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Case Report

Bronchial atresia in an accessory lobe caused tension pneumothorax

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ABSTRACT

We report a case in which bronchial atresia in an accessory lobe caused tension pneumothorax. The atretic bronchus was found in an accessory lobe (right S6) that had been incompletely isolated from the right lower lobe during the fetal period. The accessory lobe was overinflated, leading to bullous formation due to the trapping of air. Video-assisted excision of the posterior lobe was successfully performed without any perioperative complications. The pathological findings were compatible with congenital bronchial atresia. This is the first report of bronchial atresia in an accessory lobe. We must keep this lesson in mind because such cases can suddenly develop serious complications.

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Background

Bronchial atresia is a rare developmental anomaly characterized by focal obliteration of a bronchus in association with hyperinflation of the distal lung [1-4]. Bronchial atresia can involve any lobe but most commonly occurs at the apicoposterior segment of the left upper lobe, followed by the right upper lobe [1, 2]. We report a case of bronchial atresia in an accessory lobe (right S6) that had been isolated from the right lower lobe during the fetal period.

Case Presentation

The patient was a 21-year-old man who had no remarkable medical or family history. He visited a private hospital due to sudden onset of right chest pain, cough, and dyspnea. A chest X-ray showed an almost completely collapsed right lung and mediastinal shifting to the contralateral side (Figure 1A). After chest tube drainage, the patient was referred to our hospital for further examination and surgery. Chest CT showed a hyperlucent lung area with huge bullae that were localized in the right S6 (Figure 1B). There was no B6. However, the peripheral bronchus appeared normal without mucinous impaction or bronchial dilatation. The defect of the B6, as observed by bronchoscopy (Figure

1C), was also suggestive of bronchial atresia. The S6 was almost completely isolated from the right lower lobe and was a so-called 'posterior lobe' [5]. Both the ventilation and perfusion scintigraphy showed a defect in the emphysematous area (Figure 1D). The patient was diagnosed with pneumothorax due to bronchial atresia in a posterior lobe.

Under general anesthesia, video-assisted excision of the right posterior lobe was performed with 4-cm window thoracotomy. Because the posterior lobe was incompletely separated from the basal segment, the posterior lobe had trapped air that was delivered from the basal segment via the incomplete accessory fissure (Figure 2A). There was no abnormality in the vascular branching of the posterior lobe. After dissecting the A6 and V6, we identified a band-like structure that seemed to be a regressed B6 (Figure 2B-D). The posterior lobe was resected with the regressed B6. The patient was discharged hospital without any complications on postoperative day 5 after the removal of the chest tube on postoperative day 2. A pathological examination revealed bullous changes in the resected posterior lobe. The regressed B6 tissue consisted of fibroblastic tissue and had no continuity with the bronchus or alveolus (Figure 2C D). At the time of writing this report, the patient is doing well without bullous reformation.

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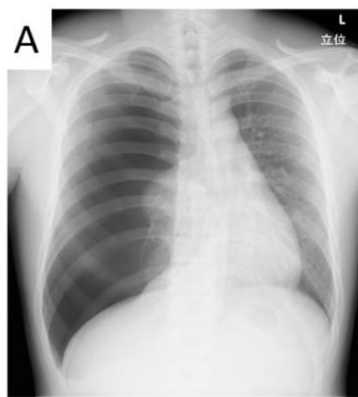


Figure 1A: Chest X-ray showing right tension pneumothorax.

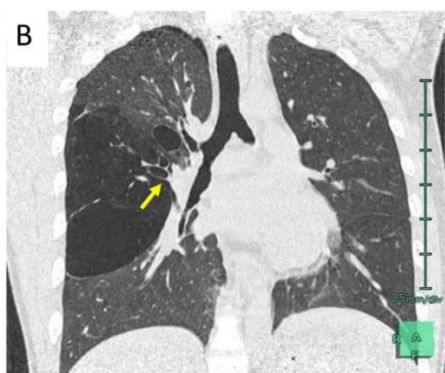


Figure 1B: Chest computed tomography showing emphysematous change in the right superior segment and atretic bronchus (arrow).

