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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

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Abstract

OBJECTIVES: To evaluate whether the overgrowth of costal cartilage may cause pectus carinatum using three-dimensional (3D) computed tomography (CT).

METHODS: Twenty-two patients with asymmetric pectus carinatum were included. The fourth, fifth and sixth ribs and costal cartilages were semi-automatically traced, and their full lengths were measured on three-dimensional CT images using curved multi-planar reformatted (MPR) techniques. The rib length and costal cartilage length, the total combined length of the rib and costal cartilage and the ratio of the cartilage and rib lengths (C/R ratio) in each patient were compared between the protruding side and the opposite side at the levels of the fourth, fifth and sixth ribs.

RESULTS: The length of the costal cartilage was not different between the more protruded side and the contralateral side (55.8 ± 9.8 mm vs 55.9 ± 9.3 mm at the fourth, 70 ± 10.8 mm vs 71.6 ± 10.8 mm at the fifth and 97.8 ± 13.2 mm vs 99.8 ± 15.5 mm at the sixth; $P > 0.05$). There were also no significant differences between the lengths of ribs. (265.8 ± 34.9 mm vs 266.3 ± 32.9 mm at the fourth, 279.7 ± 32.7 mm vs 280.6 ± 32.4 mm at the fifth and 283.8 ± 33.9 mm vs 283.9 ± 32.3 mm at the sixth; $P > 0.05$). There was no statistically significant difference in either the total length of rib and costal cartilage or the C/R ratio according to side of the chest ($P > 0.05$).

CONCLUSIONS: In patients with asymmetric pectus carinatum, the lengths of the fourth, fifth and sixth costal cartilage on the more protruded side were not different from those on the contralateral side. These findings suggest that overgrowth of costal cartilage cannot explain the asymmetric protrusion of anterior chest wall and may not be the main cause of pectus carinatum.

Keywords: Pectus carinatum • Costal cartilage • Rib • Three-dimensional computed tomography

INTRODUCTION

Pectus carinatum is an outward protrusion deformity of the sternum and/or ribs. It is the second most common congenital deformity of the anterior chest wall and comprises 5–20% of all anterior chest wall deformities [1, 2]. The exact cause of pectus carinatum is uncertain. Among the several proposed causes of pectus carinatum, exaggerated growth of the costal cartilage is the most widely accepted explanation, similar to pectus excavatum [3]. However, evidence for this hypothesis is lacking, and the aetiology of the potential cartilage overgrowth is also uncertain [2].

Nakaoka *et al*. [4] have reported that, in patients with asymmetric pectus excavatum, the lengths of the ribs and costal cartilages in the more depressed side were not longer than those in the contralateral side. They had hypothesized that, if overgrowth of the costal cartilage was the cause of pectus excavatum, the costal cartilage lengths on the more depressed side would be longer than those on the opposite side. Their findings suggest, however, that overgrowth of the costal cartilage might not be the aetiology of that asymmetric chest deformity. That a similar hypothesis has been articulated but not tested regarding patients with asymmetric pectus carinatum motivated our examination of the length of costal cartilage and ribs in pectus carinatum. Therefore, we evaluate these measurements in patients with asymmetric pectus carinatum using three-dimensional (3-D) computed tomography (CT) images to determine whether overgrowth of the costal cartilage could explain the asymmetry of pectus carinatum.

MATERIALS AND METHODS

Patients

Patients with asymmetric pectus carinatum who underwent CT scans between January 2006 and February 2013 were included in

Figure 1: Measurement of the sternal rotation angle. The sternal rotation angle was measured as the maximum angle of the sternal slope against the baseline of the thorax. Patients with a >10° angle of sternal rotation were regarded as having asymmetric pectus carinatum. In this patient, the sternal rotation angle is 27° and the right side is the more protruded than the left.

the study. The study received approval from the institutional review board at our hospital. Their imaging and clinical data were retrospectively reviewed from medical records, and the requirement for informed consent was waived. Asymmetric pectus carinatum was defined as pectus carinatum with 10° or greater angle of sternal rotation on the CT images (Fig. 1). Patients with a sternal rotation angle <10° were excluded. To reduce bias, those patients with previous chest surgery or other thoracic cage deformities were excluded.

CT scan protocol

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, Erlangen, Germany), or a 128-slice MDCT scanner (Somatom Definition AS+; Siemens Medical Solutions, Erlangen, Germany). Scanning was performed in the supine position from the lung apices to the level of the adrenal glands during inspiration. After acquiring the scout image to determine the field of view, conventional CT scanning was performed without contrast enhancement using a helical technique, with a 1 or 5 mm reconstruction interval in the mediastinal window setting. The exposure parameters for the CT scans were as follows: 80–100 kVp, 50∼130 mA, 1- or 5-mm slice thickness and 1- or 5-mm reconstruction increment. Image reconstruction for conventional CT scan was performed on the scanner's workstation. All CT images were retrieved on a picture archiving and communication system (PACS) (Centricity 1.0; GE Medical Systems, Mt Prospect, IL, USA) and then analysed using the mediastinal window settings (level, 50 HU; width, 400 HU).

