CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 37 (2017) 205–207

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Idiopathic spontaneous pulmonary torsion of the lingula: A case report

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ARTICLE INFO

Article history:
Received 27 March 2017
Received in revised form 16 June 2017
Accepted 17 June 2017
Available online 29 June 2017

Keywords:
Idiopathic
Spontaneous pulmonary torsion
Lingula
Case report

ABSTRACT

INTRODUCTION: Spontaneous pulmonary torsion is an extremely rare event and is known to occur as a complication of thoracic surgery and traumatic injuries.
PRESENTATION OF CASE: An 18-year-old man presented to our hospital with pain in the left back region. Clinical examination, computed tomography and bronchoscopy are crucial for diagnosis of pulmonary torsion. During thoracotomy, the lingula segment was observed to be bent on the head side and turned 180° counterclockwise; subsequently, lingulectomy was performed.
DISCUSSION: Spontaneous pulmonary torsion may occur in pulmonary conditions such as pneumothorax, atelectasis, infection, pleural effusion, congenital defect, or tumor. Furthermore, it can be speculated that torsion of the segment is possible only in the patients with an accessory fissure or those who have undergone a segmentectomy.
CONCLUSION: We have reported an extremely rare case with respect to the fact that the pulmonary torsion occurred spontaneously in an unseparated segment, and that the etiological factor could not be identified.

1. Introduction

Spontaneous pulmonary torsion is an extremely rare event and is known to occur as a complication of thoracic surgery and traumatic injuries. However, spontaneous cases without any associated histories are extremely rare. Furthermore, pulmonary torsion of a segment has been reported to occur only after segmentectomy [1,2]; this is because, unlike between lobes, there are no fissures between the various segments. We report a case of spontaneous pulmonary torsion of the lingula of unknown etiology.

The work in this case has been reported in line with the SCARE criteria [3].

2. Presentation of case

An 18-year-old man was referred to our hospital because of pain in his left back region that occurred suddenly at rest. His medical history and examination revealed that he had a funnel chest and scoliosis. The left back pain aggravated on motion and breath sounds of the lower left lung were slightly attenuated. The patient had mild fever and laboratory parameters showed presence of an increased inflammatory reaction. Chest radiography showed an infiltrative shadow in the lower left lung field. Chest contrast-enhanced computed tomography (CT) revealed a convoluted appearance (Fig. 1A, a) in the central side of the inferior segment of the lingula (S5) and consolidation in the peripheral side (Fig. 1). Bronchus, pulmonary artery, and the vein of S5 became constricted, bent, and rolled up and formed a convoluted shadow in the image (Fig. 1B and C). There was a border with linear structure thought to be the accessory fissure between the superior segment of the lingula (S4) and S5 (Fig. 1A, b). Bronchoscopy showed a stenosis of B5 and foamy secretion from the peripheral side. The patient had no history of surgery or trauma; thus, we made a tentative diagnosis of spontaneous pulmonary torsion of S5 with the accessory fissure, and performed emergency surgery for definitive diagnosis and treatment. During thoracoscopy, the lingula segment (S4 + S5) was observed to be bent on the head side and turned 180° counterclockwise (Fig. 2). An accessory fissure was not found; the linear structure mimicking an accessory fissure on CT was, in fact, the twisted lingula. As in the CT findings, S5 showed a blackish appearance and seemed to be necrotized; however, S4 showed no change in color even though it was twisted. First, we corrected the torsion of the lingula after ligating the lingular vein (V4 + 5) for prevention of thrombus isolation; then, lingulectomy was performed according to standard procedure by anterior thoracotomy.

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http://dx.doi.org/10.1016/j.ijscr.2017.06.043

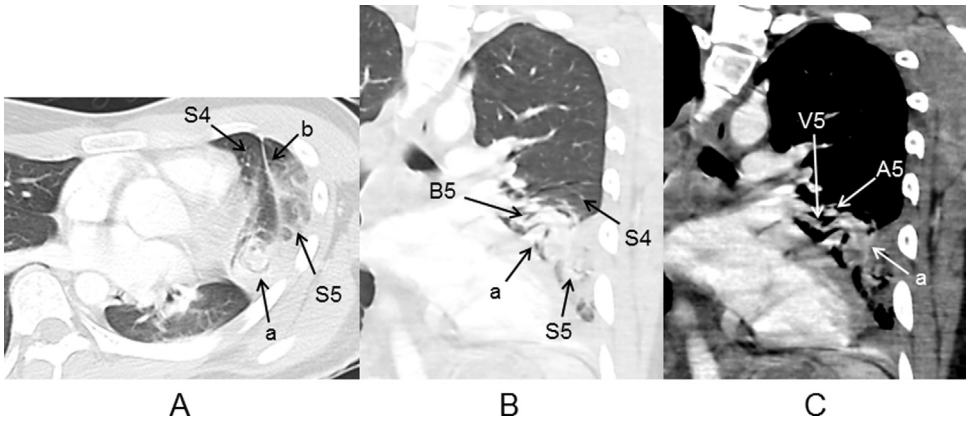

Fig. 1. Chest contrast-enhanced CT showing a convoluted appearance (a) on the central side of S5 and consolidation on the peripheral side. There was a border with a linear structure (b) thought to be an accessory fissure between S4 and S5 (A). Bronchus, pulmonary artery, and vein of S5 became constricted, bent and rolled up to form a convoluted shadow (B, C).

