

Spontaneous segmental pulmonary torsion. A case report

Torsión pulmonar segmentaria espontánea. Presentación de un caso

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Summary

Background: Lung torsions have been primarily reported as complications of thoracic surgeries or traumatic injuries. Spontaneous lung torsion is an exceptionally rare event, and cases without predisposing factors are even rarer. It has been suggested that it could be associated with the presence of other pulmonary conditions that disrupt the normal dynamics and configuration of the lung, such as infections, pneumothorax, atelectasis, tumors, among others. ***Case presentation:*** A case of spontaneous lung torsion of the lingula is presented in a 59-year-old woman with no pre-existing pathologies. The patient sought medical attention due to chest pain. The diagnostic suspicion was suggested through imaging, later confirmed through surgery with anatomopathological findings consistent with the diagnosis. ***Conclusions:*** Early diagnosis is crucial, as prompt surgical intervention significantly improves the patient's prognosis. This report emphasizes the importance of considering this pathology among diagnostic possibilities, even in the abscence of predisposing conditions.

Key words (MeSH)

Torsion mechanical
Lung diseases
Respiratory system
Multidetector computed tomography

Palabras clave (DeCS)

Torsión mecánica
Enfermedades pulmonares
Sistema respiratorio
Tomografía computarizada multidetector

Resumen

Antecedentes: Las torsiones pulmonares se han informado principalmente como complicaciones de cirugías torácicas o lesiones traumáticas. La torsión pulmonar espontánea es un evento excepcionalmente raro y los casos sin antecedentes que predispongan son aún menos comunes. Se ha sugerido que podría asociarse con otras afecciones pulmonares que perturben la dinámica y configuración normales del pulmón, como infecciones, neumotórax, atelectasias y tumores. ***Presentación del caso:*** Se presenta un caso de torsión pulmonar espontánea de la língula en una mujer de 59 años de edad sin patologías preexistentes. La paciente consultó por dolor torácico. La sospecha diagnóstica sugerida por las imágenes se corroboró en intervención quirúrgica, con hallazgos anatomopatológicos consistentes. ***Conclusiones:*** El diagnóstico precoz es fundamental, ya que la intervención quirúrgica rápida mejora significativamente el pronóstico del paciente. Este informe destaca la relevancia de considerar esta patología en las posibilidades diagnósticas, incluso en ausencia de condiciones predisponentes.

Introduction

Spontaneous pulmonary torsion is an extremely rare event and is known to occur as a complication of thoracic surgery and traumatic injuries. Spontaneous cases with no associated history are extremely rare. Furthermore, segmental pulmonary torsion has only been described after segmentectomy, probably because there are no fissures between the different segments, unlike what occurs between the lobes, so it can be speculated that torsion of a single segment is only possible in patients with an accessory fissure (1, 2).

Spontaneous pulmonary torsion could occur in association with other pulmonary conditions that disrupt the normal dynamics and configuration of the lung, such as atelectasis or pneumothorax, as well as infection or pleural effusion, and less common conditions such as congenital abnormalities or tumors. We report a case of spontaneous lung torsion of the lingula, with no apparent etiology at the time of presentation, in which imaging played a fundamental role in allowing timely surgical treatment and subsequent histological confirmation.

Case report

A 59-year-old woman with no chronic diseases and a history of two cases of flu in the last month presents with 48 hours of dorsal chest pain radiating to the front, associated with intermittent dry cough, without dyspnea. Physical examination revealed tachycardia of 118 bpm, saturation of 92% (ambient FiO2), no fever, and no abnormalities on pulmonary evaluation. Laboratory tests showed a positive respiratory panel for metapneumovirus and adenovirus, as well as elevated D-dimer. Due to suspicion of pulmonary thromboembolism (PTE), a chest CT angiogram was performed, which showed no

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signs of PTE. However, abnormalities were found in the lower lingular segment suggesting segmental torsion (Figures 1 and 2), which is why the patient underwent surgery.