CT image analysis

Two radiologists (T.H.K. and C.H.P.) with over 20 and 8 respective years of experience in chest radiology interpretation assessed the CT images. On the PACS, the angles of sternal rotation were measured using the electronic protractor as the maximum angle of sternal slope against the baseline of the thorax (Fig. 1). All CT images of those patients with asymmetric pectus carinatum were transferred to a commercially available reconstruction programme (Aquarius iNtuition Ver. 4.4.6 TeraRecon, Foster City, CA, USA), and 3-D volume rendering (VR) and curved multi-planar reformatted (MPR) images were constructed to measure the full lengths of the ribs and coastal cartilage. Unnecessary regions, including the bilateral scapulae, were carefully manually deleted. The full length of each rib and costal cartilage was semi-automatically traced and measured using a curved MPR technique with manual correction at the fourth, fifth and sixth rib levels on both sides (Figs. 2 and 3). The summation of the rib length and costal cartilage length and the ratio of the cartilage length to the rib length (C/R ratio), defined as (cartilage length/rib length × 100, %), were also calculated levels at the fourth, fifth and sixth rib levels. The length of rib, the length of costal cartilage, the total combined length of the rib and costal cartilage and the C/R ratio were compared across sides. The lateral differences were calculated by subtracting the values obtained on the non-protruding side from those obtained for the more prominent side.

Statistical analysis

Continuous variables were expressed as mean ± standard deviation (SD), and categorical variables were expressed as frequency or percentage. The paired *t*-test was used to compare the rib length, the cartilage length, the total combined length of the rib and costal cartilage and the C/R ratio on the more protruded side with that of the less protruded side at the fourth, fifth and sixth rib levels. A *P*-value of <0.05 was considered to be statistically significant. All statistical analysis was performed with commercially available software (SPSS 20; Statistical Package for the Social Science, Chicago, IL, USA).

RESULTS

The records of 24 patients with asymmetric pectus carinatum were reviewed; 2 patients were excluded due to asymmetric sternal defects or a bifid rib, leaving 22 patients enrolled in this study (Table 1). Nineteen of the patients were male and 3 were female. The mean age of the enrolled patients was 13.7 ± 2.9 years (range: 5–18 years old). The mean angle of sternal rotation was 13.3 ± 3.8° (range: 10–27°). The right side was the more protruded side in 11 patients, while the other 11 patients had greater prominence of the left side.

The mean cartilage lengths at the fourth rib level were 55.8 ± 9.8 mm vs 55.9 ± 9.3 mm, on the more prominent and opposite sides, respectively. Also at this level, the mean rib lengths and the total combined cartilage and rib lengths on the protruded side and the opposite sides were 265.8 ± 34.9 mm vs 266.3 ± 32.9 mm, 321.1 ± 40.3 mm vs 322.5 ± 37.7 mm, respectively. The mean C/R ratios in the more protruded side and in the opposite side at the fourth rib level were 21.2 ± 3.5% vs 21.1 ± 3.4%, retrospectively. At the fifth rib level, the mean cartilage lengths were 70 ± 10.8 mm vs 71.6 ± 10.8 mm, the mean rib lengths were 279.7 ± 32.7 mm vs 280.6 ± 32.4 mm, the mean total combined cartilage and rib lengths were 349.8 ± 39.7 mm vs 352.2 ± 39.3 mm, and the mean C/R ratio was 25.1 ± 3.3% vs 25.6 ± 3.3%, on the more

