) group.

Posteroanterior, lateral lung graphs and three-dimensional (3D) thorax CT examinations were performed in all cases except for routine laboratory examinations during the operation preparation phase. Preoperatively, cardiac evaluation was performed through electrocardiogram, while respiratory function was evaluated with respiratory function tests. Although the indications for surgery were usually psychosocial problems due to cosmetic impairment independent of the degree of deformity, HI, which was developed to make a more objective decision, was calculated.

Nuss procedure was applied to 70 patients with PE in PEC group and to three patients with Abramson procedure and PC history. Patients were retrospectively screened. Name, surname, age, gender, right and left rib length-costal cartilage length (sixth rib) at the deepest point of the sternum; the thorax transverse diameter, the anteroposterior (AP) diameter of the sternum-vertebra corpus, the anterior-posterior diameter of the left hemithorax and the amount of depth of depression were recorded. However, a patient with Nuss procedure history was excluded from the study as the patient had a Ravitch procedure previously, making it impossible to measure costal cartilage length precisely. Thus, the PEC group included 72 patients (64 males, 8 females; mean age 18.5±6 years; range, 6 to 40 years).

Based on the data given, the following indexes were measured: *(i)* Haller Index (HI), *(ii)* CT depression index (DI), *(iii)* correction index (CI), and *(iv)* rib length-costal cartilage length ratio index. These indexes were compared between PEC and N groups in terms of the results of the measurements and the means. Costal cartilage lengths and rib lengths were also compared between both groups.

Haller index

Although the indications for surgery are usually psychosocial problems due to cosmetic impairment independent of the degree of deformity, HI was calculated to make a more objective decision. The result is derived by dividing the lateral diameter of thorax to anterior-posterior diameter of sternum at the deepest point.[8] Haller Index is widely used today (Figure 1).

A= Transverse diameter at the level where deformity is deepest,

B= Anterior-posterior diameter at the same level.

Haller index= A/B HI is 2.6±0.4 for normal people.

The threshold value for surgical indication is accepted as 3.25. Although there are authors recommending surgical correction in patients with HI higher than 3.25, it is not a standardized practice to determine operation indications by calculating an index.

Computed tomography depression index

In the area where the sternum is deepest, the ratio of the largest area of the AP diameter of the left hemithorax to the distance between the sternum posterior and vertebral anterior surface of the thorax gives the DI (Figure 2). A significant positive correlation was detected between the severity of chest wall deformities, the amount of sternal depression, and cardiac pressure findings in evaluations performed by CT.[10]

Depression index is expressed as DI=L/AP. This value is classified as mild when below 2.4, moderate when between 2.4-2.9 and severe when over 2.9.

Correction index

In order to evaluate the correction, an imaginary horizontal line is drawn in sternum posterior and from this line, the distance to the posterior of sternum is calculated from where sternum is deepest (D). Following this, the distance from sternum posterior to vertebral corpus anterior (AP) is calculated (Figure 2). CI is expressed as CI=D/(D+AP) X 100. This ratio gives the correction ratio.

A percentage above 28% may be regarded as an operation indication, since this value correlates with the value of 3.25 of the HI.[11] The correlation of HI and CI is well in patients with standard chest wall deformities, yet it is not well in nonstandard chest wall deformities.

Rib length-costal cartilage length ratio index

This was calculated based on the measurements of rib length and costal cartilage length in the sixth rib in PEC group, where deformity was severe, and in N group.[12] Three dimension reconstruction of the chest CTs of patients and curved axial sections of the multiplanar images (3D volume rendered and curved multi-planar reformatted [MPR] images) were taken and the rib length from the costovertebral joint to the costochondral joint were measured separately on the

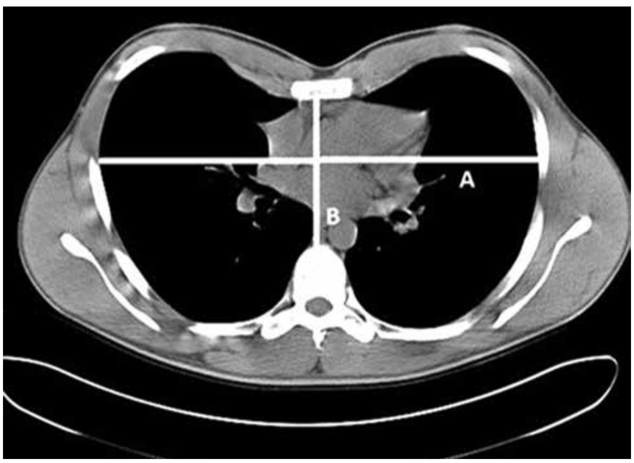

Figure 1. Haller index.

