



The etiology of pectus carinatum involves overgrowth of costal cartilage and undergrowth of ribs



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ABSTRACT

Purpose: We compared the length of costal cartilage and rib between patients with symmetric pectus carinatum and controls without anterior chest wall protrusion, using a 3-dimensional (3D) computed tomography (CT) to evaluate whether the overgrowth of costal cartilage exists in patients with pectus carinatum.

Subjects and methods: Twenty-six patients with symmetric pectus carinatum and matched twenty-six controls without chest wall protrusion were enrolled. We measured the full lengths of the 4th–6th ribs and costal cartilages using 3-D volume rendering CT images and the curved multiplanar reformatted (MPR) techniques. The lengths of ribs and costal cartilages, the summation of rib and costal cartilage lengths, and the costal index [length of cartilage/length of rib * 100 (%)] were compared between the patients group and the control group at 4th–6th levels.

Results: The lengths of costal cartilage in patient group were significantly longer than those of control group at 4th, 5th and 6th rib level. The lengths of ribs in patient group were significantly shorter than those of control group at 4th, 5th and 6th rib level. The summations of rib and costal cartilage lengths were not longer in patients group than in control group. The costal indices were significantly larger in patients group than in control groups at 4th, 5th and 6th rib level.

Conclusion: In patients with symmetric pectus carinatum, the lengths of costal cartilage were longer but the lengths of rib were shorter than those of controls. These findings may supports that the overgrowth of costal cartilage was not the only factor responsible for pectus carinatum.

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Pectus carinatum is the second most common congenital anterior chest wall deformity representing protrusion of the sternum and ribs [1,2]. The etiology of pectus carinatum is uncertain and still under debate. Among the several hypotheses for the etiology of pectus carinatum, defective elongated costal cartilage is the generally accepted theory [3,4]. However, evidence for this hypothesis is not robust [2]. Our previous study revealed that the lengths of ribs and costal cartilage in patients with asymmetric pectus carinatum were not different between the more protruded side and the opposite side at the 4th, 5th and 6th rib levels [5]. These findings supported that the overgrowth of costal cartilage could not be the main cause of pectus carinatum. However there was no study which compared the lengths of ribs or cartilages between the patients with pectus carinatum and the controls without chest wall deformity. So, in this study, we aimed to compare the length of cartilage and ribs between patients with symmetric pectus carinatum and controls without anterior chest wall protrusion using three-dimensional computed tomography (3D-CT)

imaging to identify whether overgrowth of costal cartilage exists in patients with pectus carinatum.

1. Materials and methods

1.1. Patients

This study was performed retrospectively and informed consent from the patients was waived with approval from the institutional review board. Between January 2012 and August 2013, patients with pectus carinatum who underwent chest CT scans were reviewed. Symmetric pectus carinatum was defined as pectus carinatum with a 5° or smaller angle of sternal rotation on CT images (Fig. 1A). Patients with a sternal rotation angle of greater than 5° were excluded. To reduce bias, patients with previous chest surgery or thoracic cage deformity were excluded. Finally 26 patients were enrolled. We also enrolled 26 matched controls without chest wall deformity, considering age, sex, height and weight, who underwent chest CT in our institution. The clinical indications for CT scans of the control group were pneumothorax in 12, pneumonia in 8, trauma in 3, tumor in 1, pneumomediastinum in 1 and abnormality on chest radiography in 1. Clinical data were reviewed retrospectively from the medical records of patients and controls.

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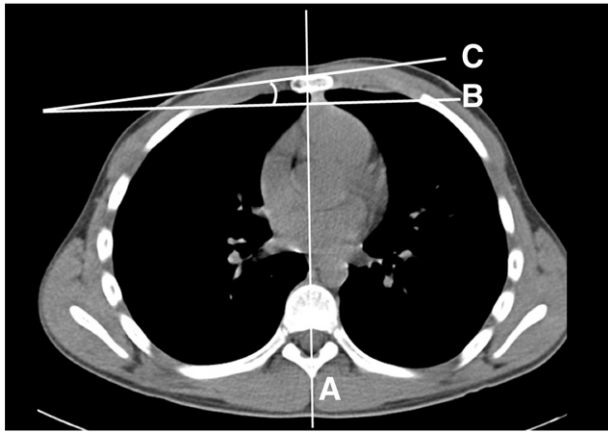


Fig. 1. Measurement of the sternal rotation angle. (a) The sternal rotation angle is measured as the angle of sternal slope (Line C) against the baseline of the thorax (Line B) at a slice with a largest sternal slope. The baseline of thorax was defined as the perpendicular line to the vertical line traversing the T spine's center (Line A).

1.2. CT scanning

CT scans were performed using one of three scanners: a 16-slice multidetector CT (MDCT) scanner (Somatom Sensation 16; Siemens Medical Solutions, Erlangen, Germany), a 64-slice MDCT scanner (Somatom Sensation 64; Siemens Medical Solutions), or a 128-slice MDCT scanner (Somatom Definition AS+; Siemens Medical Solutions). Subjects were scanned in the supine position from the lung apices to the level of the adrenal glands during breath hold at the end of inspiration. After acquiring a scout image to determine the field of view (FOV), conventional CT scanning was performed without contrast enhancement using a helical technique, with a 1-mm or 5-mm reconstruction interval in the mediastinal window setting. The exposure parameters for the CT scans were 80–120 kVp, 50–130 mA, 1-mm or 5-mm slice thickness, and 1-mm or 5-mm reconstruction increment. Image reconstruction for conventional CT scan was performed on the scanner's workstation. All CT images were sent to the picture archiving and communication system (PACS) (Centricity 1.0; GE Medical Systems, Mt Prospect, IL).