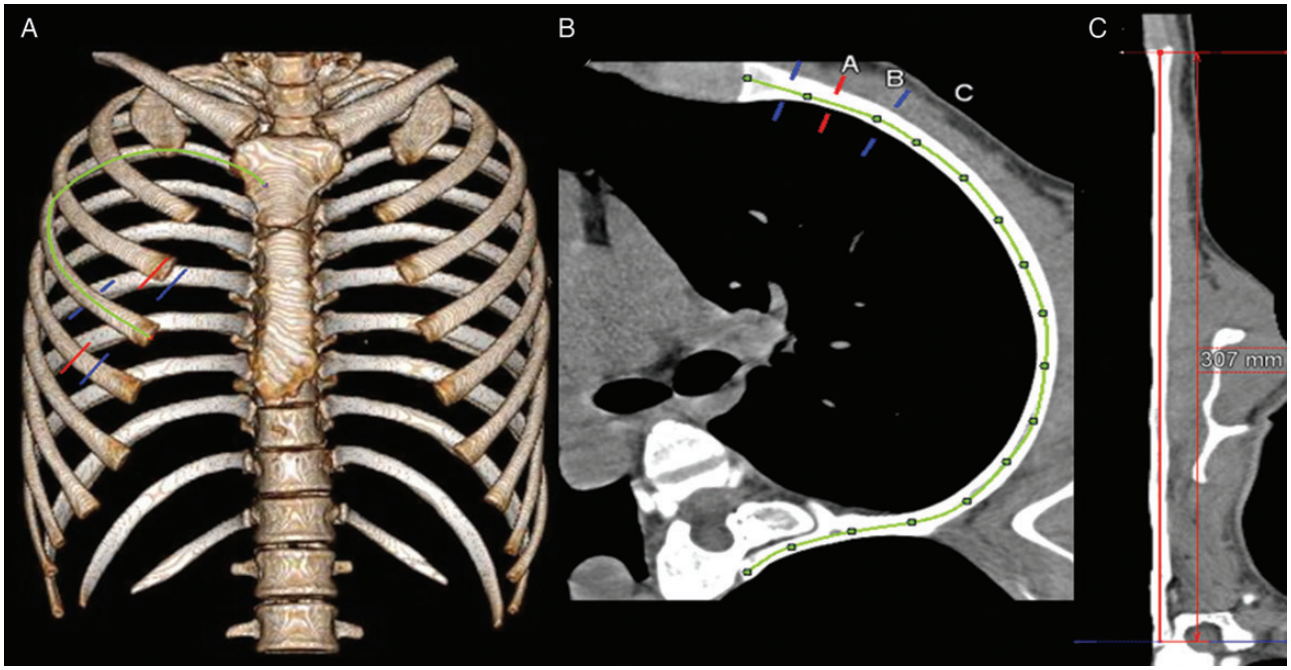

Figure 2: Measurement of the full length of the rib. All CT images of patients with asymmetric carinatum were transferred to a commercially available reconstruction programme (Aquarius iNtuition Ver.4.4.6 TeraRecon, Foster City, CA, USA). 3-D VR image was reconstructed, and unnecessary regions including bilateral scapulars were carefully deleted manually (**A**). The full length of the right fourth rib was semi-automatically traced using a curved multi-planar reformatted technique with manual correction (**B**) and then measured by electronic caliper (**C**).

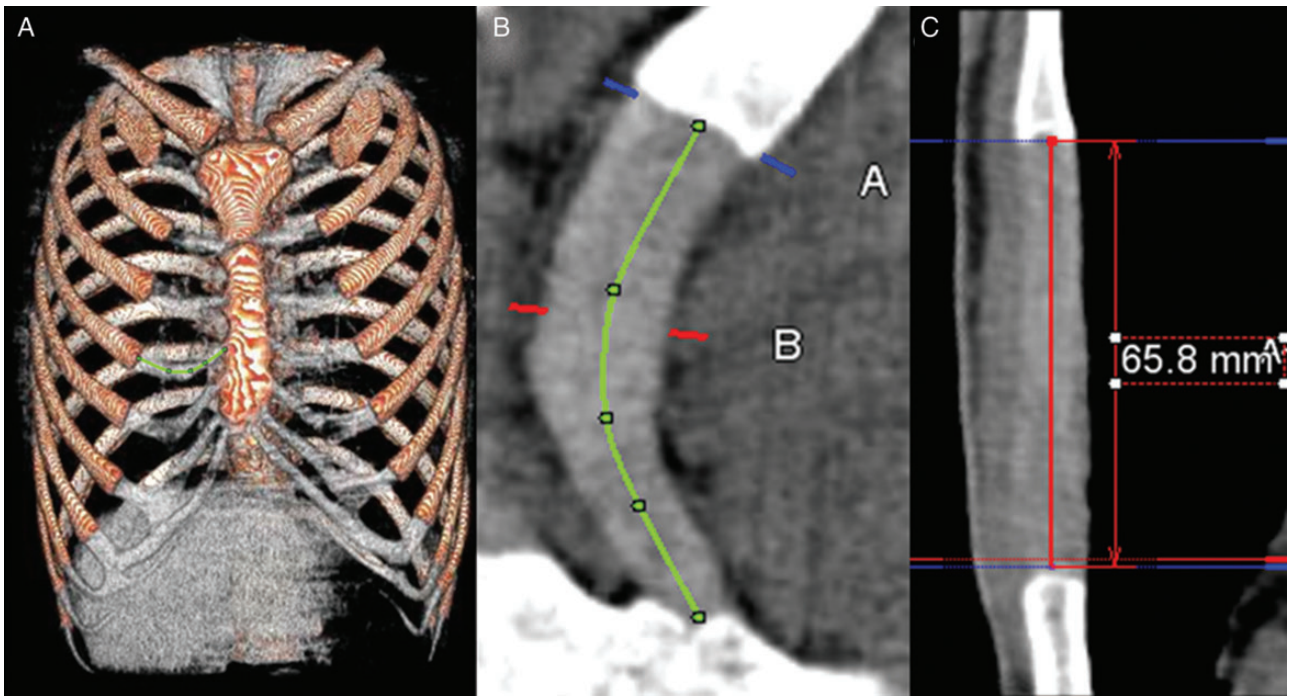

Figure 3: Measurement of the costal cartilage. 3-D VR image was reconstructed for the costal cartilage (**A**). The full length of the right fourth costal cartilage was semi-automatically traced using a curved multi-planar reformatted technique with manual correction (**B**) and then measured by electronic caliper (**C**).

