

# Airway deformation in patients demonstrating pectus excavatum with an improvement after the Nuss procedure

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## Abstract

**Purpose** This study analyzed the inside appearance of the trachea and the main bronchus at the time of performing the Nuss procedure to test the hypothesis that the trachea and the main bronchus might be deformed by compression from great vessels due to sternal depression.

**Methods** A retrospective cohort study included 36 patients with pectus excavatum, who were treated using the Nuss procedure between July 2001 and December 2009. The primary outcome measures were the oblateness of the trachea and bilateral main bronchus. The oblateness in patients with pectus excavatum was compared with that of the control group. Their postoperative changes and the relationship between the Haller CT index were also analyzed.

**Results** The oblateness of the trachea and the left main bronchus in patients with pectus excavatum was significantly greater than that of the control group. A negative correlation was recognized between the percent vital capacity and the oblateness of the left main bronchus. The oblateness of the bilateral main bronchus significantly improved during the 2 years of bar placement.

**Conclusion** A significant deformation of the airways was demonstrated in patients with pectus excavatum, which improved after correcting it by means of the Nuss procedure.

**Keywords** Pectus excavatum · Nuss procedure · Bronchoscopy · Airway · Deformation · Pulmonary function

## Introduction

Abnormal electrocardiogram findings and systolic heart murmurs were first described in patients with pectus excavatum more than five decades ago [1, 2]. In addition to decreased pulmonary function [3–9], physiological impairment and exercise intolerance, a decreased cardiac function with postoperative improvement have been reported in pectus excavatum patients [4, 7, 10–12]. Also, mitral valve prolapse and regurgitation with cardiac chamber compression have been frequently detected [13], which have recovered after corrective surgery [14]. These cardiac abnormalities indicate that the heart is compressed and displaced leftward with concomitant rotation due to sternal depression [15, 16]. The compression, displacement or rotation of the heart may affect the position of great vessels such as the ascending aorta and the pulmonary trunk in connection with the bilateral main pulmonary arteries, which may press the trachea or the bilateral main bronchus, resulting in the deformation of the airways [17–19].

Despite these aforementioned observations, the influence of sternal depression on the airways in patients with pectus excavatum is still unknown. The purpose of this study was to analyze the inside appearance of the trachea and the main bronchus by bronchoscopy at the time of the Nuss procedure [20] and the bar removal, to test the hypothesis that the trachea and the main bronchus might be deformed by the compression due to sternal depression, and that this deformation might improve by correcting such a chest deformity by means of the Nuss procedure.

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## Materials and methods

### Study population

This cohort study included 36 patients with pectus excavatum who were treated by the Nuss procedure in the Department of Pediatric Surgery, Osaka University Hospital between July 2001 and December 2009. None of these 36 patients demonstrated dyspnea. Nine patients who were treated for other diseases (subglottic stenosis,  $n = 2$ ; laryngomalacia,  $n = 1$ ; pulmonary alveolar proteinosis,  $n = 1$ ; primary pulmonary hypertension,  $n = 1$ ; foreign body,  $n = 1$ ; bronchial pleomorphic adenoma,  $n = 1$ ; mediastinal tumor,  $n = 1$ ; and pulmonary sequestration,  $n = 1$ ), accompanied by the absence of chest wall deformities during the same period, were also analyzed as a control. The medical records including videotapes of a bronchoscopic examination performed at the time of the primary Nuss procedure and the subsequent bar removal were retrospectively studied. The cases whose recorded video images did not meet the requirement for analysis, as outlined below, were excluded. This study was performed in accordance with the rules of the Institutional Review Board of Osaka University Hospital.

