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ORIGINAL ARTICLE

Secondary spontaneous pneumothorax associated with emphysema and ruptured bullae at the azygoesophageal recess

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Abstract
Purpose. With secondary spontaneous pneumothorax (SSP) associated with emphysema, lesions responsible for pneumothorax can be located anywhere along the lung surface. Among such lesions, ruptured bullae at the azygoesophageal recess (AER) have received little attention thus far.
Methods. We conducted a retrospective study of 38 right SSP patients with emphysema who underwent surgery. Among them, we reviewed the clinical characteristics and technical problems of patients with surgically proven ruptured bullae at the AER.
Results. Ruptured bullae at the AER were found in 10 of 38 patients. They accounted for 26.3% of all 38 patients and for 66.7% of 15 patients whose bullae at the AER were identified by preoperative computed tomography (CT). On CT, all the bullae were relatively large and oriented in a predominantly vertical axis. At surgery, they were confirmed as white, thin-walled structures originating from the mediastinal part of the apical segment of the right lower lobe. Surgery typically consisted of stapling bullectomy with video-assisted thoracic surgery. Technical problems in surgical treatment included poor mobilization of the base of the bulla and a restricted working space.
Conclusion. Bullae at the AER are common and possibly lead to rupture. The presence of a bulla at the AER seen by CT can be predictive of rupture. Although the AER is a unique location, video-assisted bullectomy is the method of choice for treating these lesions.

Key words Azygoesophageal recess · Computed tomography · Emphysema · Secondary spontaneous pneumothorax · Video-assisted thoracic surgery (VATS)

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Introduction

The treatment strategies of secondary spontaneous pneumothorax (SSP) with emphysema, the prevalent underlying disease, are still controversial.[1,2] Even if surgery is indicated, the surgical intervention is sometimes more difficult and operative morbidity and mortality higher than after intervention for primary spontaneous pneumothorax (PSP).[1,3–5] In SSP patents with emphysema (unlike young PSP patients), the ruptured lesions can be located anywhere on the lung surface. Among such lesions, ruptured bullae at the azygoesophageal recess (AER), a space characteristic of the right pleural cavity, has received little attention as a peculiar cause of pneumothorax thus far. We conducted a retrospective study of right SSP emphysematous patients associated with ruptured bullae at the AER confirmed at the time of surgery. The aim of this study was to clarify the peculiarity of bullae at the AER in patients with SSP with emphysema by reviewing the frequency of rupture, the morphological computed tomography (CT) features, and the technical problems associated with the surgical procedures.

Patients and methods

Patients and surgical indication

From February 2003 to April 2007 a total of 265 consecutive patients (293 procedures) underwent surgical intervention for spontaneous pneumothorax (SP) at the Department of General Thoracic Surgery, Numazu City Hospital. Among the patients, 187 had PSP and 78 SSP. Emphysema was the predominant underlying pulmonary disease in patients with SSP, with 69 of 78 (90%) SSP patients having emphysema. Among these 69 patients, 38 occurred on the right side and 31 on the left. Among these 38 right-side SSP patients, 37 were evaluated with a preoperative chest CT.

Our main indications for surgical intervention for SSP with emphysema included persistent (≥7 days) air leaks or short-interval recurrence, tension pneumothorax, relatively good performance status (0–2), and tolerance of general anesthesia. The CT diagnosis of emphysema relied on the detection of areas of low attenuation. Preoperative spirometry was not performed. We measured the three-dimensional diameters of the bullae at the AER on preoperative CT. Diameter measurements of the bullae were recorded as the mean ± SD.

Student's *t*-test was used for comparisons between dimension groups. $P < 0.05$ was considered statistically significant.

Surgical procedures

In SSP patients with emphysema, our surgical strategy can be summarized as follows: Video-assisted thoracic surgery (VATS) was the surgical approach of choice (with prompt conversion to mini-thoracotomy if needed) with minimal dissection of pleural adhesions; buttress stapling bullectomy; excision or closure of nonruptured white, thin-walled bullae if present; and no preventive pleurodesis.

