Case report: Pulmonary sequestration with gastric fistula in a 20-year-old woman

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ABSTRACT

Bronchopulmonary sequestration (BPS) is a rare condition characterized by the presence of lung tissue without function and without connection with the airways and may present in two forms: intralobar or extralobar.1 More rarely, BPS may be associated with a gastrointestinal fistula. We report the case of a 20-year-old patient with a history of recurrent pneumonia and dependent ventilatory chest pain who, after contrast-enhanced chest computed tomography and upper digestive endoscopy, was diagnosed with pulmonary sequestration of the medial basal segment and presence of gastric fistula in the cardia region. The treatment was successfully performed through a left lobectomy.

Keywords: Bronchopulmonary sequestration, Gastric fistula, Congenital malformation.

1 INTRODUCTION

Bronchopulmonary sequestration (BPS) is defined as a congenital malformation of the lower respiratory tract in which there is the presence of non-functioning lung tissue, with no connection to the airways and systemic arterial blood supply.[1] The incidence is rare, corresponding to 0.15 to 6.4% of lower respiratory malformations and is responsible for about 1.1 to 1.8% of lung resections.[2-3]

This condition can present itself in two forms that are differentiated by the origin of the visceral pleura adjacent to the malformation. The intralobar form presents visceral pleura of the adjacent lung, while extralobar sequestration is contained in its own visceral pleura, separate from the involved lung.[4] In rare cases, there may be a connection of this anomalous lung tissue to the gastrointestinal tract, being called bronchopulmonary malformation of the primitive intestine, which is related to the abnormal differentiation of the respiratory and upper gastrointestinal tracts during early embryonic development.[5]

2 REPORT

A 20-year-old woman was referred to the Hospital de Clínicas de Curitiba complaining of low back pain on the left beginning 6 days ago, with a ventilatory-dependent pattern. He also had a productive cough and fever of 37.9 ºC. On pulmonary auscultation, reduced vesicular murmur on the left, without adventitious noises. He had no gastrointestinal complaints.

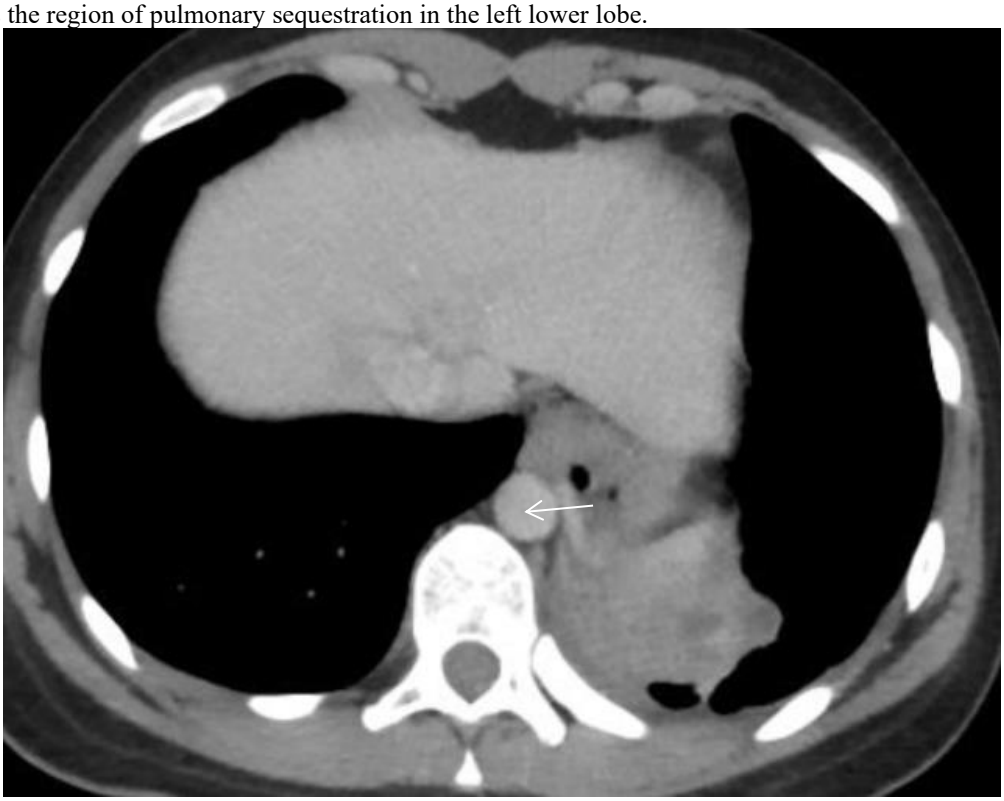
The patient reported a history of recurrent lower airway infections in one of these episodes, hospitalized with a diagnosis of lung abscess treated with antibiotic therapy for a prolonged time.