The surgical specimen was sent for pathological examination. Macroscopic examination revealed an ovoid fragment of lung tissue measuring 4.5 × 4 × 2.3 cm, diffusely hemorrhagic and necrotic in appearance, attached via a pedicle to a lung wedge measuring 7 × 3.4 × 1 cm with no apparent lesions, both with apparently independent pleural covering (Figure 3). Microscopic examination confirmed the diagnosis of hemorrhagic infarction secondary to torsion in lung tissue with characteristics that, associated with the macroscopic findings, could correspond to extralobar sequestration (Figure 4). The patient had a favorable postoperative course, with normal inflammatory parameters and no abnormalities in follow-up checks.

Discussion

The evidence available in the literature on this pathology is scarce and limited to case series. In 1987, Felson reported that spontaneous pulmonary torsion could occur mainly in three scenarios: after traumatic events associated with other pathologies such as traumatic pneumothorax; after surgery, for example, following lung resection or lung transplantation; and also spontaneously, which is much rarer and usually associated with another pulmonary abnormality (3).

Spontaneous pulmonary torsion without known underlying abnormalities is extremely rare. According to a search of the English-language literature, eleven cases have been documented to date (1, 4).

Spontaneous pulmonary torsion may occur in pulmonary conditions such as pneumothorax, atelectasis, infection, pleural effusion, congenital defect, or tumor (3). In the case described here, the patient had no underlying pulmonary abnormalities or chronic diseases; however, she did have a history of two respiratory viral infections in the last month, and the viral panel was positive for metapneumovirus and adenovirus. Therefore, despite the absence of clinical or imaging signs of pneumonia and the lack of any other obvious factors that could have caused this pathology, the previous viral infections could have contributed to the occurrence of spontaneous pulmonary torsion through some unknown mechanism. In addition, the anatomopathological findings from the surgical sample revealed a pleural lining independent of the infarcted segment, suggesting the possibility of extralobar pulmonary sequestration. Although it cannot be confirmed, the presence of accessory fissures should be considered as part of the differential diagnosis. However, it is plausible that this finding is the predisposing factor.

Diagnosis is challenging, as clinical and laboratory findings are nonspecific, so imaging plays a key role. Contrast-enhanced CT and bronchoscopy are the most useful tests for identifying pulmonary torsion. CT can show an abnormal bronchial or pulmonary artery pathway, which will be more or less evident depending on the extent of the affected segment, lobe, or lung. As for bronchoscopy, it has been reported that it is also possible to see flexion, obstruction, or torsion (1, 5). In this case, pulmonary torsion was suspected after performing a chest CT angiogram, which showed flexion and an abnormal distal course of the segmental bronchus. The curious thing about this case is that the torsion occurred only in the lower lingular segment and, according to the literature, torsions affect the entire lung or an individual lobe; therefore, it is speculated that segmental torsion would only be possible in patients with an accessory fissure or in those who have undergone a segmentectomy (5, 6). In the case described, the previous presence of an accessory fissure cannot be determined because the patient had not undergone any previous studies.

Regarding treatment, it should be noted that pulmonary torsion involves rotation of the bronchovascular structure, resulting in airway obstruction and impaired blood flow. When this occurs, the affected segment often suffers an infarction and requires resection, as was the case in the present case. Sudden blood flow deficiency can cause acute respiratory failure, hemorrhagic infarction, and subsequently gangrene of the twisted lung; therefore, delay in treatment would also cause the formation of an arterial or venous thrombus, which could result in pulmonary or cerebral embolism (5).