protruded side vs the opposite side, respectively. At the level of the sixth rib, the mean cartilage lengths were 97.8 ± 13.2 mm vs 99.8 ± 15.5 mm, the mean rib lengths were 283.8 ± 33.9 mm vs 283.9 ± 32.3 mm, the combined total cartilage and rib lengths were 381.7 ± 41.8 mm vs 383.7 ± 43.1 mm, and the mean C/R ratios were 34.7 ± 4.5% vs 35.2 ± 4.5%, on the more prominent and opposite sides, respectively (Figs. 4–7). There were no statistically significant differences between the two sides (Table 1).

Table 1: Comparison of the cartilage length, rib length, total combined cartilage and rib length, and the mean C/R ratio between the more protruded side and the opposite side at the fourth through sixth rib levels

Level	More protruded side (mean ± SD)	Opposite side (mean ± SD)	Lateral difference (mean ± SD)	*P*-value
The cartilage length (mm)				
4th	55.8 ± 9.8	55.9 ± 9.3	−0.086 ± 2.71	0.883
5th	70 ± 10.8	71.6 ± 10.8	−1.49 ± 4.39	0.125
6th	97.8 ± 13.2	99.8 ± 15.5	−1.95 ± 7.56	0.239
The rib length (mm)				
4th	265.8 ± 34.9	266.3 ± 32.9	−1.27 ± 3.58	0.111
5th	279.7 ± 32.7	280.6 ± 32.4	−0.86 ± 4.41	0.369
6th	283.8 ± 33.9	283.9 ± 32.3	−0.09 ± 5.07	0.934
The combined cartilage and rib length (mm)				
4th	321.1 ± 40.3	322.5 ± 37.7	−1.36 ± 4.26	0.150
5th	349.8 ± 39.7	352.2 ± 39.3	−2.36 ± 6.26	0.092
6th	381.7 ± 41.8	383.7 ± 43.1	−2.05 ± 8.43	0.268
The C/R ratio (%)				
4th	21.2 ± 3.5	21.1 ± 3.4	0.07 ± 1.1	0.761
5th	25.1 ± 3.3	25.6 ± 3.3	−0.48 ± 1.8	0.233
6th	34.7 ± 4.5	35.2 ± 4.5	−0.55 ± 2.8	0.377

Lateral difference = (value from the more protruded side)-(value from the opposite side).