### Determination of the airway oblateness

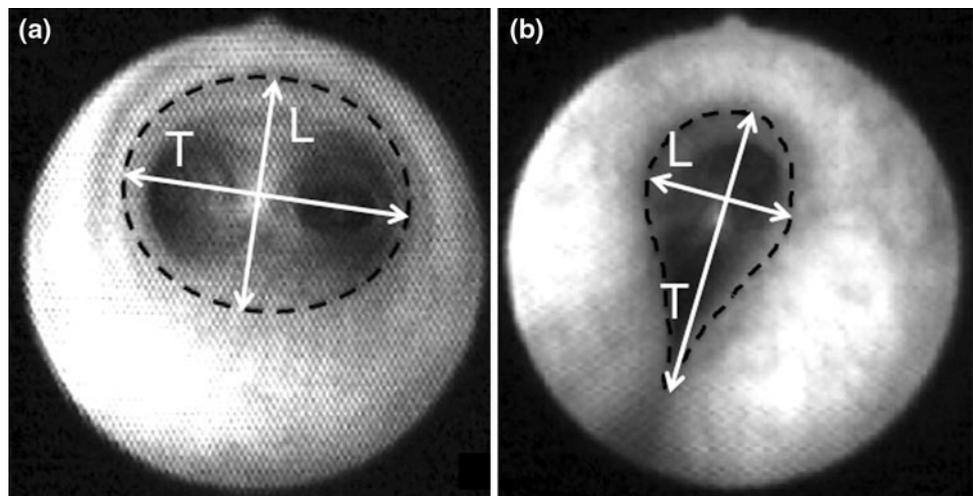
The primary outcome measures were the oblateness of the trachea and the bilateral main bronchus. The oblateness measurements were made from the still images of bronchoscopy. Bronchoscopic examinations were performed with a flexible fibroscope (FB-8V, FI-10BS; PENTAX, Tokyo, Japan) having an outer diameter of 2.8 or 3.5 mm, and a directable tip, by insertion through an endotracheal tube under general anesthesia without spontaneous respiration. The airway pressure at the time of measurement

was equal to the atmospheric pressure. The distal end of the trachea and the bilateral main bronchus were observed from the center of the proximal lumen to view the inner face of the airways with an equal endoscopic angle. The oblateness of the airways was determined at the distal end of the trachea and at the most humilis portion in the bilateral main bronchus. Motion pictures of these airways were recorded using digital videotape; the still images were subsequently analyzed. In the analysis, an inner face of the measurement portion along the lines of the individual tracheal or bronchial cartilage ring were traced on each still image, and the longitudinal and transverse diameters belonging to the closed trace were measured (Fig. 1a, b). The oblateness of the airway was defined as the difference of the transverse diameter and the longitudinal diameter divided by the transverse diameter. The value represented the average of three measurements from three different still images in a portion of the airway. A bronchoscopic examinations was performed in a same manner at the time of the subsequent operation just following the bar removal.

### Collected data and analysis

The patients' preoperative demographics including ages, gender and Haller computed tomography (CT) index [20, 21] were collected. The results of pulmonary function tests including preoperative percent vital capacity (% VC) and the percent forced expiratory volume in the first 1 s (% FEV 1.0) were also determined and recorded. The oblateness of the airways in the patients with pectus excavatum was compared with that of the control group of patients. The relationships between the Haller CT index and the oblateness of each airway, and between the pulmonary function test and the Haller CT index or the oblateness were analyzed. The oblateness after the bar

**Fig. 1** Measurement of the longitudinal diameter ( $L$ ) and the transverse diameter ( $T$ ) of the trachea (a) and the left main bronchus (b). The trace was along the lines of the inner face of the individual tracheal or bronchial cartilage ring (dashed line). The oblateness of the airways was defined as the difference between the transverse diameter and the longitudinal diameter divided by the transverse diameter ( $(T - L)/T$ )



**Table 1** The demographics of patients with pectus excavatum and those of the control group

	Pectus excavatum	Control group	<i>p</i> value
Number	36	9	
Gender (male/female)	27/9	5/4	0.411
Age (years) <sup>a</sup>	8.7 ± 4.0	9.4 ± 4.4	0.681
Haller CT index <sup>b</sup>	5.0 (3.2–11.7)	NA	
Preoperative % VC (%) <sup>a</sup>	81.0 ± 11.2 ( <i>n</i> = 12)	NA	
Preoperative % FEV 1.0 (%) <sup>a</sup>	88.2 ± 11.5 ( <i>n</i> = 12)	NA	

NA not available

<sup>a</sup> Mean ± standard deviation

<sup>b</sup> Median and range

removal was compared with that measured before the Nuss procedure was performed.

**Statistical analyses**

The mean and standard deviation, or the median and range, were used to describe continuous variables, and the frequency was used to describe the categorical data. Student’s *t* test was used for comparison of continuous variables. A Wilcoxon signed rank test was used for comparison of pre- and post-operative values of the oblateness. Fisher’s exact test was used for analysis of categorical data. *P* values of <0.05 were considered to indicate statistical significance.

**Results**

Table 1 shows the demographics of the patients with pectus excavatum and those of the control group. There was no significant difference in age and gender of the patient group (Table 1). Pulmonary function test was conducted in 12 patients with pectus excavatum (average age, 11.3 ± 3.9 years), who could be reliably examined. The mean value of the preoperative % VC was 81.0 ± 11.2%; four patients did not measure up to 80% VC. The mean value of the preoperative % FEV 1.0 was 88.2 ± 11.5% and one patient did not measure up to 70% FEV 1.0.

