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# Nuss procedure for patients with pectus excavatum with a history of intrathoracic surgery

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## KEYWORDS

Pectus excavatum;  
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Congenital diaphragm  
hernia;  
Pleural adhesion;  
Postoperative  
complication

**Abstract** *Background:* The aim of this study was to demonstrate the feasibility and safety of the Nuss procedure for patients with pectus excavatum (PE) with a history of intrathoracic surgery.

*Patients:* From April 2010 to December 2013, we performed 6 cases of PE repair in patients with a history of intrathoracic surgery. The causes of previous operations were congenital cystic adenomatoid malformation in 4 patients and congenital diaphragmatic hernia in 2. The patients' median age was 5 years (range, 4–9 years) and median preoperative pectus severity index was 4.63 (range, 3.42–10.03). Their intraoperative and postoperative courses were reviewed retrospectively.

*Results:* The mean overall operation time was  $127.5 \pm 17.0$  minutes, and the mean operation time for endoscopic pneumolysis was  $28.8 \pm 12.3$  minutes. Intraoperative exploration for pleural adhesion revealed that the endoscopic approach in the previous operation was associated with low pleural adhesion, and the open thoracotomy or laparotomy approach was associated with low to high pleural adhesion. One patient developed a pneumothorax on the first postoperative day. All the other patients had uneventful postoperative courses. All the patients received bar removal 2–3 years after bar insertion. One patient developed atelectasis after bar removal. All the other patients had an uneventful postoperative course. The mean postoperative follow-up time after bar removal was  $20.1 \pm 14.7$  months.

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**Conclusions:** History of intrathoracic surgery seems not a contraindication for the Nuss procedure. However, perioperative complications should be carefully monitored in both the bar insertion and removal operations.

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**Table 1** Patients' characteristics. The indications for the previous operations were congenital cystic adenomatoid malformation in 4 patients and congenital diaphragmatic hernia (CDH) in 2. The lateralities of the previous surgeries were in the right side in 3 patients and in the left side in the other 3 patients. An endoscopic approach was used in 2 patients, and either an open thoracotomy or a laparotomy approach was used in 4 patients. Five patients had asymmetric deformities, and 4 patients showed a more dominant depression on the previously operated side.

Case	Age	Sex	PSI	Asymmetry	Previous Ope.	Endoscopy/Open surgery
1	5	M	5.81	None	CCAM (R)	Endoscopy
2	4	F	4.47	R > L	CDH	Open laparotomy
3	5	M	4.79	L > R	CCAM (L)	Endoscopy
4	5	F	10.03	R > L	CCAM (R)	Open thoracotomy
5	9	F	3.42	R > L	CCAM (R)	Open thoracotomy
6	6	M	4.11	L > R	CDH	Open laparotomy
Median	5.0		4.63			

CCAM: congenital cystic adenomatoid malformation, CDH: congenital diaphragmatic hernia, RL: right lower lobe, LL: left lower lobe, RU: right upper lobe.

## Introduction

Since minimally invasive repair of pectus excavatum (PE) was introduced by D. Nuss in 1998,<sup>1</sup> this procedure (Nuss procedure) became the gold standard treatment for PE in prepubertal patients. The application of the Nuss procedure was initially relatively limited; however, accumulated experiences have widened the application to more difficult cases, such as adult,<sup>2</sup> severe and/or asymmetry deformity,<sup>3</sup> and recurrent cases.<sup>4-7</sup> Along with this expansion of application, the relative risk of complications such as bar rotation, pneumothorax, and severe cardiopulmonary injury has increased.<sup>5</sup> Various attempts have been made to reduce the risk of severe complications recently, including utilizing the originally designed sternum elevator or bar insertion with the extrathoracic approach.<sup>8,9</sup>

History of intrathoracic surgery in the infant stage possibly impairs the physical growth of the chest wall and is a risk factor of thoracic deformities such as PE and pectus carinatum.<sup>8-10</sup> With the recent advancement of the survival rate in such patients, surgeons may encounter more frequently cases requiring treatment for chest deformity in patients with a history of intrathoracic surgery. In this report, we demonstrate the feasibility and safety of the Nuss procedure for PE patients with a history of intrathoracic surgery.