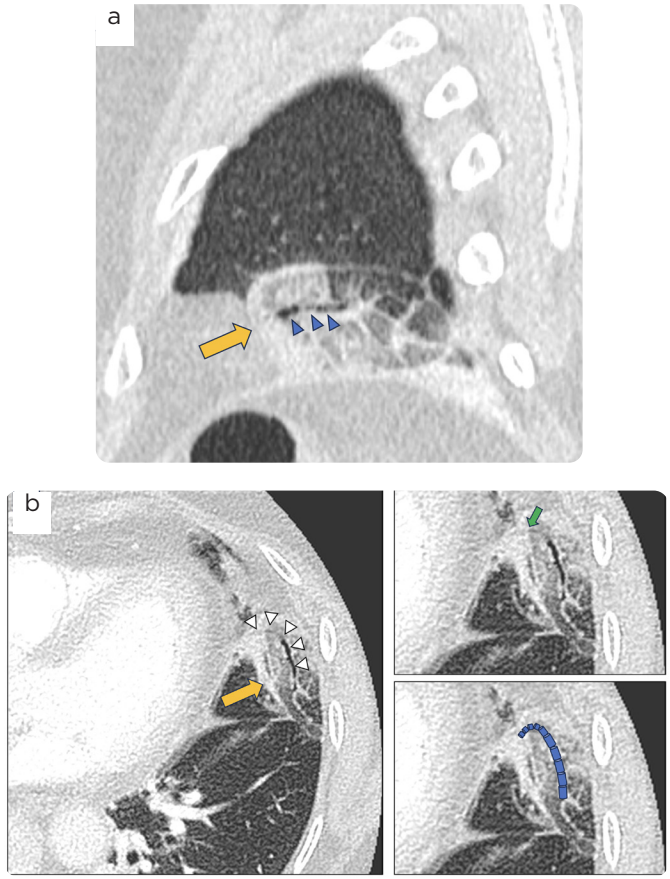


Figure 1. Chest CT angiography in lung window. a) MPR reconstruction in oblique sagittal plane: lower lingular segment with ground-glass opacities (yellow arrow) with a dilated bronchus inside (blue arrowheads); b) Axial section: "ground-glass" area (yellow arrow) and, within it, an abnormal lateral and posterior bend in the lower lingular segmental bronchus (arrowheads) at an angle of approximately 90°. A close-up of the same image is also shown, which better highlights the occlusion at the point of flexion (green arrow) and with a graphic representation of the altered bronchus (in blue).

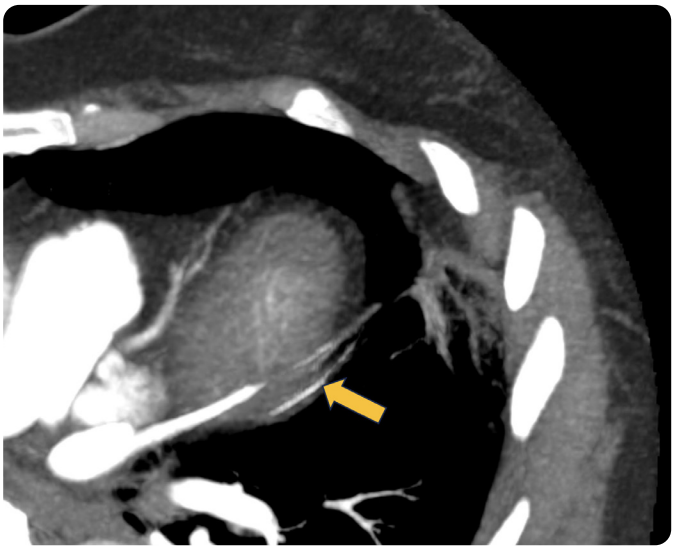
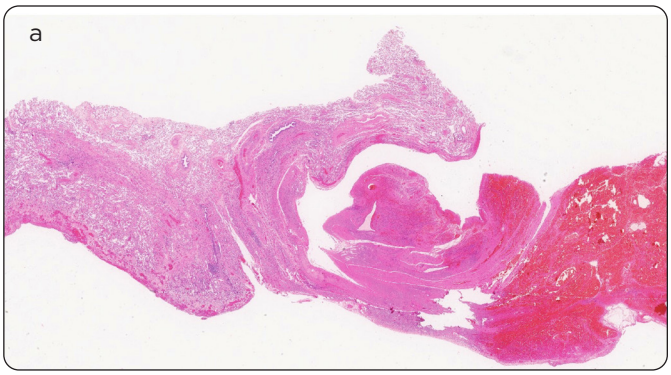


Figure 2. MIP reconstruction in soft tissue window of the same tomography: segmental artery with loss of distal representation prior to the point of torsion (yellow arrow).