The oblateness of the trachea and the left main bronchus of the children with pectus excavatum were significantly greater than that of the control group (Table 2). There was no significant difference in the oblateness of the right main bronchus between the patients with pectus excavatum and the control group (Table 2).

Although there was no correlation between the Haller CT index and the oblateness of the trachea or the left main bronchus (Fig. 2a, b), a positive correlation with a correlation coefficient of 0.443 was recognized between the Haller CT index and the oblateness of the right main bronchus (Fig. 2c).

Although no correlation was observed between the Haller CT index and the % VC, a negative correlation was seen with a correlation coefficient of –0.320 between the

**Table 2** The oblateness of the trachea, the left bronchus and the right bronchus

	Pectus excavatum	Control group	<i>p</i> value
Trachea	0.299 ± 0.097	0.217 ± 0.090	0.032*
Left main bronchus	0.418 ± 0.098	0.190 ± 0.048	<0.001*
Right main bronchus	0.310 ± 0.085	0.274 ± 0.069	0.198

The mean ± standard deviation

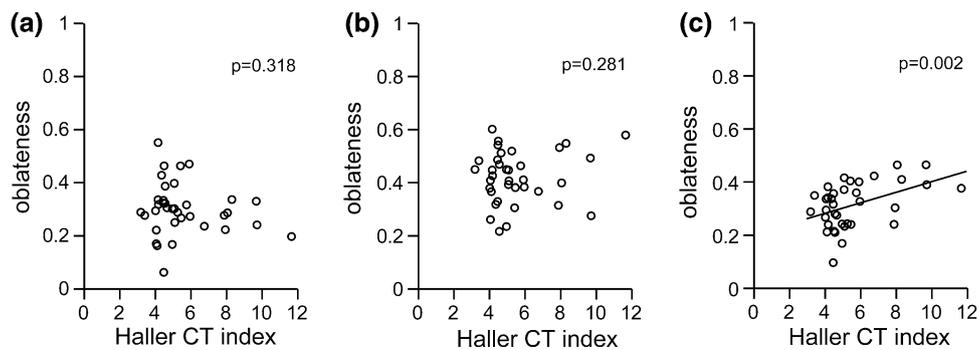
\* *p* < 0.05

Haller CT index and the % FEV 1.0 (Table 3, Fig. 3a). Concerning the relationship between the pulmonary functions and the oblateness of the airways, there was a negative correlation only between the oblateness of the left main bronchus and the % VC (correlation coefficient of –0.671) (Table 3, Fig. 3b).

The postoperative oblateness of the airways was measured in nine patients following bar removal at 2 years (24 ± 1.7 months) after the primary Nuss procedure, and the examination was recorded on videotape to meet the requirements for this analysis. Although no change was observed in the oblateness of the trachea (Fig. 4a), the oblateness of the right and left main bronchus significantly decreased during 2 years of the bar placement (Fig. 4b, c), thus indicating an improvement of the airway deformation by the correction of chest deformity with the Nuss procedure.

**Discussion**

Cardiovascular functions, which may influence both physiological impairment and exercise intolerance, have been investigated in pectus excavatum patients. Abnormalities in cardiac function, such as decreases in the stroke volume, cardiac output, maximum oxygen uptake and metabolic threshold, and mitral valve prolapse or regurgitation have been reported in such patients [7, 10–14, 22]. The causes of these cardiac dysfunctions are thought to be the compression of the heart due to sternal depression [7, 10, 13]. It has also been reported that there can be a leftward displacement of the heart with concomitant rotation, even if a functional impairment of the heart is not evident [15, 16]. These



**Fig. 2** Relationship between the Haller CT index and the oblateness of the trachea (a), the left main bronchus (b) and the right main bronchus (c). There was no correlation between the Haller CT index and the oblateness of the trachea or the left main bronchus. The

correlation coefficient between the Haller CT index and the oblateness of the right main bronchus was 0.443 with a linear regression of oblateness = 0.20 + (0.020 × Heller CT index)

**Table 3** Relationship between pulmonary function and the Haller CT index or the oblateness of the airways in patients with pectus excavatum