1.3. CT image analysis

On the PACS, sternal rotation angles were measured using an electronic protractor. The sternal angle of rotation was defined as the sternal slope against the baseline. The sternal angle of rotation was measured at the point of the greatest angle as described in the previous studies [6–8]. The baseline was defined as the perpendicular line to the vertical line traversing the center of T-spine (Fig. 1). The Haller index was defined as maximal internal transverse diameter of

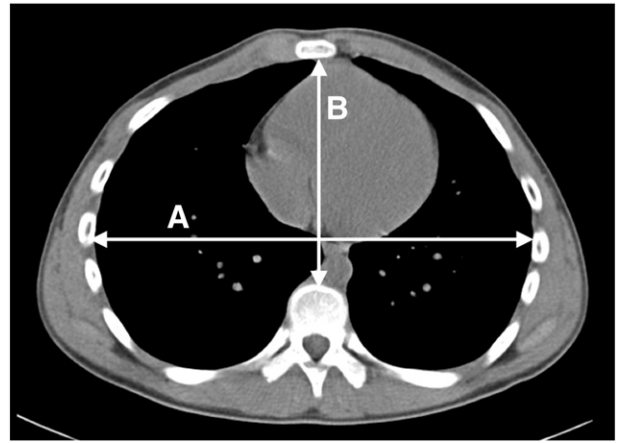


Fig. 2. Measurement of the Haller index. The Haller index is defined as maximal internal transverse diameter of the chest (A) divided by the anteroposterior diameter between the sternum and the vertebral bodies (B) on axial CT images.

the chest divided by the anteroposterior diameter between the sternum and the vertebral bodies at the same level on axial CT images [9,10] (Fig. 2). All CT images of patients with symmetric pectus carinatum and controls were transferred to a commercially available reconstruction program (Aquarius iNtuition Ver.4.4.6 TeraRecon, Foster City, CA) and then 3D volume rendering (VR) and multiplanar reformatted (MPR) images were built to measure the full length of the ribs and costal cartilage by an automatic segmentation and reconstruction technique. Unnecessary images of other regions, like bilateral scapulas, were manually deleted. The full length of the ribs and costal cartilage was traced semiautomatically and measured using the curved MPR technique with manual correction at the fourth, fifth, and sixth rib levels (Figs. 3 and 4). The total combined rib and costal cartilage length was the summation of the rib and costal cartilage length. The costal index (%) was defined as the ratio of the cartilage length to the rib length ((cartilage length/rib length) * 100) [11]. The length of each rib and costal cartilage, the total combined length of the ribs and costal cartilage, and the costal index were compared between the patient group and control group at the fourth, fifth, and sixth rib levels. The differences between two groups were calculated by subtracting the values of the control group from those obtained from the patient group.

1.4. Statistical analysis

Continuous variables were expressed as mean \pm standard deviation (SD) and categorical variables were expressed as frequencies or percentages. Paired *T*-test or Wilcoxon signed rank test was used to compare the age, height, weight, body mass index (BMI), the Haller index between the patient and control groups. Paired *T*-test was used

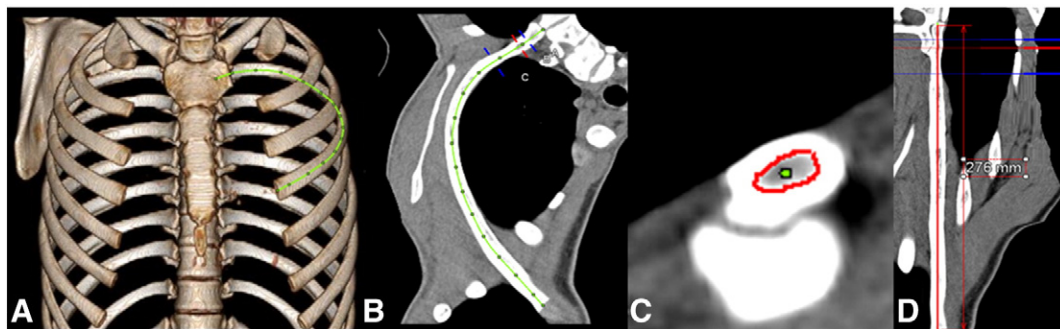


Fig. 3. Measurement of the full length of the rib. (A) Three-dimensional volume rendering (VR) image is reconstructed automatically with a commercially available reconstruction program (Aquarius iNtuition Ver.4.4.6 TeraRecon, Foster City, CA). Unnecessary regions are manually deleted. (B) The full length of the left 4th rib is semiautomatically traced using the curved MPR technique with manual correction. (C) The tracing line passes through the center of rib. (D) The full length of the rib is measured by electronic caliper.