During their care in our service, laboratory tests that suggested an infectious condition were requested. A contrast-enhanced computed tomography scan of the chest showed a lobulated image in the lower lobe of the left lung, containing a small arterial branch from the celiac trunk, dilated bronchi and cavities in the left pulmonary base. In its internal margin, a linear path was observed that suggested a fistula to the distal esophagus and, because of this finding, an upper digestive endoscopy was performed, which confirmed the presence of a fistula in the cardia region.

After the examinations and discussion of the case, the diagnostic conclusion of pulmonary sequestration of the medial basal segment with arterial supply of the celiac trunk and presence of gastric fistula in the cardia region was reached.

Left inferior lobectomy was performed with cautious dissection of the esophageal fistula path, oversuture of its origin and local reinforcement with mediastinal pleura tissue. Uneventful procedure.

Figure 1. Computed tomography with intravenous contrast injection showing a small arterial branch from the celiac trunk that nourishes the region of pulmonary sequestration in the left lower lobe.



3 DISCUSSION

The presence of pulmonary sequestration associated with communication with the gastrointestinal tract (usually esophagus or stomach) is very rare and poorly described in the literature. Its embryological formation comes from a defect in the formation of the primitive intestine.[5]

The diagnosis is usually made during childhood, but in rare cases it can be diagnosed in adulthood. The most frequent symptoms are cough, hemoptysis, and pleural effusion.[6] In addition, the presence of BPS is related to a history of recurrent pneumonia, which may be the only form of presentation that will lead to the diagnosis.[7]

The SBP is divided into intralobar and extralobar form and has a systemic rather than pulmonary arterial supply.[8] A small retrospective study, which evaluated 8 cases of BPS, showed a predominance of this condition in the left lower lobe and in the intralobar form.[9] In another retrospective study, in which patients with BPS were evaluated, 15 adults and 13 children, the arterial circulation of the malformation came mainly from the thoracic aorta, originating from the abdominal aorta only in 35% of the patients analyzed.[2]

Imaging tests are essential for diagnosis. Among them, computed tomography of the chest stands out, which demonstrates the presence of a well-defined mass with possible cystic changes and surrounding emphysematous changes in the lung parenchyma10. The presence of aberrant vessels at pulmonary sequestration can be seen on tomography, but it is suggested that more specific tests, such as arteriography (gold standard), be performed to detail the characteristics of the arterial branch that supplies the lesion, facilitating the subsequent surgical approach.[11th]

Treatment depends on the patient's condition and his symptomatology. Resection of pulmonary sequestration, either by lobectomy or segmentectomy, is indicated for the prevention of infections and/or for symptom control, and its surgical treatment is controversial in asymptomatic patients.[2] This procedure has minimal morbidity, and most of it has a good prognosis.[12th]

4 CONCLUSION

Despite being a rare condition and little described in the literature, its early diagnosis is essential for the prevention of recurrent respiratory infections and improvement of symptoms, when present. Surgical treatment brings good results.

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