	% VC		% FEV 1.0	
	c.c.	p	c.c.	p
Haller CT index	0.172	0.257	−0.320	0.032*
Oblateness of the trachea	−0.178	0.241	−0.077	0.615
Oblateness of the left main bronchus	−0.671	<0.001*	−0.131	0.391
Oblateness of the right main bronchus	−0.065	0.669	−0.275	0.068

c.c. Correlation coefficient

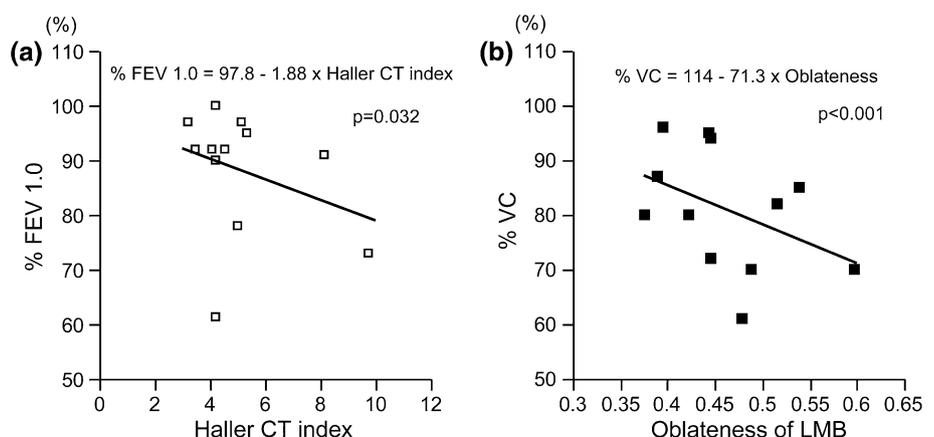
\*  $p < 0.05$

influences of the cardiac compression and displacement can be improved by a correction of the pectus excavatum [10, 11, 14, 22]. With regard to the pulmonary function, a respiratory impairment concerning vital capacity, total lung capacity, forced vital capacity and FEV 1.0 in pectus excavatum patients has been well described [3–6, 8, 9, 23]. However, the effect on the pulmonary function of a corrective surgery is still controversial. Some investigators have reported an

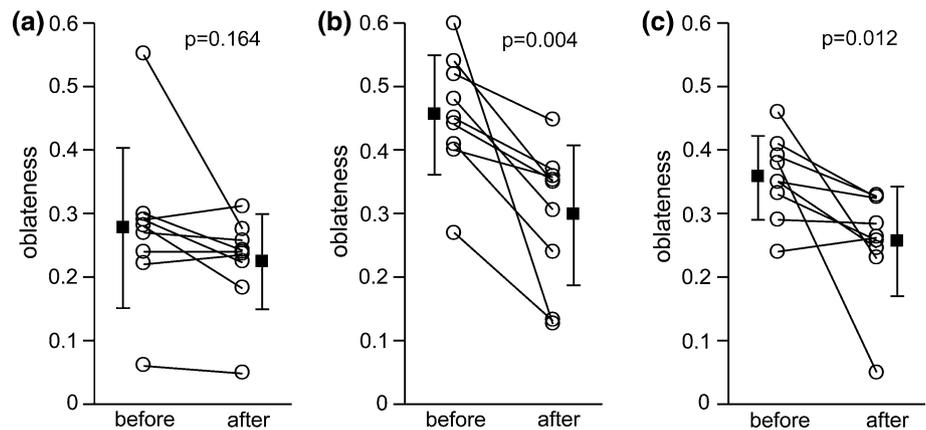
improvement of pulmonary function [4, 9, 22], while others have reported that no effect or even a deterioration was observed after the repair of pectus excavatum [6, 11, 23, 24]. In the present study, an obstructive lung defect was found in 4 patients, and a restrictive lung defect in 1 out of 12 patients who underwent pulmonary function test preoperatively. Therefore, while there have been numerous systematic studies concerning the relationship between chest deformity and cardiopulmonary function in pectus excavatum patients, the influence of chest deformity on the airways has not hitherto been revealed.