## Patients and methods

The study was approved by internal ethics committee of Nagoya University Hospital. From April 2010 to December 2013, we performed 6 cases of PE repair in patients with a history of intrathoracic surgery. The case summaries are shown in Table 1. Of the patients, 2 were male and 4 were female. The causes of the previous operations were congenital

ital cystic adenomatoid malformation (CCAM) in 4 patients and congenital diaphragmatic hernia (CDH) in 2 patients. The lateralities of the previous surgeries were in the right side in 3 patients and in the left side in the other 3. In the previous operation, an endoscopic approach was used in 2 of the 6 patients, and either an open thoracotomy or a laparotomy approach was used in 4 patients. The first operation for CCAM or CDH was performed in the neonatal stage, during which time the chest deformity was not yet clear. The deformity gradually developed during the growth stages, and the patients were referred to our department at the age of 3-8 years for consultation regarding their chest deformities. Five patients had asymmetric deformities, and 4 had a more dominant depression on the previously operated side.

The median age at PE repair was 5 years (range, 4-9 years), and the median preoperative pectus severity index (PSI) was 4.63 (range, 3.42-10.03). The intraoperative and postoperative courses of the patients were reviewed retrospectively.

## Operative procedure

All 6 patients underwent the Nuss procedure. The patient was positioned supine with both arms raised to the head. Before the operation, ultrasonography (US) examination was performed to evaluate for pleural adhesion. If adhesion was detected, it was marked and trocars were inserted to avoid the adhesion site. A skin incision was made on both sides of the chest, and an under-muscle dissection was made where the bar was to be placed. Two 5-mm Endopath XCELs (Ethicon Inc., USA) were placed in the sixth intercostal space (endoscope) and in the incision that was created for bar insertion. These trocars were placed in the



**Figure 1** Preoperative appearance (left, middle) and computed tomography image (right) of the patient. The preoperative pectus severity index was 3.42.

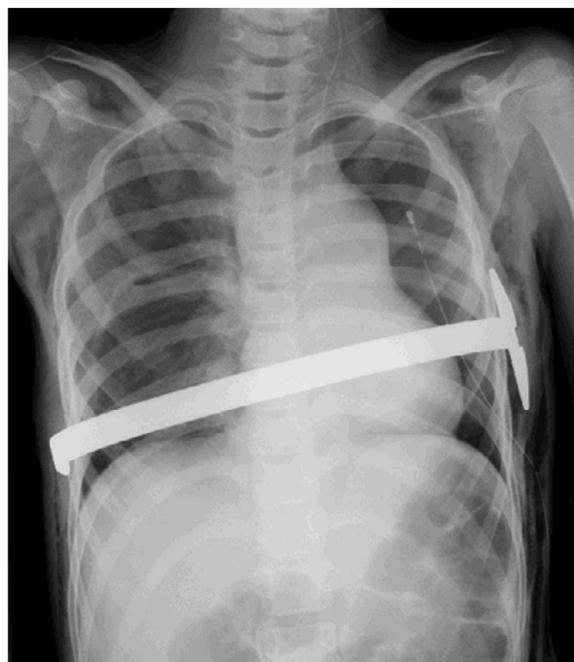
previously operated side. The thoracic endoscope was inserted, and endoscopic observation of the adhesion was classified into mild, moderate, or severe, where mild adhesion indicates a localized small adhesion without restriction of lung movement, moderate adhesion indicates a localized adhesion with restriction of lung movement, and severe adhesion indicates adhesion of the entire ipsilateral lung surface. The pleural adhesion was then dissected, if needed, with Maryland bipolar forceps. The visceral pleura of the lung or mediastinal organs was freed from the thoracic wall, which was elevated after the pectus bar insertion. In severe adhesion cases, an additional port was made for the dissector when necessary. After the pneumolysis, the Nuss procedure was performed following the standard procedure, utilizing a pectus bar (Biomet Microfixation Inc., USA). In our case series, no thoracic drainage was placed because the intraoperative bleeding was minimum.