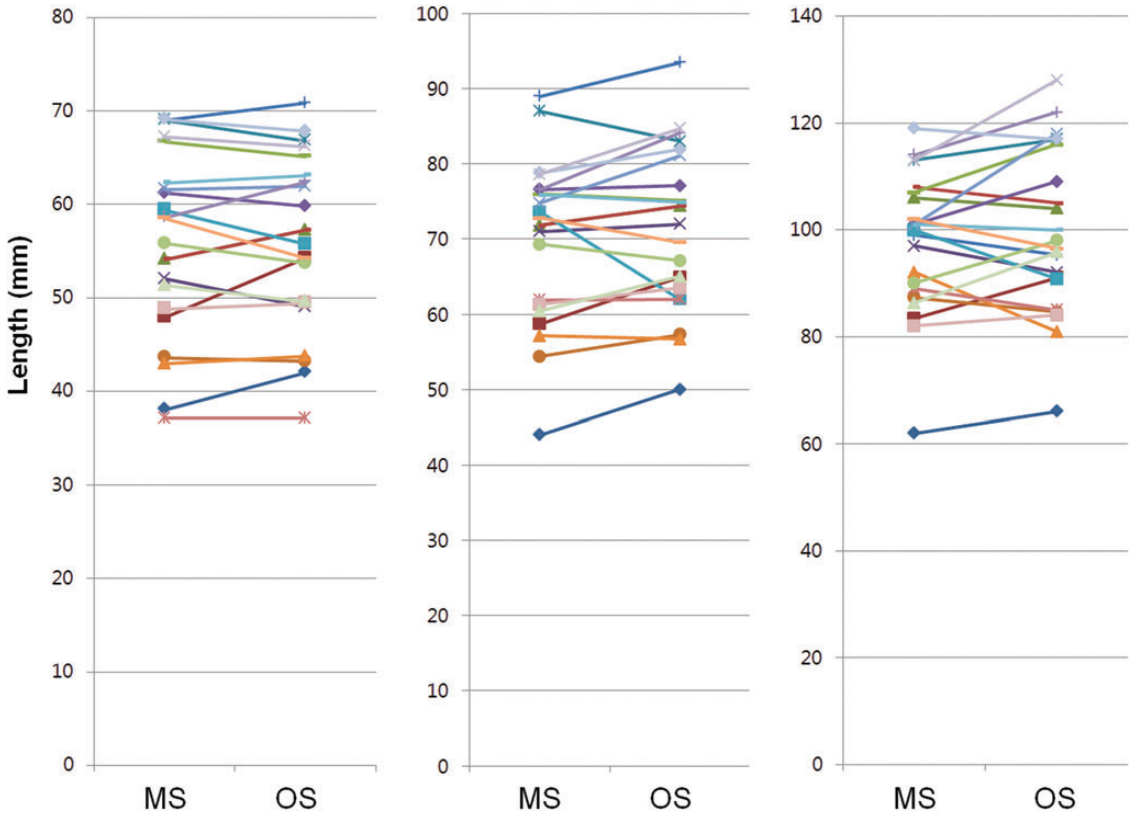

Figure 4: Differences in costal cartilage length. (**A**) Fourth cartilage, (**B**) fifth cartilage and (**C**) sixth rib. The lengths of the cartilage of the more protruded side at the fourth (A), fifth (B) and sixth (C) rib levels were not different from those of the opposite side (55.8 ± 9.8 vs 55.9 ± 9.3 at the fourth, 70 ± 10.8 vs 71.6 ± 10.8 at the fifth and 97.8 ± 13.2 vs 99.8 ± 15.5 at the sixth, $P > 0.05$). *MS: more protruded side; *OS: opposite side.

DISCUSSION

Our study measured the lengths of the ribs and the costal cartilage in patients with asymmetric pectus carinatum using 3-D CT images. Our results demonstrate that neither the rib length nor the cartilage lengths were different between the more protruded side and the opposite side in these patients.

The pathophysiology of pectus carinatum is not well established. About 25% of patients with pectus carinatum have a family history of some type of chest wall defect [2, 5]. Various diseases