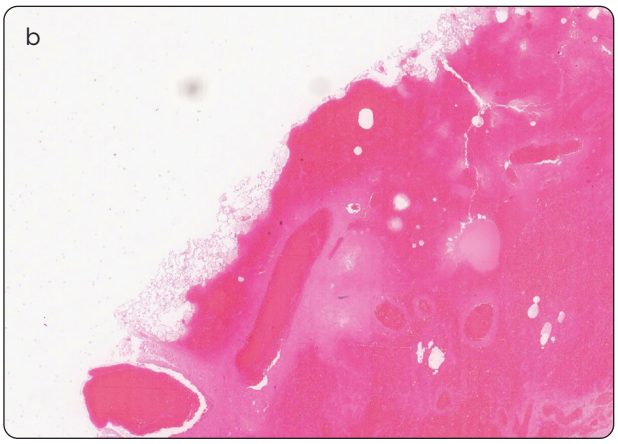


Figure 4. Microscopic examination. a) Low magnification view of histological section at the junction of lung tissue with preserved architecture and part of the adjacent tissue with hemorrhage and diffuse necrosis; b) At higher magnification, it can be seen that the area of the sample with necrosis and hemorrhage corresponds to lung tissue with few preserved alveoli towards the periphery and, in the central area, vessels of varying sizes, hyperemic, without recognizable bronchovascular or alveolar structures, consistent with hemorrhagic infarction secondary to vascular pedicle torsion.

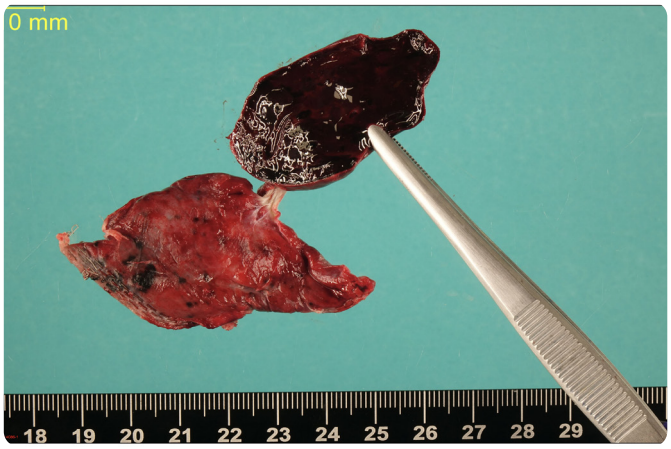


Figure 3. Macroscopic examination: segmental resection with macroscopically preserved lung fragment attached by pedicle to ovoid fragment with diffusely hemorrhagic and necrotic cut surface, both covered by pleura.

Conclusions

Pulmonary torsion is a rare condition, but it can cause serious complications once it occurs; therefore, early diagnosis is crucial because rapid surgical intervention provides the affected person with a good prognosis, as was the case with the patient documented here.

This case is particularly rare because the torsion occurred spontaneously in one segment and the etiological factor could not be identified. However, these reports are relevant because they raise awareness about this pathology and allow those who are familiar with it to keep it in mind within their range of differential diagnoses.

References

1. Kanayama M, Osaki T, Nishizawa N, Nakagawa M, So T, Kodate M. Idiopathic spontaneous pulmonary torsion of the lingula: A case report. Int J Surg Case Rep. 2017;37:205-7. https://doi.org/10.1016/j.ijscr.2017.06.043.
2. Wong PS, Goldstraw P. Pulmonary torsion: a questionnaire survey and a survey of the literature. Ann Thorac Surg. 1992;54(2):286-8. https://doi.org/10.1016/0003-4975(92)91386-n.
3. Felson B. Lung torsion: radiographic findings in nine cases. Radiology. 1987;162(3):631-8. https://doi.org/10.1148/radiology.162.3.3809475.
4. Ohde Y, Nakagawa K, Okumura T, Kondo H. Spontaneous pulmonary torsion secondary to pseudo-Meigs' syndrome. Interact Cardiovasc Thorac Surg. 2005;4(1):59-60. https://doi.org/10.1510/icvts.2004.096594.
5. Jalota Sahota R, Anjum F. Lung Torsion. En: StatPearls (Internet). Treasure Island (FL): StatPearls Publishing; 2023. PMID: 32809737.
6. Kanza RE, Heniche Y, Hubert J, Bérubé M. Segmental lung torsion. Radiol Cardiothorac Imaging. 2023;5(3):e220258. https://doi.org/10.1148/ryct.220258.

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