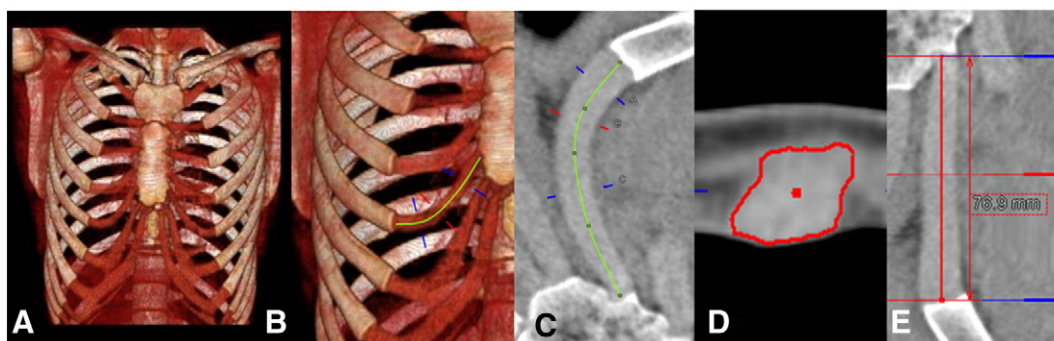


Fig. 4. Measurement of the full length of the cartilage. (A) The cartilage is less contrasted than the bone in CT. However the methodology for cartilage measuring was exactly same with that of the rib. (B) Three-dimensional volume rendering (VR) image is reconstructed automatically with a commercially available reconstruction program. (C) The full length of the right 4th cartilage is semiautomatically traced using the curved MPR technique with manual correction. (D) The tracing line passes through the center of cartilage. (E) The full length of the cartilage is measured by electronic caliper.

to compare the length of ribs, length of costal cartilage, total combined length of ribs and costal cartilage, and the costal index between right side and left side of each group. Paired *T*-test was used to compare the same parameters between the patient group and the control group at the same level of same sides. Intraclass correlation coefficients (ICCs) were used to evaluate interobserver reliability of the Haller index, rib lengths and cartilage lengths between two radiologists (C.H.P and I.J). A *p*-value less than 0.05 was considered statistically significant. All statistical analysis was performed with commercially-available software (SPSS 20; Statistical Package for the Social Sciences, Chicago, IL).

2. Results

Twenty-six patients with symmetric pectus carinatum and twenty-six matched controls were enrolled in this study. The demographic data of patients and controls are summarized in Table 1. All patients and controls were male. The age, height, weight and BMI were not significantly different in two groups ($p > 0.05$). The Haller index of the patient group was significantly smaller than that of the controls (2.15 ± 0.17 vs. 2.59 ± 0.28 , $p < 0.001$).

All the ribs and cartilages of level 4th, 5th and 6th were evaluated on both right and left sides. A total of 312 costal cartilages and 312 ribs were measured.

The laterality effects of the rib and cartilage lengths were summarized in Tables 2 (patient group) and 3 (control group). The lengths of ribs or cartilages at 4th, 5th and 6th levels were not different between the right side and left side, except the 6th costal cartilage. The lengths of the left 6th costal cartilage were longer than those of the right 6th cartilage in patient group as well as in control group (99.3 ± 9.1 mm vs. 102.0 ± 9.1 mm, $p = 0.001$ in patient group. 90.8 ± 6.8 mm vs. 93.6 ± 8.6 mm, $p = 0.002$ in control group).

The comparison of the cartilage length, rib length, total combined cartilage and rib length, and the mean costal index between patient

group and control group at the fourth through sixth rib levels are summarized in Table 4.

The lengths of costal cartilage in the patient group were significantly longer than those of control group at the 4th, 5th and 6th rib levels. (57.3 ± 3.8 mm vs. 51.7 ± 3.9 mm at 4th level, 72.7 ± 5.2 mm vs. 66.0 ± 3.8 mm at 5th level, and 99.3 ± 9.1 mm vs. 90.8 ± 6.8 mm (Right side)/ 102.0 ± 9.1 mm vs. 93.6 ± 8.6 mm (Left side) at 6th level) (Fig. 5).

The lengths of ribs in patient group were shorter than those of controls at the 4th, 5th and 6th rib levels. (262.9 ± 18.6 mm vs. 275.8 ± 20.4 mm at 4th level, 276.6 ± 19.9 mm vs. 289.7 ± 21.0 mm at 5th level, and 283.4 ± 20.8 mm vs. 295.0 ± 21.8 mm (Right side)/ 282.5 ± 20.1 mm vs. 294.0 ± 21.1 (Left side) at 6th level) (Fig. 6).

The total combined lengths of rib and costal cartilage were not longer in patients group than in controls at the 4th, 5th and 6th rib levels (320.3 ± 19.7 mm vs. 327.5 ± 20.4 mm at 4th level, 349.3 ± 22.9 mm vs. 355.8 ± 20.4 mm at 5th level and 382.8 ± 23.2 mm vs. 385.8 ± 22.5 mm (Right side)/ 384.7 ± 22.0 mm vs. 387.7 ± 24.9 mm (Left side) at 6th level) (Fig. 7).

The costal indices were significantly larger in patients than in controls at the 4th, 5th and 6th rib levels ($21.9 \pm 1.8\%$ vs. $18.9 \pm 2.1\%$ at 4th level, $26.3 \pm 1.8\%$ vs. $22.9 \pm 2.0\%$ at 5th level, and $35.2 \pm 4.1\%$ vs. $30.9 \pm 3.2\%$ (Right side)/ $36.3 \pm 4.1\%$ vs. $31.9 \pm 3.4\%$ (Left side) at 6th level) (Fig. 8).

Table 2

Comparison of the cartilage length, rib length, total combined cartilage and rib length, and the costal index between right side and left side in the patient group.