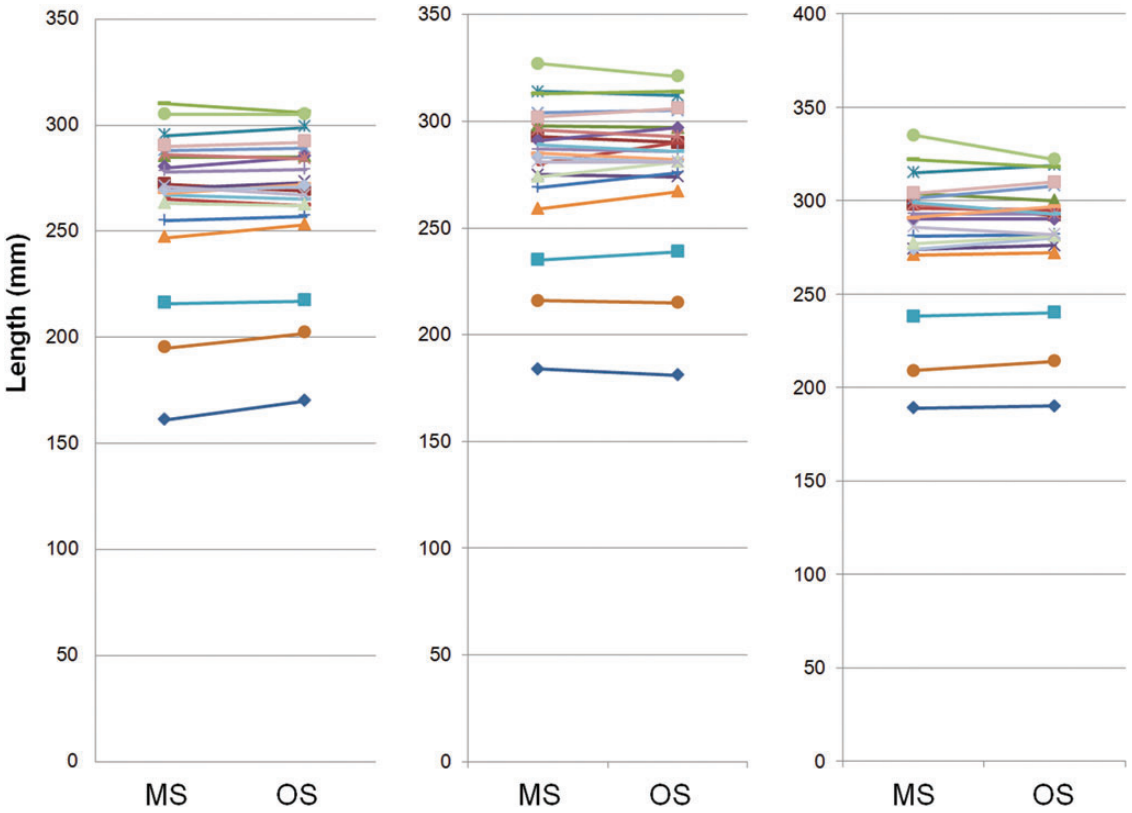

Figure 5: Differences in rib length. (**A**) Fourth rib, (**B**) fifth rib and (**C**) sixth rib. The rib lengths of the more protruded side at the fourth (A), fifth (B) and sixth (C) rib levels were not different from those of the opposite side (265.8 ± 34.9 vs 266.3 ± 32.9 at the fourth, 279.7 ± 32.7 vs 280.6 ± 32.4 at the fifth and 283.8 ± 33.9 vs 283.9 ± 32.3 at the sixth, $P > 0.05$). *MS: more protruded side; *OS: opposite side.

such as scoliosis, Marfan syndrome, osteogenesis imperfecta, Noonan syndrome, as well as some types of congenital heart disease have been associated with pectus carinatum [2, 6, 7]. Proposed explanatory hypotheses for pectus carinatum include the overgrowth of cartilage, the overgrowth of the ribs, the anomalous development of the diaphragm, a deformity of the sternum and a connective tissue abnormality [3, 8, 9]. Among these hypotheses, the exaggerated growth of costal cartilage is the most widely accepted potential mechanism for pectus carinatum as well as pectus excavatum. This theory is also the basis of surgical correction of pectus carinatum [3, 6]. Despite this, evidence supporting this hypothesis has been insufficient, and a potential aetiology for the cartilage overgrowth has not been established [2, 3].

In patients with asymmetric pectus excavatum, the rib length and cartilage length on the more severely depressed side have been shown to be either significantly shorter or not different from those on the opposite side [4]. In patients with symmetric excavatum, however, the C/R ratios were not larger than those in healthy controls [10]. These findings suggest that the overgrowth of costal cartilage may not sufficiently explain the occurrence of pectus excavatum. We hypothesized that, if overgrowth of the costal cartilage is the cause of the outward protrusion of the anterior chest wall seen in pectus carinatum, the costal cartilage lengths on the more protruded side would be greater than that on the opposite side. However, the lengths of the cartilage and ribs, the total combined length of the cartilage and rib and the C/R ratio in our study were not different between the more protruded side and the opposite side at any of the rib levels examined. Based on these findings, we conclude that overgrowth of the costal cartilage might not be the responsible factor in pectus carinatum.

Our study has several limitations that can be addressed in future studies. First, our sample size was rather small, as we included only 22 patients. Secondly, we measured only the fourth, fifth and sixth ribs and associated cartilage and did not include any other rib measurements. This could have been insufficient for proper analysis, as there are two major types of pectus carinatum, upper and lower, although lower pectus carinatum is more common [3]. In our study, all of the patients showed the lower type of pectus carinatum, in which the fourth through sixth costal cartilages contribute primarily to the observed sternal rotation. The 7th through 10th costal cartilages are typically fused together, and the 11th and 12th ribs are floating [4]. Therefore, we concluded that the measurement of the fourth, fifth and sixth ribs and the associated costal cartilages might be sufficient to evaluate asymmetric pectus carinatum. Finally, we only compared the length of the ribs and cartilages in patients with asymmetric pectus carinatum, without any controls. In our next study, the differences in these measurements in patients with symmetric pectus carinatum and those in healthy subjects will be evaluated.

In conclusion, the fourth, fifth and sixth rib lengths and costal cartilage lengths are not different between the more protruded side and the opposite side in patients with asymmetric pectus carinatum. These findings suggest that overgrowth of costal cartilage cannot explain the asymmetric protrusion of anterior chest wall and may not be the main cause of pectus carinatum.