It has been reported that either tracheobronchial compression or secondary tracheobronchomalacia occurs because of abnormalities of the great vessels [25–27]. Tracheal flattening has also been recognized in patients with straight back syndrome, which has a similar type of chest deformity as pectus excavatum [19]. In these cases, two different mechanisms of airway compression by the great vessels were assumed from an anatomical aspect: one was the compression by an ascending aorta to the lower trachea, including the origin of the right main bronchus; and the other, by a pulmonary trunk together with the left pulmonary artery to the carina through the end of the left

**Fig. 3** Data on the CT index and oblateness. **a** The relationship between the Haller CT index and % FEV 1.0 (open squares) indicated a negative correlation with a correlation coefficient of  $-0.321$ . **b** The relationship between the oblateness of the left main bronchus (LMB) and % VC (closed squares) indicated a negative correlation with a correlation coefficient of  $-0.671$



**Fig. 4** The changes in the oblatenesses before and after the Nuss procedure of the trachea (a), the left main bronchus (b) and the right main bronchus (c) in the nine patients who underwent subsequent measurements of oblateness at the time of bar removal. The *open circles* represent each of the values, the *closed squares* express the mean values and the *error bars* depict the standard deviation



main bronchus. If the backward shift of great vessels due to sternal depression has a similar mechanism acting on the airways, pectus excavatum may likely be a cause of tracheobronchomalacia. A previous study reported five cases of segmental bronchomalacia of the left main bronchus associated with pectus excavatum [28]. We also previously reported pexis of the ascending aorta and pulmonary trunk to the sternum at the time of sternal elevation to improve tracheobronchomalacia in patients with pectus excavatum [29].

As there has been no systematic study regarding the effect of sternal depression to the airways in pectus excavatum, we tested the hypothesis that the trachea and the main bronchus might be deformed by the compression due to sternal depression, which might be improved by an operative correction. To estimate the airway deformation and their improvement, we analyzed the inside appearance of the airways using bronchoscopy before and after the corrective surgery. Although other modalities such as CT may be available to assess the inside deformation of the airways, an appropriate specialized technique using three-dimensional reconstruction may be necessary to measure the inner diameter at a cross section orthogonal to the axis of the airway lumen. In the bronchoscopic examination, we placed the observing point at the center of the airway lumen with due attention paid to equal inspection of the airways to trace the individual cartilage ring at an even distance.

In the present study, the oblateness of the right main bronchus demonstrated a positive correlation with the Haller CT index, while it was not greater than that of other diseases. On the other hand, the oblateness of the trachea and the left main bronchus of patients with pectus excavatum were greater than that of other diseases, although no correlation with the Haller CT index was observed (Fig. 2). These contrasting results may therefore indicate that the right main bronchus is directly affected by the backward displacement of the ascending aorta, which is located beneath the sternum,

just above the right main bronchus. On the contrary, the lower trachea including carina through the left main bronchus may be compressed by the left main pulmonary artery in connection with the pulmonary trunk, which is displaced leftward and rotated concomitantly independent of the degree of sternal depression [17, 18, 28].

The compression to the right main bronchus might not influence the restrictive lung defects, in spite of the negative correlation between the Haller CT index and the % FEV 1.0, because the correlation between the oblateness of the right main bronchus and the FEV 1.0 did not reach significant difference (Fig. 3, Table 3). It has been reported that the degree of sternal depression correlated with the decrease of vital capacity [5, 6]. However, our results demonstrate a significant negative correlation of the % VC with the oblateness of the left main bronchus independent of the Haller CT index (Table 3, Fig. 2), which may imply that, not the sternal depression itself, but the cardiac leftward displacement directly influences the decreased % VC. One study reported that children with pectus excavatum have decreased perfusion and loss of volume in the left lung, which correlated well with the degree of left displacement of the heart, while there was no correlation with the degree of sternal depression [30]. The narrowing of the left main bronchus is probably not a cause of the decrease in % VC, but a result of cardiac leftward displacement. This may explain the loss of lung capacity.

Our study has demonstrated that the deformation of the trachea and left main bronchus, which are relevant to decreased pulmonary function, can be improved by the Nuss procedure (Fig. 4). These results indicate that the airways of the patients can be reversibly recovered. In several patients, we compared the deformation of the trachea and the left main bronchus just before and after the bar placement during the Nuss procedure and found no change during the bar placement (data not shown). The oblateness of the bilateral main bronchus improved, probably by degrees, during the 2-year period of bar placement.

The mean oblateness of the bilateral main bronchus in the nine cases (left main bronchus,  $0.455 \pm 0.095$ ; right main bronchus,  $0.355 \pm 0.066$ ) in whom the bars were removed was higher than that of the other 27 cases. Nevertheless, no statistically significant difference was observed between the oblateness of the 9 patients and the other 27 patients. The improvement of the cardiopulmonary function after the Nuss procedure in pectus excavatum patients has been well demonstrated [9, 11, 14, 22]. Our results may also suggest the concomitant recovery of the airway deformation along with the recovery of pulmonary function, although, because of the small number of patients who were presently studied, our results await verification by a prospective study in a larger series.

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