Level	Right side	Left side	Differences	P-value
The cartilage length (mean \pm SD, mm)				
4th	57.0 ± 3.9	57.6 ± 4.0	-0.5 ± 2.1	0.212
5th	72.1 ± 5.6	73.3 ± 5.5	-1.3 ± 3.5	0.076
6th	99.3 ± 9.1	102.0 ± 9.1	-2.6 ± 3.7	0.001
The rib length (mean \pm SD, mm)				
4th	263.9 ± 17.8	261.9 ± 20.0	2.0 ± 7.6	0.198
5th	277.3 ± 19.9	275.9 ± 20.3	1.4 ± 5.0	0.174
6th	283.4 ± 20.8	282.5 ± 20.1	0.9 ± 5.0	0.379
The combined cartilage and rib length (mean \pm SD, mm)				
4th	321.0 ± 18.6	319.5 ± 21.3	1.4 ± 7.0	0.312
5th	349.4 ± 23.2	349.3 ± 23.1	0.1 ± 6.4	0.932
6th	382.8 ± 23.2	384.7 ± 22.0	-1.8 ± 6.7	0.196
The costal index (mean \pm SD,%)				
4th	21.7 ± 1.9	22.1 ± 1.9	-0.4 ± 1.3	0.135
5th	26.0 ± 1.8	26.7 ± 2.1	-0.6 ± 1.4	0.028
6th	35.2 ± 4.1	36.3 ± 4.1	-1.0 ± 1.4	0.001

●Difference = (value from right side) – (value from left side).

Table 1

Demographic data of patients with symmetric pectus carinatum and controls without chest wall deformity.

Characteristics	Patient group (mean \pm SD)	Control group (mean \pm SD)	P-value
Male–female ratio	26: 0	26: 0	
Age (y)	15.0 ± 2.7	15.3 ± 3.2	0.069
Height (cm)	169.1 ± 9.4	169.2 ± 9.5	0.699
Weight (kg)	52.5 ± 9.1	53.5 ± 8.7	0.088
Body mass index	18.2 ± 2.0	18.6 ± 1.9	0.085
Haller Index	2.15 ± 0.17	2.59 ± 0.28	< 0.001

Table 3

Comparison of the cartilage length, rib length, total combined cartilage and rib length, and the costal index between right side and left side in the control group.

Level	Right side	Left side	Differences	P-value
The cartilage length (mean \pm SD, mm)				
4th	51.7 \pm 3.7	51.8 \pm 4.4	−0.2 \pm 2.8	0.788
5th	66.1 \pm 3.9	65.9 \pm 4.4	0.2 \pm 3.1	0.700
6th	90.8 \pm 6.8	93.6 \pm 8.6	−2.8 \pm 4.2	0.002
The rib length (mean \pm SD, mm)				
4th	276.0 \pm 21.1	275.6 \pm 19.8	0.4 \pm 4.3	0.653
5th	289.9 \pm 20.6	289.6 \pm 21.8	0.4 \pm 4.3	0.806
6th	295.0 \pm 21.8	294.0 \pm 21.1	0.9 \pm 4.2	0.271
The combined cartilage and rib length (mean \pm SD, mm)				
4th	327.7 \pm 20.9	327.4 \pm 20.2	0.2 \pm 5.5	0.828
5th	356.0 \pm 20.2	355.5 \pm 23.4	0.5 \pm 7.0	0.695
6th	385.8 \pm 22.5	387.7 \pm 24.9	−1.9 \pm 6.5	0.194
The costal index (mean \pm SD, %)				
4th	18.8 \pm 2.2	18.9 \pm 2.1	−0.1 \pm 1.1	0.754
5th	22.9 \pm 2.1	22.9 \pm 2.1	0.1 \pm 1.2	0.770
6th	30.9 \pm 3.2	31.9 \pm 3.4	−1.0 \pm 1.3	0.001

●Difference = (value from right side) − (value from left side).

The interobserver reliabilities for the Haller index, the length of cartilage, and the length of rib were summarized in the Table 5. The intraclass correlation coefficients between two readers were high (0.986 in the Haller index, 0.998 in cartilage length, 0.998 in rib length retrospectively).

3. Discussion

The aim of this study was to evaluate whether the overgrowth of costal cartilage exists in patients with pectus carinatum by comparing

Table 4

Comparison of the cartilage length, rib length, total combined cartilage and rib length, and the costal index between patient group and control group at the fourth through sixth rib levels.

Level	Pectus carinatum	Controls	Difference	P-value
The cartilage length (mean \pm SD, mm)				
4th Mean	57.3 \pm 3.8	51.7 \pm 3.9	5.6 \pm 4.8	<0.001
5th Mean	72.7 \pm 5.2	66.0 \pm 3.8	6.7 \pm 5.1	<0.001
6th Right	99.3 \pm 9.1	90.8 \pm 6.8	8.5 \pm 10.1	<0.001
Left	102.0 \pm 9.1	93.6 \pm 8.6	8.3 \pm 12.5	0.002
The rib length (mean \pm SD, mm)				
4th Mean	262.9 \pm 18.6	275.8 \pm 20.4	−12.9 \pm 15.9	<0.001
5th Mean	276.6 \pm 19.9	289.7 \pm 21.0	−13.1 \pm 15.3	<0.001
6th Right	283.4 \pm 20.8	295.0 \pm 21.8	−11.5 \pm 13.5	<0.001
Left	282.5 \pm 20.1	294.0 \pm 21.1	−11.5 \pm 14.4	<0.001
The combined cartilage and rib length (mean \pm SD, mm)				
4th Mean	320.3 \pm 19.7	327.5 \pm 20.4	−7.3 \pm 15.4	0.024
5th Mean	349.3 \pm 22.9	355.8 \pm 22.0	−6.4 \pm 17.3	0.069
6th Right	382.8 \pm 23.2	385.8 \pm 22.5	−3.0 \pm 15.5	0.332
Left	384.7 \pm 22.0	387.7 \pm 24.9	−3.2 \pm 20.0	0.429
The costal index (mean \pm SD, %)				
4th Mean	21.9 \pm 1.8	18.9 \pm 2.1	3.0 \pm 2.3	<0.001
5th Mean	26.3 \pm 1.8	22.9 \pm 2.0	3.5 \pm 2.0	<0.001
6th Right	35.2 \pm 4.1	30.9 \pm 3.2	4.3 \pm 4.1	<0.001
Left	36.3 \pm 4.1	31.9 \pm 3.4	4.3 \pm 4.6	<0.001

●Difference = (value from patient's group) − (value from control group).

the lengths of ribs and costal cartilage in patients with symmetric pectus carinatum and controls without chest wall deformity. Our results demonstrate that, in patients, costal cartilages are longer and ribs are shorter than in controls.