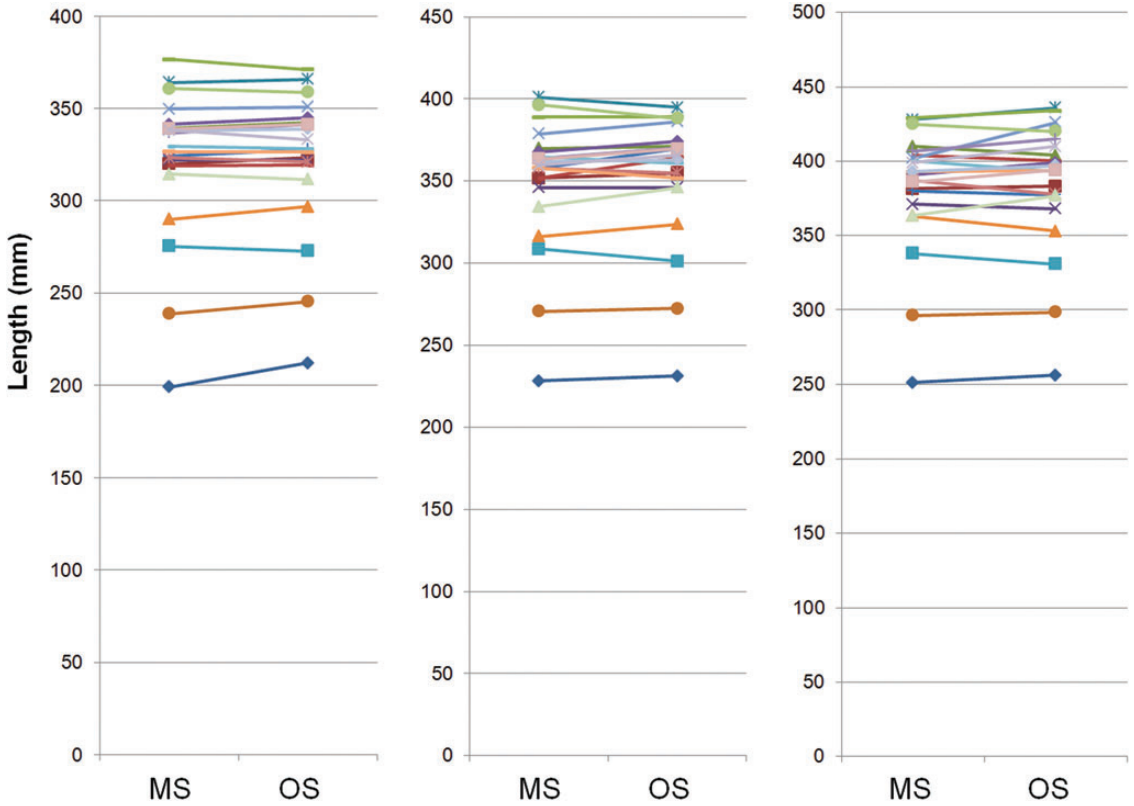

Figure 6: Differences in the total combined rib and costal cartilage length. (**A**) Fourth rib + cartilage, (**B**) fifth rib + cartilage and (**C**) sixth rib + cartilage. The total combined rib and cartilage length of the more protruded side at the fourth (A), fifth (B) and sixth (C) rib levels were not different from those of the opposite side (321.1 ± 40.3 vs 322.5. ± 37.7 at the fourth, 349.8 ± 39.7 vs 352.2 ± 39.3 at the fifth and 381.7 ± 41.8 vs 352.2 ± 39.3 at the sixth, $P > 0.05$). *MS: more protruded side; *OS: opposite side.