### Illustrative case

A 9-year-old girl was referred to our department because of chest wall deformity. She had undergone an open right lower lobectomy due to CCAM at 3 months of age. The preoperative appearance and CT scan showed PE with a CT index of 3.42 (Figure 1a). An endoscopic pneumolysis followed by the Nuss procedure was planned. Intraoperative exploration for plural adhesion revealed a severe adhesion. On the first postoperative day, she developed a pneumothorax on the contralateral side, and an 8-Fr Argyle trocar aspiration kit (Covidien, Ireland) was inserted in the pneumothorax cavity (Figure 2). She recovered with continuous drainage for 5 consecutive days. Two years after the bar placement, bar removal was performed. Twenty-four months after the bar removal, no complication occurred. The configuration of her chest wall was good (Figure 3).

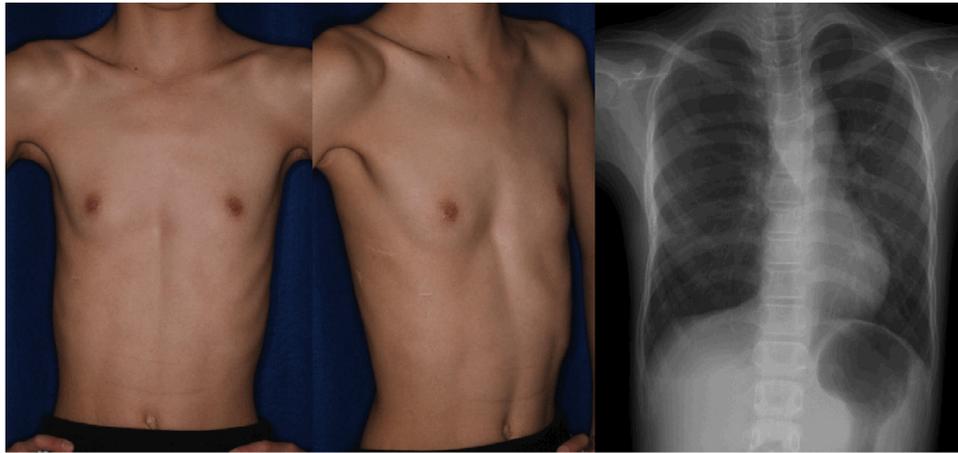
### Results

The results are summarized in Table 2. The mean ( $\pm$ SD) operation time for the Nuss procedure was  $127.5 \pm 17.0$  minutes, and the mean dissection time for endoscopic pneumolysis was  $28.8 \pm 12.3$  minutes. Intraoperative exploration to determine the pleural adhesion severity revealed that the



**Figure 2** Radiography image of the patient after bar insertion. Pneumothorax developed in the contralateral side of the previously operated side.

endoscopic approach for the previous operation was associated with mild pleural adhesion, and the open thoracotomy or laparotomy approach was associated with mild to severe pleural adhesion. In the postoperative course, 1 patient (case 5) developed a pneumothorax on the contralateral side (left side) of the previously operated side (right side) on the first postoperative day. All the other patients experienced uneventful postoperative courses after bar insertion. All the patients underwent bar removal 2-3 years after the Nuss procedure. One patient (case 3) developed atelectasis after bar removal and was treated with physiotherapy. In this patient, mild pleural adhesion was observed during the bar placement operation; however, severe adhesion was observed in the bar removal operation; therefore, extensive pneumolysis was performed before bar removal. The other 5 patients experienced uneventful



**Figure 3** Appearance (left, middle) and radiography image (right) of the patient after bar removal. Pectus excavatum has been corrected.

**Table 2** Operation time, dissection time, adhesion level, and postoperative complications in the bar insertion and removal. The mean postoperative observation period was  $20.1 \pm 14.7$  months.

Case	Previous Ope.	Ope. Time, min	Dissection Time, min	Adhesion Severity	Postoperative Complication Bar insertion	Bar removal, yrs	Postoperative Complication Bar removal	Follow-up Period, mo
1	CCAM (R)	132	17	Mild	None	2.1	None	24.0
2	CDH	111	8	Mild	None	3.1	None	48.6
3	CCAM (L)	137	43	Mild	None	3.4	Atelectasis	12.1
4	CCAM (R)	158	36	Moderate	None	4.0	None	12.1
5	CCAM (R)	117	31	Severe	Contralateral Pneumothorax	3.0	None	12.0
6	CDH	110	38	Severe	None	2.7	None	12.0
Mean		127.5	28.8			3.0		20.1

postoperative courses after bar removal. The mean follow-up period after bar removal was  $20.1 \pm 14.7$  months.