The costal cartilage overgrowth is a generally accepted theory of pectus carinatum as well as pectus excavatum [12,13]. Nakaoka et al.

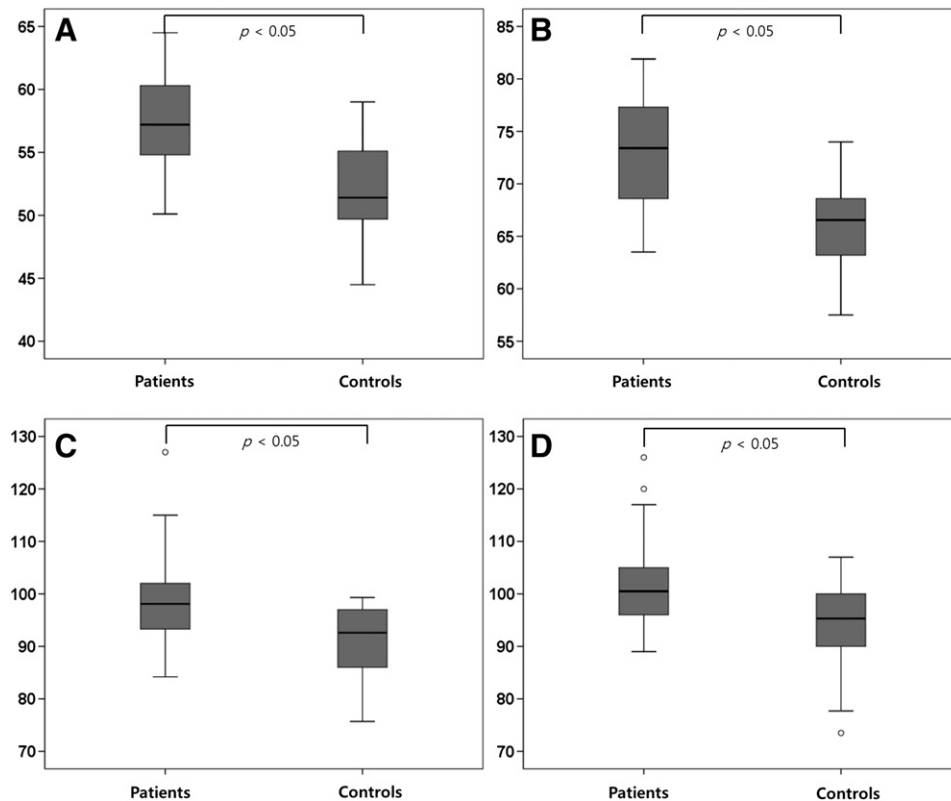


Fig. 5. Differences of cartilage lengths between patient group and control group. (A) 4th Cartilage (B) 5th Cartilage. (C) Right 6th Cartilage (D) Left 6th Cartilage. The lengths of costal cartilage in the patient group were significantly longer than those of control group at the 4th, 5th and 6th rib levels in both sides. ((A) 57.3 \pm 3.8 mm vs. 51.7 \pm 3.9 mm at 4th level, (B) 72.7 \pm 5.2 mm vs. 66.0 \pm 3.8 mm at 5th level, and (C) 99.3 \pm 9.1 mm vs. 90.8 \pm 6.8 mm (Right side)/(D) 102.0 \pm 9.1 mm vs. 93.6 \pm 8.6 mm (Left side) at 6th level.) Box = 1st–3rd quartiles, Bold line = median, Whiskers = minimum and maximum values, o = outlier.