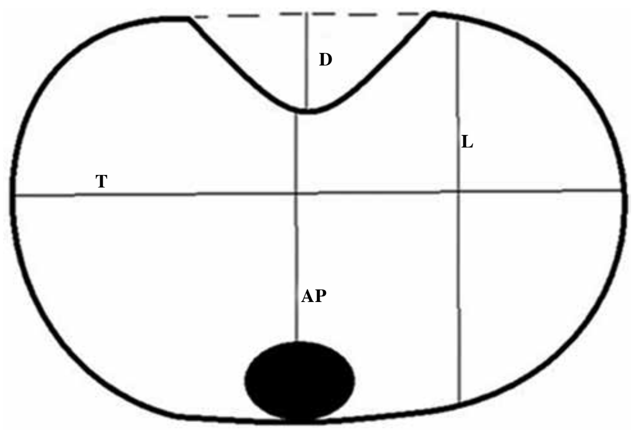

Figure 2. Computed tomography depression index and correction index.

right and left sides and recorded as rib length. Likewise, cartilage lengths from the junction of the costochondral joint to the junction of the sterno-costal joint were measured in the right and left sides and the data were collected and recorded as the costal cartilage length.

We used 3D volume-rendered CT images and curved MPR reformatting techniques on picture archiving and communication system for measuring. The costal index was determined as the proportion of rib length to costal cartilage length. This index is expressed as Rib Length (Blue)/Costal Cartilage Length (Red) (Figure 3).

Statistical analysis

The obtained data were analyzed using the Statistical Package for the Social Sciences (SPSS) version 15.0 (SPSS Inc., Chicago, IL, USA). The results were presented as mean±standard deviation and lowest and highest values. Correlation of the related parameters with each other was interpreted by Pearson correlation coefficient (r) analysis. A *p* value of <0.05 was considered statistically significant.[13]

RESULTS

In PEC group, Nuss procedure was performed due to PE in 69 patients and Abramson operation was performed due to PC in three patients. There was no respiratory (chronic obstructive pulmonary disease, interstitial lung disease, emphysema, chronic bronchitis or tuberculosis etc.) and/or cardiac disease that could affect the chest wall measurements of patients in both groups.

In PEC group, mean HI was 3.4±1, with the lowest value of 1.47 and highest value of 6.50. Mean value of CT DI was 2.0±0.5, with the lowest value of 1.00 and highest value of 3.62. CI mean was 0.3±0.1, with the lowest value of 0.04 and highest value of 0.58. Rib length-costal cartilage length ratio index was 3.1±0.5, with the lowest value of 2.20 and highest value of 4.77.

In N group, mean HI was 2.2±0.3, with the lowest value of 1.81 and highest value of 3.36. Mean CT DI was 1.5±0.1, with the lowest value of 1.26 and highest value of 1.79. Mean CI was 0.1±0.0, with the lowest value of 0.01 and highest value of 0.13. Rib length-costal cartilage length ratio index was 2.8±0.4, with the lowest value of 2.06 and highest value of 3.99.

On the basis of the threshold values of patients in PEC and N groups, HI of 35 patients was higher than 3.25 (48%). The CI of 32 patients was higher than 0.28 (44%). The CT DI of 15 patients was higher than 2.4 (20%).

A comparison of the mean rib length, costal cartilage length, HI, CI, CT DI and rib length-costal cartilage length ratio index of patients in PEC and N groups revealed that rib length, total costal length, rib length-costal cartilage length ratio index, HI, CI and CT DI were increased in PEC group compared to N group (p<0.05). There was no statistically significant difference in terms of mean costal cartilage lengths between PEC and N groups (p>0.05).