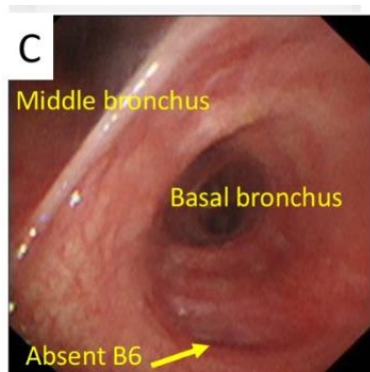


Figure 1C: Bronchoscopy showing the absence of the B6 orifice.

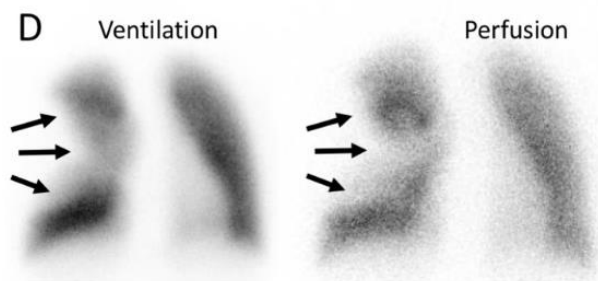


Figure 1D: Ventilation and perfusion scanning showing ventilation and perfusion defects in the emphysematous area (arrows).

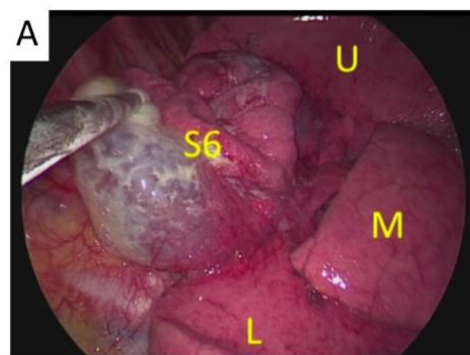


Figure 2A: A thoracoscopic view showing the isolated S6 with a prominent accessory fissure.

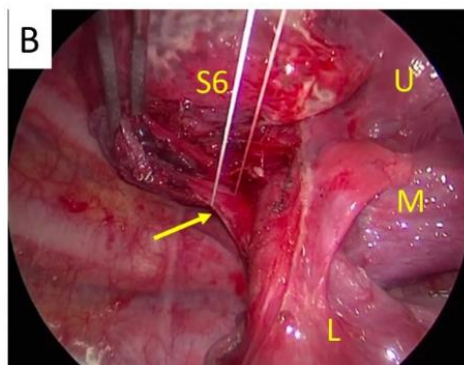


Figure 2B: U: upper lobe, M: middle lobe, L: lower lobe. An intraoperative view showing a band-like structure including vanished B6 (arrow).

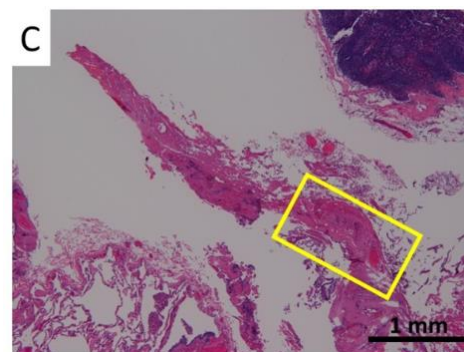


Figure 2C: A microscopic view of the vanished B6 tissue revealed fibroblastic tissues.

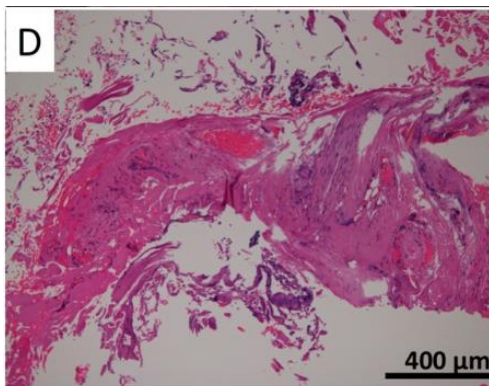


Figure 2D: A high power view of the square area in C showing fibroblastic tissue without continuity between the bronchus and alveolus.

