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CASE STUDY

Broncholithiasis and lithoptysis associated with silicosis

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Broncholithiasis and lithoptysis associated with silicosis. V.C.S. Antão, G.A. Pinheiro, J.M. Jansen. ©ERS Journals Ltd 2002.

ABSTRACT: A case of broncholithiasis associated with massive silicosis is reported, showing a rare aspect of parenchymal lesions generating broncholiths as well as the presence of recurrent lithoptysis, with subsequent regression of radiological lesions.

Aetiological, clinical, physiopathological, and radiological aspects of the disease are discussed, demonstrating the importance of the use of computed tomography in diagnosis. The mineralogical analysis of expectorated fragments is also shown.

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Broncholithiasis is characterised by bronchial erosion or distortion due to hilar or parenchymatous calcifications and is usually related to infectious aetiologies, such as tuberculosis or histoplasmosis [1]. Its association with silicosis is quite rare, and in the few reports described in medical literature, broncholithiasis has occurred only as a result of disorders of the lymph nodes [2–5].

The present authors report a unique case of a patient with silicosis, who presented parenchymatous calcified lesions that eroded into the bronchial lumen leading to lithoptysis, with elimination of >100 broncholiths and impressive reduction of radiological lesions.

Case report

A 43-yr-old male presented with productive cough with bloody sputum for 5 months, wheezing and dyspnoea on exertion. The patient also reported the expectoration of small calculi of greyish colour and stony consistency, which were ~2 mm in diameter and resembled "grape seeds" (fig. 1).

The patient had a 28-pack-yr history of cigarette use. Twenty-six yrs before presentation, the patient had worked as a stone crusher in the stone working industry for a 1-yr period, and was exposed to a large amount of granite and quartz dust, but did not use any respiratory protective equipment. The patient had also occasionally crushed stones from a nearby asbestos mine. During his subsequent jobs as a painter and doorman, there was no exposure to silica dust.

Physical examination disclosed diminished breath sounds and sparse ronchi, as well as finger clubbing. The spirometry demonstrated severe airflow obstruction with a forced expiratory volume in one second (FEV_1) of 1.17 L (41% of predicted) and FEV_1/forced vital capacity of 50% pred. The first chest radiograph revealed bilateral calcified apical coalescence areas, compatible with massive silicosis, lung hyperinflation, hilar eggshell calcifications of the lymph nodes and bilateral diaphragmatic, calcified, pleural plaques (fig. 2a). The patient underwent a fibreoptic bronchoscopy and the endoscopical appearance was normal. Sputum and bronchoalveolar lavage analyses did not reveal

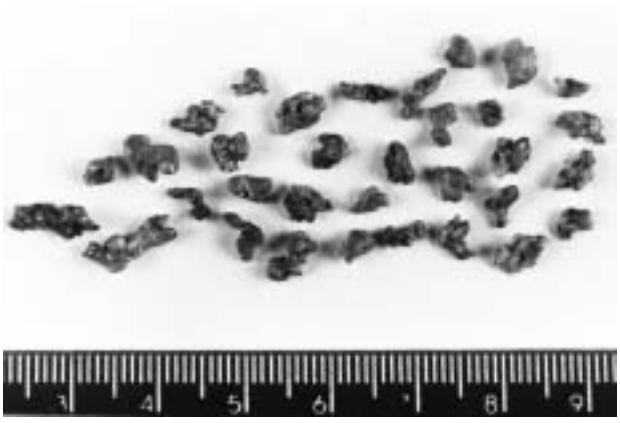

Fig. 1. – Broncholiths expectorated by the patient, ~2 mm in diameter.