For VATS procedures, three or four ports and a 10-mm flexible thoracoscope were used. Given the fragility of the underlying emphysematous tissue, a buttressed stapler with polyglycolic acid (PGA) sleeve (Neoveil, tube type; Gunze, Kyoto, Japan) for staple-line reinforcement was routinely used. Endoscopic staplers, with or without knife (Echelon 60, Endopath EZ45 or NK45; Ethicon Endo-Surgery, Cincinnati, OH, USA), were utilized. Mini-thoracotomy VATS consisted of a 5- to 8-cm skin incision and division of as little muscle as possible.

Postoperative follow-up

Follow-up after discharge continued for 1–2 months, and patients were then referred back to their primary care setting unless problems were detected at the initial follow-up. Long-term data about pneumothorax recurrence and current status were obtained by telephone interviews.

Results

The distribution of the ruptured bullae is shown in Table 1. Characteristics of all 38 right SSP patients with emphysema are summarized in Table 2. Bullae at the AER were identified by preoperative CT in 15 (39.5%) of 37 patients (Fig. 1). The lesions responsible for the air leak (the ruptured bulla) were intraoperatively identified in 35 (92.1%) of 38 patients. All the bullae localized at the AER originated from the most dorsal part of the apical segment of the lower lobe. In 10 patients, the bullae at

Table 1 Distribution of ruptured bullae in the right lung

Location of ruptured bulla	No. of patients
Upper lobe	18 (47.4%)
Apex	13 (34.2%)
Other than apex	5 (13.2%)
Middle lobe	4 (10.5%)
Lower lobe	13 (34.2%)
AER	10 (26.3%)
Other than AER	3 (7.9%)
Not identifiable	3 (2.6%)

AER, azygoesophageal recess

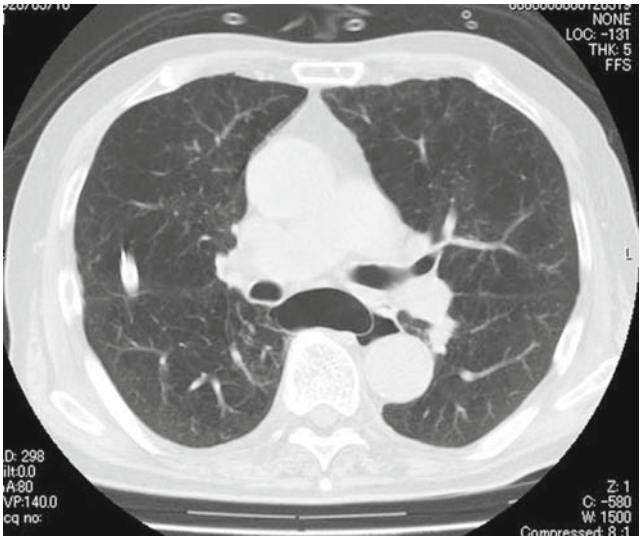

Fig. 1 Chest computed tomography (CT) showing a bulla at the azygoesophageal recess

Table 2 Characteristics of all 38 right SSP patients associated with emphysema

Air leak site	AER	Other than AER	Unidentifiable	Total
No. of patients (male/female)	10 (10/0)	25 (24/1)	3 (3/0)	38 (37/1)
Age (years)[a]	70.0 (60–81)	66.8 (45–84)	75.3 (61–85)	68.3 (45–85)
Preop. identification of the AER bulla	10 (100%)	4 (16.0%)	1 (33.3%)	15 (39.5%)[b]
Surgical approach (VATS/M-T VATS)[c]	6/4	17/8	0/3	23/15
Chest tube removal (days)[a]	1.5 (1–3)	1.25 (1–4)[c]	2.3 (1–4)	1.4 (1– 4)[c]
Operating time (min)[a]	82.7 (37–210)	74.8 (29–142)	116.3 (90–162)	80.2 (29–210)
Postop. hospital stay (days)[a]	5.1 (3–7)	5.1 (1–25)[c]	8.0 (5–9)	5.3 (1–25)[c]
Postop. in-hospital death	0	1 (4.0%)	0	1 (2.6%)
Postop. complications	1 (10.0%)	3 (12.0%)	2 (66.6%)	6 (15.8%)
	Renal dysfunction (1)	Persistent air leak and empyema (1)	Hypoxemia 2	
		SP recurrence and arrhythmia (1)		
		Gastric ulcer bleeding (1)		
Follow-up (months)[a]	23.9 (10.9–43.1)	29.3 (9.8–58.9)[c]	26.1 (21.9–29.3)	27.6 (9.8–58.9)[c]
Postdischarge recurrences	0	2 (8.0%)	1 (33.3%)	3 (7.9%)