Discussion

Congenital bronchial atresia is a rare disease that was first reported by Ramsay in 1953 [1]. Obstruction at the bronchial origin causes enlargement of the peripheral bronchus, mucous accumulation, and/or emphysematous changes in the peripheral lung. The obstructed lung field traps air thorough the pores of Kohn and bronchiolar-alveolar channels of Lambert. The collateral airway works as a check valve, resulting in hyperinflation and pneumothorax in some cases.

Jederlinic reviewed 86 cases of bronchial atresia. Bronchial atresia most commonly occurs in young patients (mean age, 17 years [range, 0–44 years]) [2]. Bronchial atresia tends to occur in males more frequently than in females (64 vs 36%). Fifty percent of bronchial atresia cases occurred in the left superior lobe and 49–51% of the patients were

asymptomatic at the time of evaluation [2, 3]. According to the reports of Kanzaki and Ishiwata in Japan, the etiology is almost the same as above [3, 4].

A PubMed search using the key words, ‘bronchial atresia AND pneumothorax’ revealed 7 case reports (Table 1). Chest tube drainage alone was performed in two cases, while surgical excision of the affected lung was performed in the remaining five cases (lobectomy, n=1; segmentectomy, n=4). Preservation of the lung parenchyma by segmentectomy is reasonable in cases of benign disease, particularly in the current case in which the patient had a prominent accessory fissure between the resected lung and the preserved lung. We believe that surgical therapy was also reasonable because the posterior lobe had neither perfusion nor ventilation, and because the patient had a considerable risk of death from tension pneumothorax.

Authors	Age/sex	Year	Location	Treatment	Bulla
Beckman et al.	20/M	1996	Left B6	Drainage	Unknown
Okamoto and Okubo	25/M	2002	Left superior	Drainage	Unknown
Yoon et al.	36/F	2004	Right B2	Upper lobectomy	Present
Kameyama et al	36/F	2006	Left superior	Superior segmentectomy	Present
Niimi and Gotoh	25/F	2010	Right B4	S4 segmentectomy	Present
Igai et al.	18/F	2012	Right B2	S2 segmentectomy	Present
Tanaka et al.	26/M	2013	Left superior	Superior segmentectomy	Present
Maeda (current case)	21/M	2016	Right B6	S6 segmentectomy	Present

The pathological mechanism of the bronchial obstruction remains unknown. There are several possible theories. Bronchial atresia was caused by a disturbance in the bronchial branching during the 5th week of the fetal period when the central airways were being formed [6]. However, a normal bronchial structure is generally observed in the peripheral field of occluded bronchi, as was observed in our case. Thus, other investigators have suggested that bronchial atresia is caused after the 16th week of the fetal period, at the completion of bronchial branching. The present case showed emphysematous change without mucoid impaction in the occluded S6 area. Alveolar glands mature during weeks 20–24 of the fetal period. Furthermore, development of collateral ventilation is accomplished by 8 years of age. If bronchial atresia is caused before the sufficient development of mucous production by the alveolar glands, mucous collection is not observed: the high pressure of the occluded bronchioles prevents mucous collection [7]. Thus, in the present case, bronchial atresia might have appeared at around week 20 of the fetal period.

Summary

A superior accessory fissure that divides the lower lobe into the basal segment and superior segment, is present in approximately 5% of individuals examined by CT [8, 9]. However, the incidence of such a prominent accessory fissure as was observed in the current case may be less than 1%. This report showed that bronchial atresia in the isolated accessory lobe may induce tension pneumothorax. This should be a grave warning for physicians involved in therapeutic decision-making in similar cases.

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