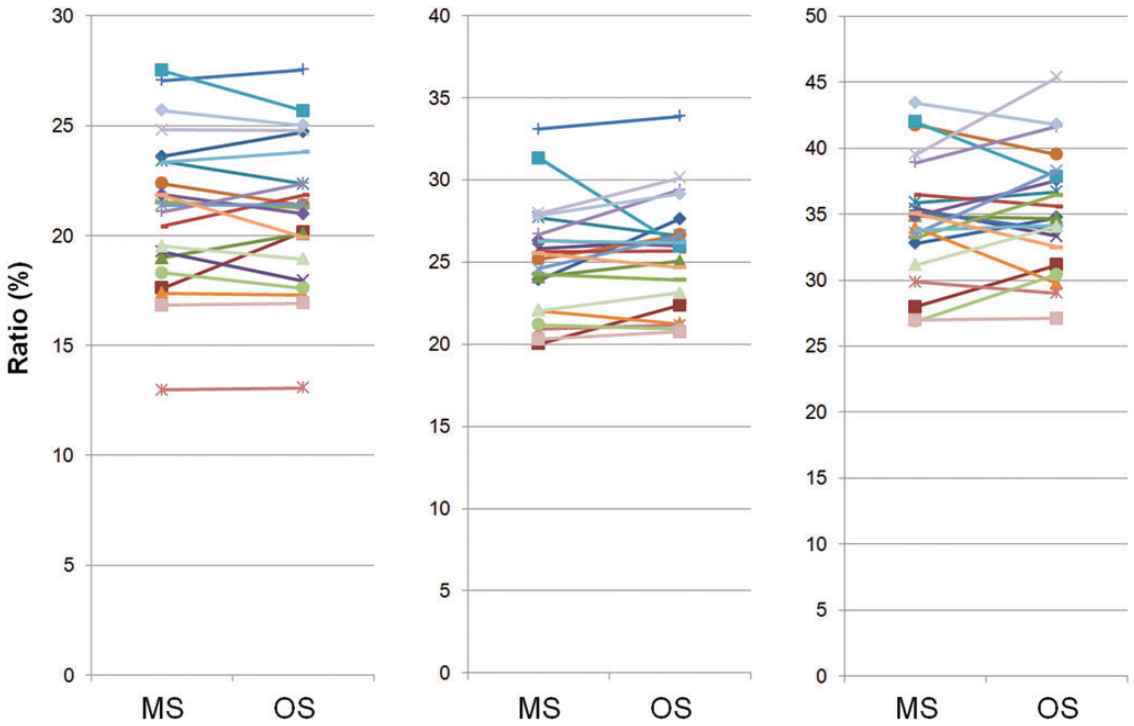

Figure 7: Differences of the C/R ratio. (**A**) Fourth C/R ratio and (**B**) fifth C/R ratio and (**C**) sixth C/R ratio. The C/R ratio on the more protruded side at the fourth (A), fifth (B) and sixth (C) rib levels were not different from those on the opposite side (21.2 ± 3.5 vs 21.1 ± 3.4 at the fourth, 25.1 ± 3.3 vs 25.6 ± 3.3 at the fifth and 34.7 ± 4.5 vs 35.2 ± 4.5 at the sixth, $P > 0.05$). *MS: more protruded side; *OS: opposite side.

Conflict of interest: none declared.

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eComment. Is overgrowth of costal cartilages the unique cause of pectus deformities?

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In their recent article, Park and coworkers hypothesized that 'if overgrowth of the costal cartilage is the cause of the outward protrusion of the anterior chest wall seen in pectus carinatum (PCar), the costal cartilage lengths on the more protruded side would be greater than that on the opposite side'. By studying the length of three paired rib/costal cartilages in 22 patients with asymmetric PCar, Park and coworkers discovered no statistically significant differences between the two sides and concluded 'overgrowth of the costal cartilage might not be the responsible factor in PCar' [1]. This is an interesting and meticulous study, which, however, raises some issues, as they outlined in their article. First, a rather small number of patients were enrolled. Second, only three paired rib/cartilages (fourth to sixth) were measured while either the third to seven paired rib/cartilages are involved in the standard form of PCar deformities; and second to fourth/five in the upper form (pouter pigeon breast) [2]. Last, no data are provided regarding the morphology/distortion of costal cartilages. Upon re-examining Figure 1 of the article (thoracic CT, axial view of an asymmetric PCar) it appears to us that the right cartilage area (more protruded side) is convex-shaped, while the contralateral cartilage area is concave, leading to sternum rotation, volume reduction of the rib cage on the left and, consequently, asymmetric deformity. Therefore this issue impedes a clear demonstration of the authors' hypothesis. In fact, the pathogenesis of anterior chest wall deformities remains elusive so far. Apart from connective tissue disorders or severe scoliosis, the cause of pectus deformities has been thought to be abnormal elongation of the ribs [3], and then costal cartilages [4], while Park and coworkers, and others [1, 5] claim that overgrowth of costal cartilages is not responsible, since there is no significant change in the costal cartilage length/rib length ratio (C/R ratio). In our opinion, this latter finding might be suggestive of a developmental cartilage dysfunction including the growth centre at the costochondral junction, leading to proportionate overgrowth of both cartilage and adjacent rib (thus with no change of C/T ratio) as shown bilaterally in patients with symmetric pectus deformities, and only on the more protruded side in patients with severe asymmetric pectus excavatum (PEx); or, on the contrary, proportionate hypoplasia of both cartilage and adjacent rib on the more depressed side (generally on the right) in patients with severe asymmetric PEx. Furthermore, this developmental cartilage dysfunction might be responsible for cartilage distortion and/or enlargement as shown in patients with protruding lowermost costal cartilage in a PEx context. Finally, overgrowth of costal cartilages is not the unique cause of pectus deformities but this cartilage structure plays a key role in the pathophysiologic and clinical features of the disease.

Conflict of interest: none declared

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eComment. Dilemma of the costal cartilage overgrowth in chest wall deformities

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We have read with interest the article by Chul Hwan Park *et al.* [1]. The authors suggest a hypothesis for the cause of pectus carinatum deformity. We wish to add our comment on this topic.

In the Results part of the article, only the mean values of the fourth, fifth, and 6th ribs are given. We think that the values of each rib compared with the rib on the opposite side would be more illustrative than the mean values. The mean values of the ribs may not be statistically important, but if the length of a rib is different from its opposite matching rib, this is important and should be mentioned. Also, the patients should be classified as having sternal rotation, scoliosis, or neither of these, and each of these groups should be evaluated separately. The ribs start from the vertebral body and slope down to the sternum with a unique angle in each individual. If a deformity in a vertebral body is present, a deformity of the thoracic cage is likely in that individual. Also, if sternal rotation is present, the costal cartilages will be depressed or will protrude to reach the sternum. We think that the length of the ribs is the same on the depressed and the protruded side in patients who have scoliosis or sternal rotation and that the primary deformity is the vertebral or sternal deformity.

Conflict of interest: none declared

Reference

[1] Park CH, Kim TH, Haam SJ, Lee S. 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.