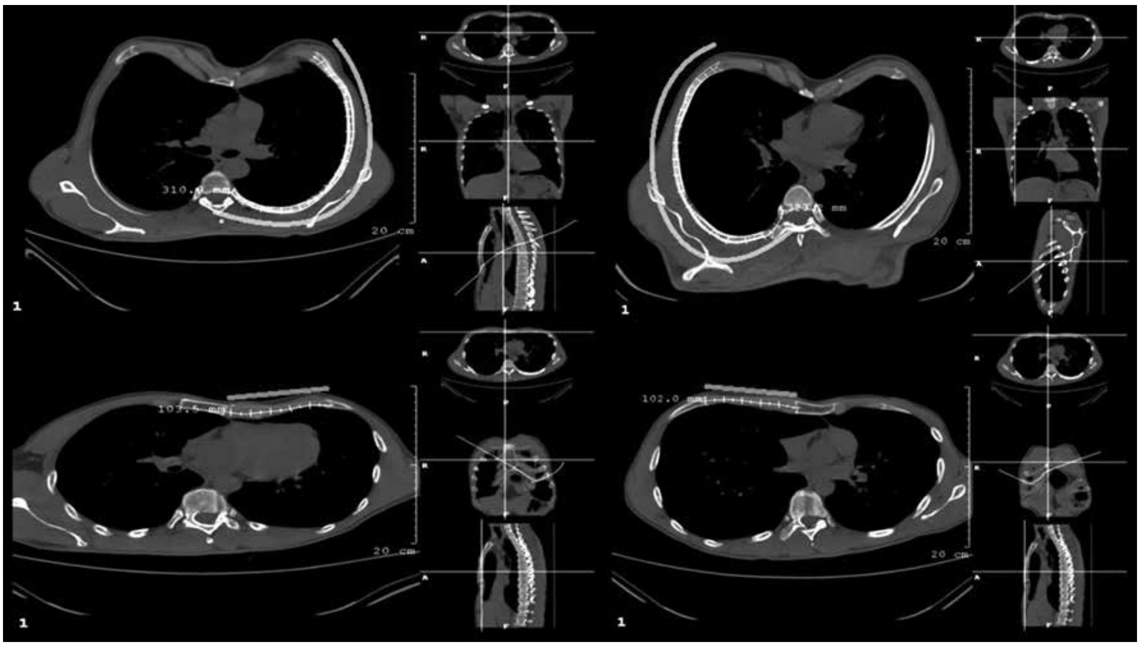

Figure 3. Rib length-costal cartilage length ratio index.

Table 1. Comparison of indexes in pectus excavatum and pectus carinatum group (p<0.05)

Group PEC (r-p)	R/C	HI	DI	CI
R/C	1	-0.183	-0.283*	-0.205
		0.124	0.016	0.084
HI		1	0.958*	0.912*
			0.000	0.000
DI			1	0.936*
				0.000
CI				1

R/C: Rib Lenght-Costal Cartilage Lenght Ratio Index; HI: Haller Index; DI: CT Depression Index; CI: Correction Index.

Table 2. Comparison of indexes in normal group (p<0.05)

Group N (r-p)	R/C	HI	DI	CI
R/C	1	-0.005	-0.093	0.024
		0.974	0.580	0.886
HI		1	0.782*	0.448*
			0.000	0.005
DI			1	0.680*
				0.000
CI				1

R/C: Rib Lenght-Costal Cartilage Lenght Ratio Index; HI: Haller Index; DI: CT Depression Index; CI: Correction Index.

Relationship Between Measured Indexes

When the rib length-costal cartilage length ratio index and the other indexes including HI, CT DI, and CI of patients in PEC group were scanned comparatively, there was no correlation with HI and CI, while there was a mild negative correlation with CT DI (p<0.05). There was a significant positive correlation between HI and CT DI and CI (p<0.05). There was also a significant positive correlation between CT DI and CI (p<0.05) (Table 1).

When the rib length-costal cartilage length ratio index, HI, CT DI, and CI were scanned comparatively in N group, no significant correlation was detected among HI, CI and CT DI. There was a significant positive correlation between HI and DI at a high level and a mild positive correlation with CI and a moderate positive correlation between CT DI and CI (p<0.05). There was also a moderate positive correlation between CT DI and CI (p<0.05) (Table 2).

A comparison of the indexes showed that they were more correlated in PEC group than N group.

DISCUSSION

A number of methods have been described in the treatment of PE and PC. However, there is no concrete criterion about when and which treatment method should be preferred. Various indexes have been defined in determining the type and severity of chest wall deformities. Although HI has been defined as the most accepted one, this index does not correlate with age, operative duration, postoperative bar infection or duration of hospitalization.[9] Haller index shows 48% overlap among normal patients and those with pectus deformity.[14] There are few studies on costal indexes in the literature.

