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Case Report: Pulmonary Sequestration in an Adult Not A Metastasis

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Abstract | **Case Report**

Introduction: Pulmonary sequestrum is a rare congenital lung anomaly defined as an area of dysplasia and nonfunctional lung tissue with abnormal systemic blood supply and various forms of venous drainage. We report the case of a 43-year-old man who was followed up for prostate adenocarcinoma with bone and lung metastases. The diagnosis of bronchopulmonary sequestration was made. ***Case report:*** The patient was 62 years old and was being followed for metastatic prostate adenocarcinoma to the bone. The frontal chest radiograph showed a poorly limited right basal opacity. Bronchial endoscopy noted an inflammatory appearance of the right lower lobar bronchus. A thoraco-abdomino-pelvic CT scan was requested as part of the extension work-up to identify a focus of intra-parenchymal condensation on the right posterior basal side vascularized by an abnormal systemic vessel emanating from the descending thoracic aorta in favor of an intra-lobar sequestration. ***Discussion and Conclusion:*** Pulmonary sequestration is a rare malformation whose diagnosis must be evoked in front of repeated infections in the same pulmonary territory, in particular the posterobasal territory. Multislice CT or MRI angiography are the examinations of choice for diagnosis and assessment. Surgical treatment remains the most important option.

Keywords: Pulmonary sequestrum, intra lobar sequestrations, extra lobar sequestrations, CT and MRI angiography, repeated infections, Surgical treatment.

INTRODUCTION

Pulmonary sequestrum is a rare congenital lung anomaly defined as an area of dysplasia and nonfunctional lung tissue with abnormal systemic blood supply and various forms of venous drainage [1]. The incidence of congenital airway anomalies ranges from 1 in 8300 to 35,000, and pulmonary sequestration accounts for 0.15% to 6.4% of congenital anomalies [2]. Pulmonary sequestration is primarily considered a childhood disease, as most cases are diagnosed early in life. Sometimes the diagnosis is not made until childhood, when affected individuals may live with mild or no symptoms. Asymptomatic pulmonary sequestrations are usually detected incidentally on radiological imaging. Pulmonary sequestration most commonly manifests as recurrent pneumonia, but also as chest pain, cough, dyspnea, and hemoptysis [3]. We report the case of a 43-year-old man who was followed up for prostate adenocarcinoma with bone and lung metastases. The diagnosis of bronchopulmonary sequestration was made.

CASE REPORT

The patient was 62 years old and was being followed for metastatic prostate adenocarcinoma to the bone. The frontal chest radiograph (Figure 1) showed a poorly limited right basal opacity. Bronchial endoscopy noted an inflammatory appearance of the right lower lobar bronchus. A thoraco-abdomino-pelvic CT scan was requested as part of the extension work-up to identify a focus of intra-parenchymal condensation on the right posterior basal side vascularized by an abnormal systemic vessel emanating from the descending thoracic aorta in favor of an intra-lobar sequestration (Figure 2).