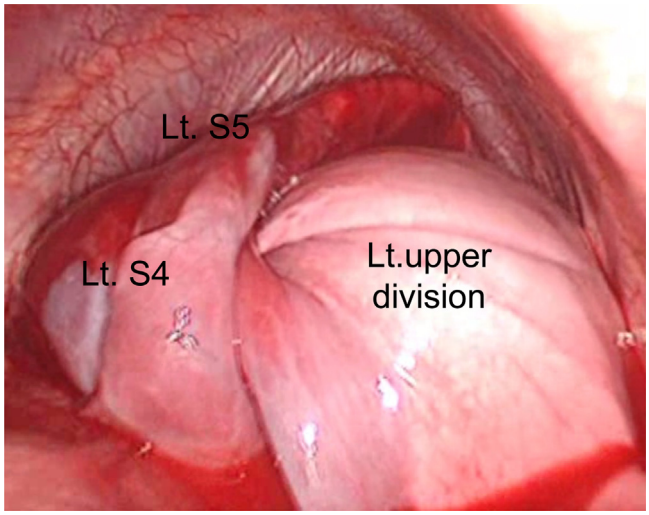

Fig. 2. Thoracoscopy showing the lingula segment (S4 + S5) bent on the head side and turned 180° counterclockwise. S5 showed a blackish appearance and seemed to be necrotized; however, S4 showed no changed in color even though it was twisted.

Pathological examination of the resected lung revealed extensive hemorrhage, congestion and vasodilatation and loss of air vesicle structure, which suggested reduced perfusion and necrosis due to the torsion. We removed the thoracic drain on postoperative day 2 and the patient recovered uneventfully. He was discharged on postoperative day 9.

3. Discussion

Pulmonary torsion refers to the rotation of a bronchovascular pedicle with resultant airway obstruction and vascular compromise. This disorder has been reported to occur after pulmonary resection, thoracic trauma, and pulmonary transplant. It may also occur spontaneously [4]. Among these, most cases occurred after pulmonary resection [5], but the torsion following pulmonary resection is a relatively rare complication (approximately 0.0086–0.3%) [6]. Furthermore, spontaneous pulmonary torsion is extremely rare; according to Ohde et al. [7], only 10 cases have been reported in scientific literature published in English language till date.

Spontaneous pulmonary torsion may occur in pulmonary conditions such as pneumothorax, atelectasis, infection, pleural effusion, congenital defect, or tumor [4]. In this patient, no causative episode was identified for spontaneous pulmonary torsion. In addition to these causes, we speculate that chest deformity may have been an additional cause because it increases the flexibility by decreasing the volume ratio of lung/pleural space. This patient had a funnel chest and scoliosis; however, the CT did not show obvious enlargement of the pleural space. Pulmonary torsion may involve the entire lung or individual lobe; therefore, it can be speculated that torsion of the segment is possible only in the patients with an accessory fissure or those who have undergone a segmentectomy. However, pulmonary torsion of a segment has been reported only after segmentectomy [1,2]. We could not find reports of cases caused by the presence of an accessory fissure. An extensive review of the literature suggests that this is the first case of spontaneous pulmonary torsion of lingula occurring in the absence of surgical history or any anatomical feature indicating an accessory fissure.

Pulmonary torsion is a rare condition but may cause serious complications once it occurs; the sudden blood flow deficiency mainly on the pulmonary vein occurs in hilar region and may result in acute respiratory failure, hemorrhagic infarction or gangrene of the torsional lung. The fatality rate has been reported to be 22% [6]. Therefore, early recognition and prompt intervention are crucial. Contrast-enhanced CT and bronchoscopy are useful for identifying pulmonary torsion. CT reveals an altered relationship between the trachea and the pulmonary arteries, and a change in the anatomical position of the lung [8]. Bronchoscopy has been reported to reveal flexure, twisting or obstruction [6]. In our case, we were able to suspect a possible pulmonary torsion by performing CT and bronchoscopy relatively early. When a pulmonary torsion is suspected, definitive diagnosis should be made immediately with the thoracoscopic examination. Once the diagnosis is confirmed, surgical detorsion or resection is indicated, depending on the viability of the injured pulmonary parenchyma [9].

4. Conclusion

We have reported an extremely rare case with respect to the fact that the pulmonary torsion occurred spontaneously in an unseparated segment, and that the etiological factor could not be identified.

Conflict of interest

All authors declare no relationship with other people and organizations of this work.

Funding

No funding has been received for the publication of this manuscript.

Ethical approval

Ethic approval not required.

Consent

Written informed consent was obtained from the patients for publication of this Case Report and any accompanying images.

Authors contribution

Masatoshi Kanayama: Corresponding author.
Toshihiro Osaki: The chief supervisor.
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Guarantors

Masatoshi Kanayama, Toshihiro Osaki.

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