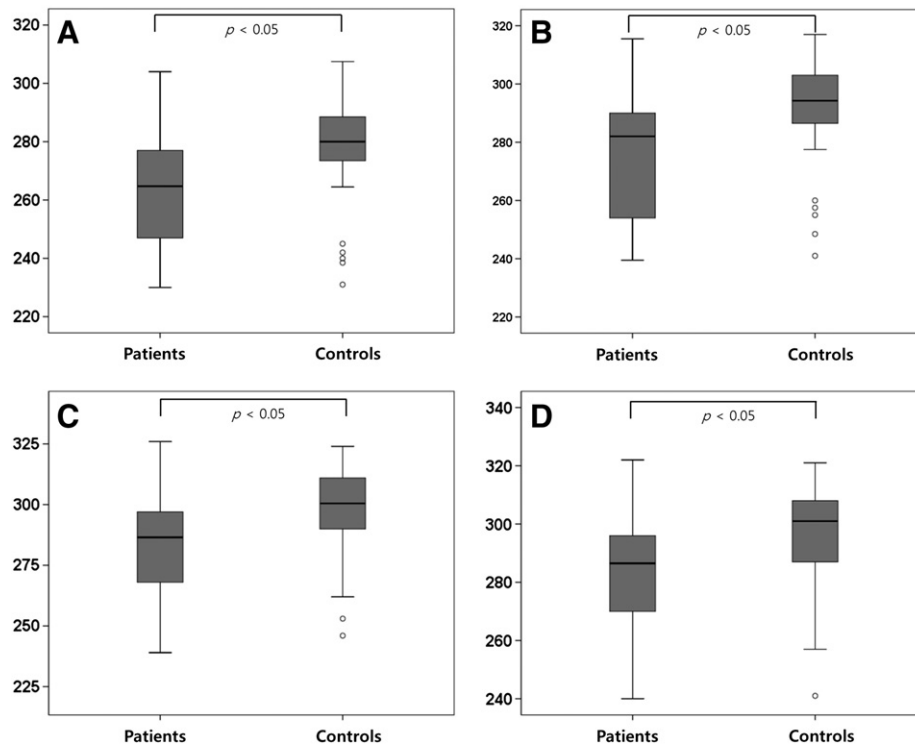


Fig. 6. Differences of the rib lengths rib between patient group and control group. (A) 4th Rib (B) 5th Rib. (C) Right 6th Rib (D) Left 6th Rib. The lengths of ribs in patient group were shorter than those of controls at the 4th, 5th and 6th rib levels on both sides. ((A) 262.9 ± 18.6 mm vs. 275.8 ± 20.4 mm at 4th level, (B) 276.6 ± 19.9 mm vs. 289.7 ± 21.0 mm at 5th level, and (C) 283.4 ± 20.8 mm vs. 295.0 ± 21.8 mm (Right side)/(D) 282.5 ± 20.1 mm vs. 294.0 ± 21.1 (Left side) at 6th level.) Box = 1st–3rd quartiles, Bold line = median, Whiskers = minimum and maximum values, o = outlier.

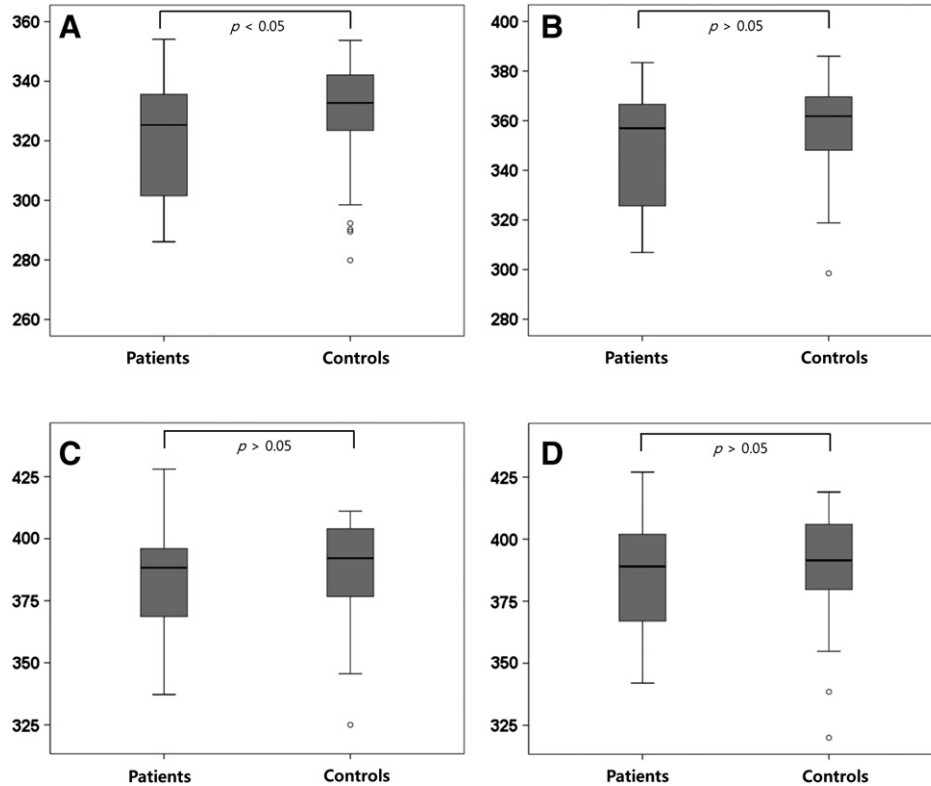


Fig. 7. Differences of the summation of the lengths of rib and costal cartilage between patient group and control group. (A) 4th Rib + Cartilage (B) 5th Rib + Cartilage. (C) Right 6th Rib + Cartilage (D) Left 6th Rib + Cartilage. The total combined length of ribs and costal cartilage was not longer in patients group than in controls at the 4th, 5th and 6th rib levels on both sides ((A) 320.3 ± 19.7 mm vs. 327.5 ± 20.4 mm at 4th level, 349.3 ± 22.9 mm vs. (B) 355.8 ± 20.4 mm at 5th level and (C) 382.8 ± 23.2 mm vs. 385.8 ± 22.5 mm (Right side)/(D) 384.7 ± 22.0 mm vs. 387.7 ± 24.9 mm (Left side) at 6th level.) Box = 1st–3rd quartiles, Bold line = median, Whiskers = minimum and maximum values, o = outlier.