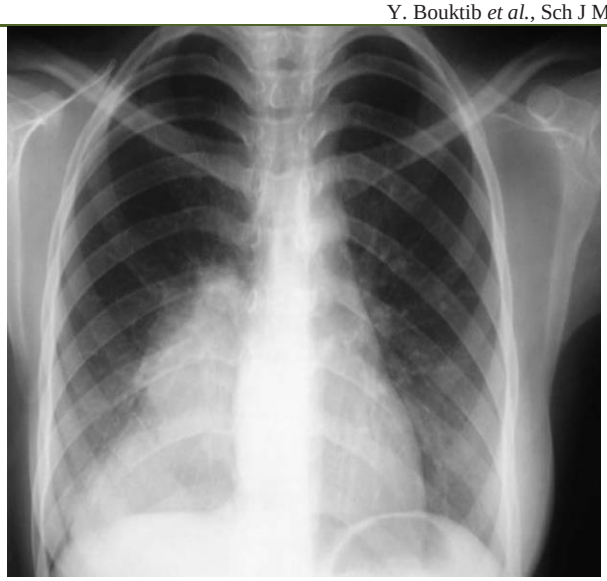

Figure 1: Frontal chest radiograph showing a limited right basal opacity

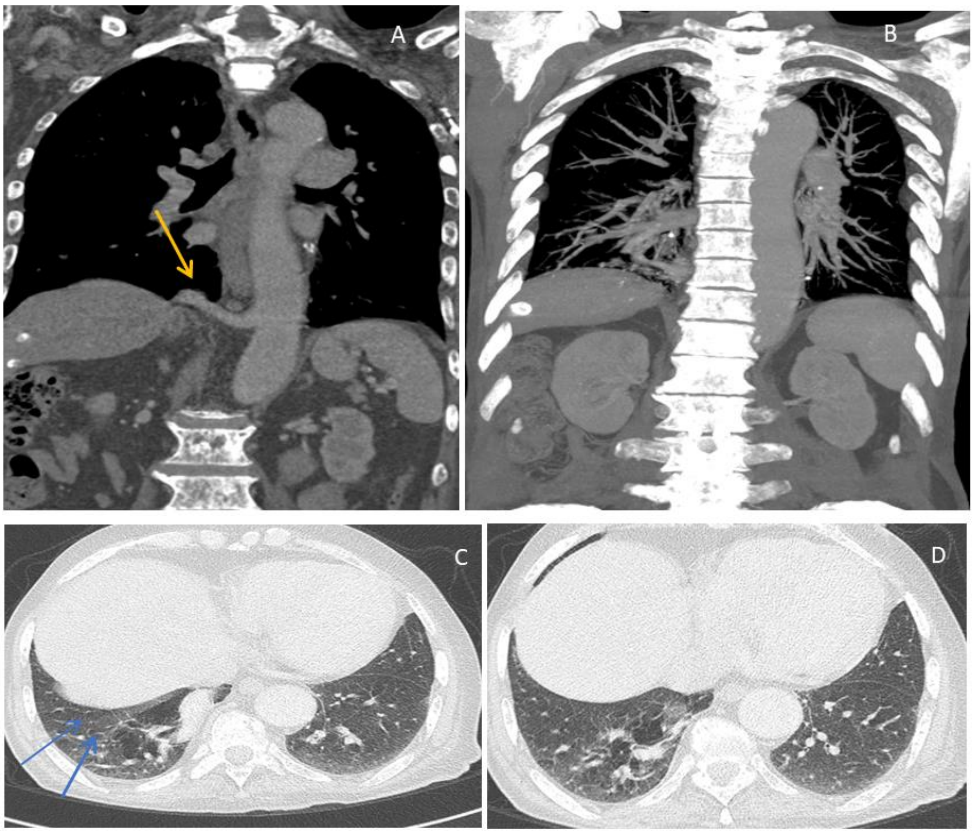

Figure 2: Thoracic CT with injection; A and B: Systemic arterial vascularization by an artery from the aorta with pulmonary venous drainage; C and D : postero-basal consolidation with emphysema bubbles

DISCUSSION

Lung sequestration is a congenital, non-hereditary malformation secondary to an abnormality occurring during lung organogenesis (day 22-24). It represents 0.15 to 6.4% of all congenital malformations of the lung and can be seen at any age with a slight predominance during the first two decades.

First described by Rokitanski and Rektorzic in 1861, it was Pryce [1] who, in 1946, gave a precise definition and distinguished two types: intra- and extralobar.

In intralobar sequestrations (ILD) the abnormal parenchyma is included in the normal lung: it is enveloped by the same visceral pleura; the usual location is the posterobasal region of the lower lobe without side predominance.