VATS, video-assisted thoracic surgery; M-T VATS, mini-thoracotomy VATS; SP, spontaneous pneumothorax
[a] Data are presented as the mean (range)
[b] The population is 37 patients who underwent preoperative CT
[c] Excludes a case of in-hospital death in which the chest tube could not be removed postoperatively

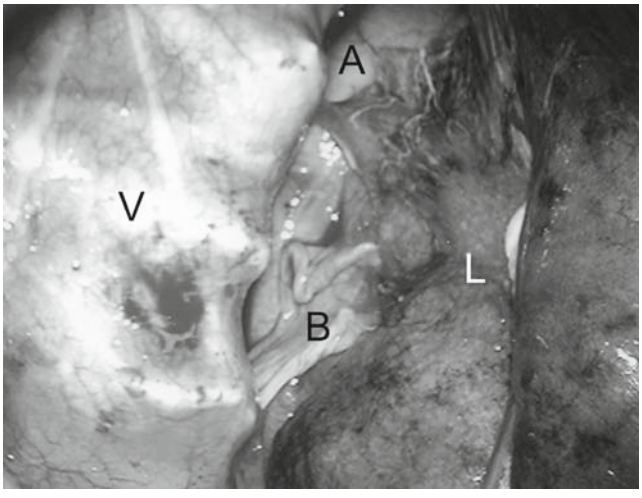

Fig. 2 Thoracoscopic view of a bulla at the azygoesophageal recess. *A*, descending aorta; *B*, bulla at the AER; *L*, lung; *V*, vertebral body

the AER were indeed ruptured (Fig. 2). They accounted for 26.3% of all 38 patients and for 66.7% of 15 patients whose bullae at the AER were identified by preoperative CT. The average size of the bullae at the AER based on CT measurements (breadth × length × height) was 38.5 ± 11.0 (22 – 55) × 19.1 ± 6.1 (10 – 28) × 85.7 ± 33.8 (40–150) mm. The long-axis of the AER bullae was predominantly vertically oriented, with the height significantly greater than the width ($P < 0.01$).

The reasons for converting from VATS to a mini-thoracotomy were pleural adhesion around the AER or poor visualization of the lesion due to a restricted working space. At surgery, all bullae at the AER were white and thin-walled. Surgical methods for managing the bullae at the AER were buttress stapling using a PGA sheet in eight, simple stapling in one, and hand suture using a PGA sheet pledget in one.

Two technical problems are peculiar to this setting. First, when we obtain a surgical view of the AER bullae by retracting the lung anteriorly, if the parenchyma underlying the bulla is under traction it is sometimes difficult to encircle the bulla's base with a stapler. Second, because of location, the working space to maneuver an endoscopic stapler, even if it is flexible, is somewhat limited.

Seven postoperative complications occurred in 6 of 38 patients (morbidity 15.8%). Among them, one patient had ipsilateral recurrence on the third postoperative day, and required reoperation. Another patient with intractable prolonged air leak and requiring long-term steroid use for interstitial pneumonia died from pneumonia and empyema on the 55th postoperative day (mortality 2.6%).