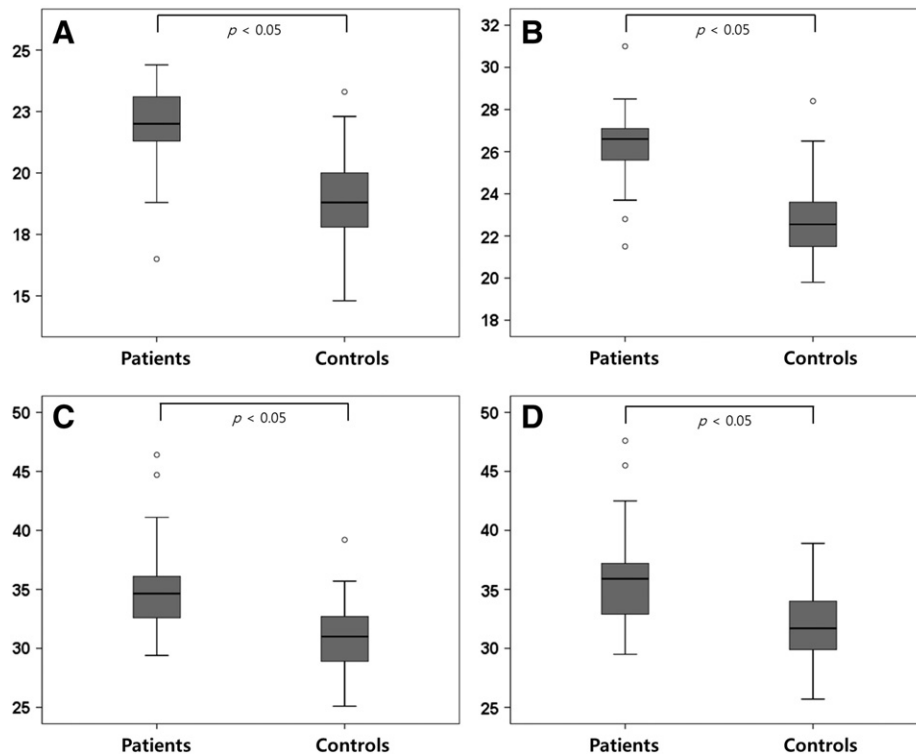


Fig. 8. Differences of the costal index in between patient group and control group. (A) 4th costal index (B) 5th costal index. (C) Right 6th costal index (D) Left 6th costal index. The costal indices were significantly larger in patients than in controls at the 4th, 5th and 6th rib levels on both sides ((A) $21.9 \pm 1.8\%$ vs. $18.9 \pm 2.1\%$ at 4th level, (B) $26.3 \pm 1.8\%$ vs. $22.9 \pm 2.0\%$ at 5th level, and (C) $35.2 \pm 4.1\%$ vs. $30.9 \pm 3.2\%$ (Right side)/(D) $36.3 \pm 4.1\%$ vs. $31.9 \pm 3.4\%$ (Left side) at 6th level.) Box = 1st–3rd quartiles, Bold line = median, Whiskers = minimum and maximum values, o = outlier.

[6,14] evaluated this theory on the pectus excavatum and reported that the length of the cartilage or ribs on the severer depressed side was not longer than those on the opposite side in patients with asymmetric pectus excavatum, and that the costal indices of patients with symmetric excavatum were similar to those of healthy controls. These results motivated our hypothesis that the overgrowth of costal cartilage might not be the main cause for pectus carinatum, either [5]. In the present study, we investigated this hypothesis by comparing the length of ribs and costal cartilage between patients with symmetric pectus carinatum and controls without anterior chest wall deformities.

For a precise comparison, all patients were matched to the controls one by one considering age, sex, height and weight. The Haller index of the patient group was significantly smaller than that of the control group. However the absolute difference between two groups was not so large (2.15 vs. 2.59). The Haller index is significantly related to age and gender. The Haller index of the control group in this study was not smaller comparing with the reported normal range of the children [9]. All patients with pectus carinatum were diagnosed on the basis of physical examination and patient's symptom by one experienced thoracic surgeon (S. Lee) who had more than 10-years' experience in

chest wall deformity. Anterior protrusion of pectus carinatum is usually more severe on physical examination than CT findings. Even though, the Haller index is a useful tool to evaluate chest wall configuration, the morphology of thoracic cage and the position of vertebra also can affect the Haller index as well as anterior chest wall protrusion or depression.

This study reveals that patients with symmetric pectus carinatum have longer costal cartilages and shorter ribs than controls at the fourth, fifth and sixth rib level. Therefore, the costal indices were significantly larger in patients than in controls. These findings may support that pectus carinatum is related to the undergrowth of rib as well as overgrowth of costal cartilage. The combined total length of ribs and costal cartilage was not longer in patient group than in control group, so the anterior protrusion of the chest wall in pectus carinatum could go beyond a simple issue of the cartilage length or rib lengths. The abnormalities of costal cartilage, rib, diaphragm, sternum were suggested as the etiology of pectus carinatum [3,15,16]. Within the cartilage abnormalities, developmental cartilage dysfunction or cartilage structure could be important factors of the pectus carinatum as well as cartilage length [17]. Various diseases including scoliosis, Marfan syndrome, Morquio syndrome, Poland anomaly, Moebius anomaly, osteogenesis imperfecta, Noonan syndrome, and congenital heart disease were related to pectus carinatum [2,12,18,19].

Our results are contrary to previous studies which measured the lengths of cartilages or ribs in patients with pectus excavatum [6,14]. Haje et al. [15] reported that the injuries of sternal growth plate could develop the pectus carinatum as well as pectus excavatum. However the pathophysiology of pectus carinatum might be different with that of pectus excavatum. Pectus carinatum usually starts or spurts with progressive growth around the puberty and rarely presents at birth but pectus excavatum tends to start from birth [2,3].