The extralobar sequestrations (ELS) do not have a connection with the normal lung because they develop from a supernumerary bronchial bud. There is therefore a complete anatomical and physiological separation between the two. SELs have their own pleural envelope. In 75% of cases they are located between the diaphragm and the lower lobe (80% on the left). Other locations in the middle and upper lobe are rare [2, 3].

The sequestered areas constitute a non-functional structure excluded from its bronchoarterial connections. The systemic origin of the vascularization is the main abnormality [1, 4, 5]. The vascularization of the sequestered territory most often comes from the thoracic aorta (SIL: 75%, SEL: 46%) or from the abdominal aorta (SIL: 19%, SEL: 32%). But the origin of the systemic artery can be a branch of the aorta (diaphragmatic artery, intercostal, celiac trunk, renal arteries, subclavian artery).

According to Pryce, three types are distinguished:

- Type I: The only abnormality is the vascularization of an area of normal pulmonary parenchyma by an artery of systemic origin ;
- Type II: The sequestered lung parenchyma is formed from a normal bronchial bud. The abnormal artery supplies an area of sequestered lung and part of the adjacent healthy lung;
- Type III: The sequestered lung bud is formed from a normal bronchial bud;
- Type IV: The bronchial bud is normal but the supernumerary artery vascularizes only sequestered lung.

Venous return varies according to the type of sequestration. In extralobar forms venous return is dependent on Venous return varies according to the type of sequestration. In extralobar forms, venous return is dependent on the azygos system, the inferior vena cava or exceptionally the portal vein. In intralobar forms, venous return is often provided by a pulmonary vein in a normal position, sometimes by a venous trunk of the vena cava system with a supra- or transdiaphragmatic course.

LIS may be manifested by recurrent episodes of pneumopathy with fever and sometimes hemoptysis and chest pain. Sometimes it is discovered during a complication: hemothorax or heart failure due to shunt effect.

The macroscopic aspect of the sequestrated parenchyma is often atelectatic or dystrophic. Histological examination shows air cavities with a bronchial structure with a collagenous wall lined with a cylindrical epithelium.

A frontal chest X-ray may suggest the diagnosis in the presence of a posterobasal opacity, particularly on the left. CT with injection (multislice angiotomodensitometry) is an excellent examination for the diagnosis and preoperative assessment of pulmonary sequestration [6, 7]. It recognizes the nature of the mass and shows the systemic artery [2, 6-8]. However, magnetic resonance imaging (MRI), and in particular MRI angiography, is probably the ideal method for diagnosing pulmonary sequestration [9, 10]. Aortography is no longer used for diagnostic purposes and its use is now reserved for embolization techniques [2, 7].

Treatment of pulmonary sequestration is necessary because of the risk of recurrent respiratory infection and spontaneous hemothorax [3, 11]. It is essentially surgical. The control of the systemic feeding artery is delicate due to the frailty of its embryonic wall and its elasticity with the risk of retraction into the mediastinum or through the diaphragm. The surgical procedure often consists of a lobectomy, rarely a segmentectomy. A simple ligation of the afferent artery may be performed whenever the pulmonary parenchyma is considered normal and there are no dystrophic lesions: Campbell *et al*., [12] and Ernst *et al*., [13] have reported cases of this. Our observation is another example. Yamanaka *et al*, [4] report a case in which the aberrant artery was transected at its origin and anas- tomosed laterally with the inferior border of the left pulmo- nary artery without pulmonary parenchymal resection.

Recently, surgical treatment by videothoracoscopy has been performed [14]. It seems technically more feasible in extralobar sequestrations and in children.

Currently, endovascular techniques are proposed for the treatment of pulmonary sequestrations, mainly in patients with an altered general condition or in patients with a lack of oxygen.

CONCLUSION

Pulmonary sequestration is a rare malformation whose diagnosis must be evoked in front of repeated infections in the same pulmonary territory, in particular the posterobasal territory (especially on the left). Multislice CT or MRI angiography are the examinations of choice for diagnosis and assessment. Surgical treatment remains the most important option.

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