Excessive growth of cartilage is widely accepted as the cause of chest wall deformities.[15,16] It is generally thought that the sternum is displaced posteriorly due to overgrowth of the lower costal cartilages, and probably the diaphragm also plays a role in this displacement.[17,18] There are also publications indicating that the cause of deformity does not depend on costal cartilage length.[19,20]

Cartilage length is not considered in HI. Therefore, a difference in terms of this condition arises in this study according to HI. The use of cartilage length in our study and rib length-costal cartilage length ratio index for this purpose suggests that the determination of chest wall deformity severity in the etiology of hypertrophic cartilage growth is more significant, while on the other hand, raises suspicion about the abnormal costal cartilage length actually found in the etiology when the available statistical data and the literature are analyzed. Similarly, there was no significant difference in the normal control group and the operated group in terms of the costal cartilage in our study.

In another study, patients with PE and control patient group's length of costal cartilage and length of the costal cartilages between the rotated and non-rotated side in asymmetric PE patients were compared. Lengths of costal cartilages were similar in PE patients and control patients. These were also similar between the rotated and non-rotated sides of the sternum in patients with asymmetric PE. Consequently, it was found that costal cartilage overgrowth cannot be a main causative factor for PE.[21]

In one study including patients with asymmetric PC, the fourth, fifth, and sixth cartilage lengths were compared with the side where more protrusion was present and with contralateral costal cartilage length measurement, which demonstrated no difference in between. These findings indicate that costal cartilage length does not explain the asymmetric protrusion of the chest wall, and that the main cause of PC is not costal cartilage length.[22]

In another study, length of rib and length of the costal cartilages were compared in symmetrical PE patients under 10 years of age and age- and sex-matched control patients group without chest wall deformity. Rib length was significantly longer on the right sixth rib and left fourth, fifth and sixth ribs compared to the control group. It was found that abnormal rib length is more responsible for the deformity than costal cartilage length. There was no significant difference between the two groups in terms of rib length-costal cartilage length ratio.[12]

Number of publications on costal indexes is limited. For this reason, no clear information can be given on their normal values. The threshold for surgery is not mentioned since there is no correlation with the most widely accepted HI.

The following findings were detected in a study about asymmetric pectus and sternal rotation: in patients who had asymmetric PE, the ribs were longer than control patients while costal cartilage was not longer than the controls. As a result, it was concluded that cartilage overgrowth is not the main factor responsible for asymmetric PE, and it is derived from abnormal rib growth.[23]

Similarly, in our study, there was no significant difference in the normal control group and the operated group in terms of the costal cartilage. The rib length and the total costal length were longer in the operated group than in the control group. The rib length-costal cartilage length ratio index was higher in the operated group because of the longer rib length. This suggests that rib length is more valuable than costal cartilage in the etiology of chest wall deformities.

In both of the above mentioned studies and in our study, evaluations with thoracic CT were performed and it was determined that while there was an expected increase in costal cartilage length in the etiology, there was no difference between the control and study groups. In our study, significant results which were negatively and mildly correlated with CT DI were obtained. Although the results were significant, they only correlated with the CT DI.

A significant correlation is determined when the severity of chest wall deformity is compared with the CT DI, HI and CI. The DI provides objective measurements independent of thoracic measurements to determine the severity of the deformity.[24]

Another study demonstrated that HI and CI correlate better in patients with standard chest wall deformities, but are highly discordant in nonstandard chest wall deformities.[11] In our study, there was a high positive significant correlation among HI, CI and CT DI compatible with the literature.

Although the restrictive spirometric data of pectus excavated patients correlated with other indexes, HI was the most valuable index.[25] The rib length-costal cartilage length ratio index was not correlated with HI or CI. Studies with larger series examining patients with different types of deformities with different courses of treatment are needed to be able to assume that this index is stronger and more significant.

Our study has some limitations. The major limitations were small sample size of study and minority of pectus carinatum cases. Nonetheless, the findings are noteworthy. More reliable scientific results can be obtained by increasing the number of cases particularly pectus carinatum cases.

In conclusion, contrary to what was expected, there was no significant difference between the normal control and operated study groups in terms of costal cartilage length. Also, the rib length and rib length-costal cartilage length ratio index were found to be statistically significantly higher in the operated study group. Although our costal index was significantly higher in the study group, it was insufficient in accordance with the literature to determine the degree of deformity with rib length-costal cartilage length ratio index. This suggests that in chest wall deformities and particularly in symmetric cases, Haller index is strong in determining the severity of deformity. Today, the use of Haller index is still effective and maintains its importance in determining the severity of chest wall deformity. The existing indexes used to determine the severity of deformity complement each other at this stage. Studies with larger series are needed to determine the severity of chest wall deformities and whether the etiology indeed arises from costal cartilage length.

Declaration of conflicting interests

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