The postoperative follow-up period ranged from 9.8 to 58.9 months (median 27.6 months). Following discharge, three patients died from exacerbation of idiopathic pulmonary fibrosis, gingival malignancy, and malignant lymphoma, respectively. Postdischarge ipsilateral recurrences were observed in three patients and were treated with surgery in one, simple aspiration in one, and chemical pleurodesis in one. Among the 10 patients with surgically proven ruptured bullae at the AER, no postoperative recurrence was documented.

Discussion

In PSP patients, the role of surgery, VATS bullectomy, and apical pleurodesis has been firmly established.[6–9] In SSP emphysematous patients, however, the role of surgery still remains controversial; this population mostly consists of elderly individuals with variable respiratory reserve and co-morbidities.[1,2] If patients are selected appropriately, surgical intervention, especially VATS, is a highly effective treatment modality for both SSP and PSP.[4,10–12] The goal of treatment varies from permanent recurrence prevention (e.g., in younger PSP patients) to temporary discharge without a chest tube. The American College of Chest Physicians (ACCP) and British Thoracic Society (BTS) have issued several recommendations on this topic.[13,14] However, because of the wide heterogeneity of this population, indications for surgical intervention should be determined individually. Location of the ruptured lesions, pleural adhesions, and the degree of emphysematous changes are important factors affecting surgical procedures and potentially outcome. Few attempts have been made to determine surgical strategies for SSP on the basis of the location of the ruptured lesions.

The AER, a pleural protrusion into the right posterior mediastinum extending from the azygos arch to the diaphragm, was initially called "cul-de-sac pleural interazygo-oesophagien."[15] Markel stated that this pleural recess contains a ridge of lung tissue of the right lower lobe, the so-called crista pulmonis.[15] The normal configuration of the AER on CT is a smooth interface convex to the left. It is accentuated with deep inspiration, pulmonary emphysema, or thoracic kyphosis.[16,17] The deep AER is one of the important CT findings consistent with lung overinflation in patients with emphysema.[18] It is reasonable to suppose that bullae at the AER also form "the crista bullae" similar to lung parenchyma. The AER is considered a potential expandable reserve for pulmonary tissue or bullae in the right pleural space. It is supposed that bullae at the AER tend to expand paravertebrally and rupture. Incidentally, likely as a consequence of the presence of the descending aorta, a similar phenomenon has not been observed in the left thorax.

Technical problems in surgical treatment of the bullae at the AER included poor mobilization of the base of the bulla and a restricted working space. These factors sometimes hinder stapling the bulla within the bulla's base at an appropriate angle of introduction, which might cause a compromised stapling line on the bulla. To resolve these problems we used a buttress stapling technique and an endoscopic stapler introduced through a port at the tenth intercostal space. When the endoscopic stapler is introduced through this port, parallel to the vertebral body, stapling of the bulla at the AER is safer and accomplished with greater ease and completeness.

Preoperative CT is useful for locating bullae and pleural adhesions and to facilitate safe port placement. Although the bullae at the AER can be identified by CT, one must make sure not to mistake the expanded bulla at the AER for an intrapleural air collection, especially when there is no free air in the AER. It was remarkable that all ruptured bullae at the AER had a predominantly vertically extended shape on CT. Because the AER is the deepest region in the right pleural space, if the bulla at the AER were completely deflated it could be missed even by thoracoscopic inspection. Thus, preoperative awareness of this bulla, revealed by CT, is important.

Postoperative recurrence of SSP has been reported in up to 12.5% of cases.[1,5,10] However, a comparative evaluation of postoperative recurrence rates by various therapeutic options cannot be generalized because SSP patients comprise a heterogeneous population. Although we did not add routine preventative pleurodesis procedures to the parietal pleura, we observed no postoperative recurrence for patients with ruptured bullae at the AER. Our results indicated that right SSP patients with bullae at the AER might be one of the SSP subgroups who benefit from surgical intervention.

Conclusion

When treating right SSP with emphysema, the possibility of a ruptured bulla at the AER should be considered as a common lesion responsible for the occurrence of pneumothorax. Preoperative awareness of this peculiar location of the bullae and CT localization is important for surgical planning. Although the AER is a unique location, bullectomy under VATS is the method of choice for treating these lesions.

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