The measuring of full length of rib or cartilage is not easy owing to its 3D configuration. For the exact measurement, we used curved MPR

Table 5
The interobserver reliability in measurement of the Haller index, rib lengths and cartilage lengths.

	Reader 1 (mean \pm SD)	Reader 2 (mean \pm SD)	Difference	Intraclass correlation coefficient
Haller Index	2.37 \pm 0.32	2.36 \pm 0.31	0.01 \pm 0.07	0.986
Cartilage length	73.9 \pm 20.6	73.6 \pm 20.1	0.4 \pm 1.6	0.998
Rib length	277.5 \pm 20.0	278.9 \pm 20.1	-1.3 \pm 1.8	0.998

●Interobserver reliability was performed between two readers (C.H.P. and I.J.).

●Difference = (value from reader 1) – (value from reader 2).

technique of which the tracing lines passing through the center of each rib or cartilage. To ensure the passage of the tracing lines, the cross-sectional images, 3D-VR images were also used. This method allowed exact measurement of 3D structures like ribs and cartilage.

Our study has several limitations. First, the study sample size was too small to generalize results. Second, this study was limited to measuring and comparing the 4th–6th ribs and costal cartilages. However enrolled patients were all diagnosed with lower-type pectus carinatum, in which the 4th–6th rib levels are the main contributors to chest wall protrusion [6]. Third, matched controls without chest wall deformity were enrolled among the patients who underwent chest CT scan. This control group might not be fully healthy and there could be a selection bias. Finally the pubertal stage of patients and controls was not considered even though pubertal stage may affect the growth of rib or cartilage.

In conclusion, the length of costal cartilage is longer and the length of rib is shorter in patients with pectus carinatum than those in controls. These findings may support that the overgrowth of costal cartilage was not the only factor responsible for pectus carinatum.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <http://dx.doi.org/10.1016/j.jpedsurg.2014.02.044>.

References

- [1] Fokin AA, Steuerwald NM, Ahrens WA, et al. Anatomical, histologic, and genetic characteristics of congenital chest wall deformities. *Semin Thorac Cardiovasc Surg* 2009;21:44–57.
- [2] Goretsky MJ, Kelly Jr RE, Croitoru D, et al. Chest wall anomalies: pectus excavatum and pectus carinatum. *Adolesc Med Clin* 2004;15:455–71.
- [3] Coelho Mde S, Guimaraes Pde S. Pectus carinatum. *J Bras Pneumol* 2007;33:463–74.
- [4] Brochhausen C, Muller FK, Turial S, et al. Pectus carinatum—first ultrastructural findings of a potential metabolic lesion. *Eur J Cardiothorac Surg* 2012;41:705–6.
- [5] Park CH, Kim TH, Haam SJ, et al. Does overgrowth of costal cartilage cause pectus carinatum? A three-dimensional computed tomography evaluation of rib length and costal cartilage length in patients with asymmetric pectus carinatum. *Interact Cardiovasc Thorac Surg* 2013;17:757–63.
- [6] Nakaoka T, Uemura S, Yano T, et al. Does overgrowth of costal cartilage cause pectus excavatum? A study on the lengths of ribs and costal cartilages in asymmetric patients. *J Pediatr Surg* 2009;44:1333–6.
- [7] Egan JC, DuBois JJ, Morphy M, et al. Compressive orthotics in the treatment of asymmetric pectus carinatum: a preliminary report with an objective radiographic marker. *J Pediatr Surg* 2000;35:1183–6.
- [8] Stephenson JT, Du Bois J. Compressive orthotic bracing in the treatment of pectus carinatum: the use of radiographic markers to predict success. *J Pediatr Surg* 2008;43:1776–80.
- [9] Daunt SW, Cohen JH, Miller SF. Age-related normal ranges for the Haller index in children. *Pediatr Radiol* 2004;34:326–30.
- [10] Haller Jr JA, Kramer SS, Lietman SA. Use of CT scans in selection of patients for pectus excavatum surgery: a preliminary report. *J Pediatr Surg* 1987;22:904–6.
- [11] Sandoz B, Badina A, Laporte S, et al. Quantitative geometric analysis of rib, costal cartilage and sternum from childhood to teenagehood. *Med Biol Eng Comput* 2013;51:971–9.
- [12] Robicsek F, Watts LT. Pectus carinatum. *Thorac Surg Clin* 2010;20:563–74.
- [13] Lester CW. The etiology and pathogenesis of funnel chest, pigeon breast, and related deformities of the anterior chest wall. *J Thorac Surg* 1957;34:1–10.
- [14] Nakaoka T, Uemura S, Yoshida T, et al. Overgrowth of costal cartilage is not the etiology of pectus excavatum. *J Pediatr Surg* 2010;45:2015–8.
- [15] Haje SA, Bowen JR, Harcke HT, et al. Disorders in the sternal growth and “pectus” deformities: an experimental model and clinical correlation. *Acta Orthop Bras* 1998;6:67–75.
- [16] Harcke HT, Grissom LE, Lee MS, et al. Common congenital skeletal anomalies of the thorax. *J Thorac Imaging* 1986;1:1–6.
- [17] Benhamed L, Hysi I, Wurtz AJ. eComment. Is overgrowth of costal cartilages the unique cause of pectus deformities? *Interact Cardiovasc Thorac Surg* 2013;17:763.
- [18] Shamberger RC, Welch KJ. Surgical correction of pectus carinatum. *J Pediatr Surg* 1987;22:48–53.
- [19] Kotzot D, Schwabegger AH. Etiology of chest wall deformities—a genetic review for the treating physician. *J Pediatr Surg* 2009;44:2004–11.