## Discussion

Congenital anomaly in the thoracic region and history of intrathoracic surgery are considered risk factors of thoracic deformities such as PE and pectus carinatum.<sup>8-10</sup> The mechanisms of such thoracic deformities are not clearly understood but may include inflammation of intrathoracic tissue due to surgical intervention or pulmonary inflammatory disease, expansion or deflation of the thoracic space, and increased breathing work during the neonatal and perioperative stages. As the survival rate has increased in such patients with congenital intrathoracic anomalies,<sup>11-13</sup> the incidence of thoracic deformity in patients with a history of intrathoracic surgery is considered to increase as well.

Reports of complication rates after the Nuss procedure vary among studies that included hemothorax (1-5%), pleural effusions (2-9%), infections (3-9%), and a requirement of additional operations (4-19%). The total complication rate also varies between 15% and 27%.<sup>2,14</sup> Several reports cited a slightly higher complication rates in redo surgery. Nuss and others<sup>5,7</sup> reported that patients who required a redo surgery had high rates of complications including pneumothorax re-

quiring a chest tube (14%), hemothorax (8%), pleural effusion requiring drainage (8%), pericarditis (4%), pneumonia (4%), and wound infection (2%). These are considered to result from pleural and pericardial adhesion and consequent difficulty of dissection on the previously operated site. As inflammation caused by surgery occurs, local resident cells produce angiogenetic/vasculogenetic factors, which result in the formation of vascular-rich scars. Furthermore, the need for extensive dissection leads to persistent postoperative pain and consequent postoperative atelectasis and pneumonia.

Various attempts have been made to manage pleural and pericardial adhesions. Miller<sup>6</sup> used a subxiphoid incision in the procedure for substernal management, and Schaarschmidt et al.<sup>20</sup> performed an extrapleural placement of the bar. Yüksel et al.<sup>10</sup> used the incision for bar placement as the port for the pneumonolysis procedure. In our case series, US examination was useful for evaluating the severity of the pleural adhesion and for deciding the placement site of the endoscopic port. The sensitivity and specificity of US for detecting pleural adhesion were reported to be 80.6% and 96.1%, respectively.<sup>11</sup> In the US examination of pleural adhesion, the degree of pleural adhesion can be assessed on the basis of the "gliding sign," which is a bright to-and-fro movement visible between the visceral and the parietal pleurae and is synchronized with breathing

movements (Videos 1 and 2). With regard to the previous intrathoracic surgery, the use of an endoscopic approach in the primary operation has been proved to decrease the pleural adhesion considerably.<sup>12</sup> This is consistent with our study, in which all cases with the endoscopic approach in the primary operation were associated with low adhesion of the pleural cavity. Several authors<sup>13-15</sup> reported the potency of antiadhesion agents administered after the intrathoracic surgery in experimental studies.

To date, only few reports have been published that describe the feasibility of the Nuss procedure for patients with a history of other intrathoracic surgery. Metzelder et al.<sup>16</sup> described 5 patients with a history of prior intrathoracic surgery among their 110 patients indicated to undergo the Nuss procedure. Of the 5 patients, 3 had CDH, 1 had CCAM, and 1 had bronchopulmonary dysplasia. Although all 5 patients required adhesiotomy during the operation, no severe perioperative or postoperative complications were reported. In our case series, the Nuss procedure was performed in 6 patients with a history of intrathoracic surgery and the bar was removed in all the patients. One patient developed a pneumothorax during the bar placement operation, and 1 patient developed atelectasis during the bar removal operation. Although these complications are not life threatening and can be managed conservatively, they are more frequent in the patients with than in those without a history of intrathoracic surgery. Careful postoperative monitoring and management should be performed in such patients.

## Conclusions

In conclusion, the Nuss procedure is a feasible option for the repair of thoracic deformities in patients with a history of intrathoracic surgery. However, postoperative complications should be monitored carefully in such patients. US examination is useful for evaluating pleural adhesion preoperatively.

## Conflict of interest

None.

## Funding

None.

## Supplementary material

Supplementary material associated with this article can be found, in the online version, at doi:[10.1016/j.bjps.2018.12.027](https://doi.org/10.1016/j.bjps.2018.12.027).

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