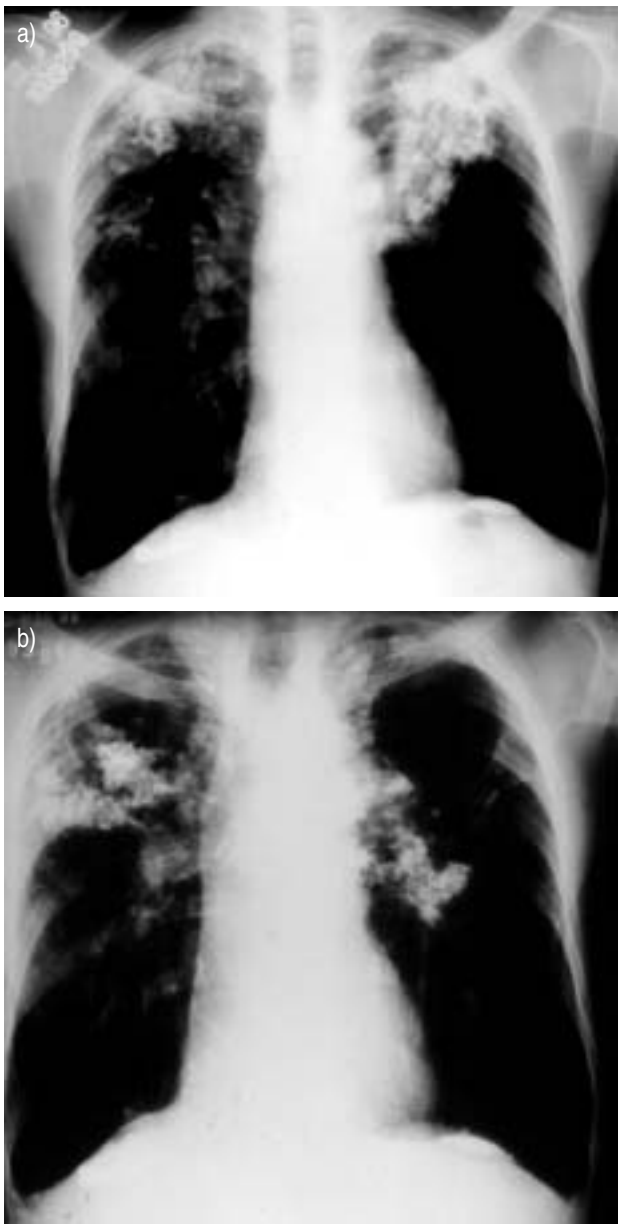

Fig. 2. – a) Postero-anterior chest radiograph showing calcified coalescence areas in upper lobes, lung hyperinflation, bilateral eggshell lymph nodes, and calcified diaphragmatic pleural plaques. b) Chest radiograph, performed 3 yrs later, showing gradual reduction of calcified opacities.

Mycobacterium tuberculosis, fungi or malignant tumour cells. The result of a tuberculin test was negative. There was no evidence of autoimmune disease or other disorders.

During the 3-yr period of follow-up, the patient presented with several episodes of lithoptysis. Chest radiograph showed gradual reduction of the calcified lesions, which had also moved towards the middle zones of the lung (fig. 2b). A helical computed tomography (CT) was performed, demonstrating the close relationship between the broncholiths and the bronchi bilaterally (fig. 3), as well as the presence of apical bullae.

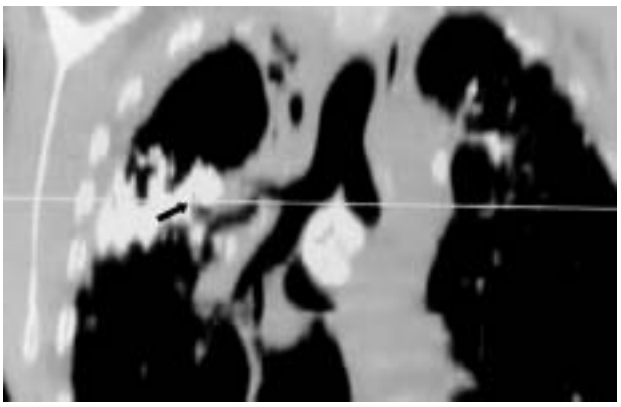

Fig. 3. – Helical computed tomography (coronal reconstruction), revealing the broncholiths that originated from the parenchyma in close relationship with the bronchi (arrow) and the calcified lymph nodes.

One sample of the expectorated material was analysed by electron microscopy and energy-dispersive spectrometry (EDS), with a NORAN-VOYAGER system (Altran Corporation, Boston, MA, USA) attached to a JEOL JXA 840A microscope (JEOL, Ltd, Tokyo, Japan), operating at 20 kV. The sample had previously been metallised with carbon in a JEOL JEE 4X (JEOL, Ltd). Nine EDS, semiquantitative chemical analyses were performed. The mean values for atomic weight, obtained through the microanalysis, were: 78.92 calcium and 21.09 phosphorus (fig. 4).

Discussion

In the USA, histoplasmosis appears as the primary cause of broncholithiasis, whereas in Europe, most broncholithiasis cases are attributable to tuberculosis [6]. Other infectious aetiologies are: cryptococcosis, aspergillosis, coccidioidomycosis, and actinomycosis [3]. Nocardiosis was also mentioned by WEED and

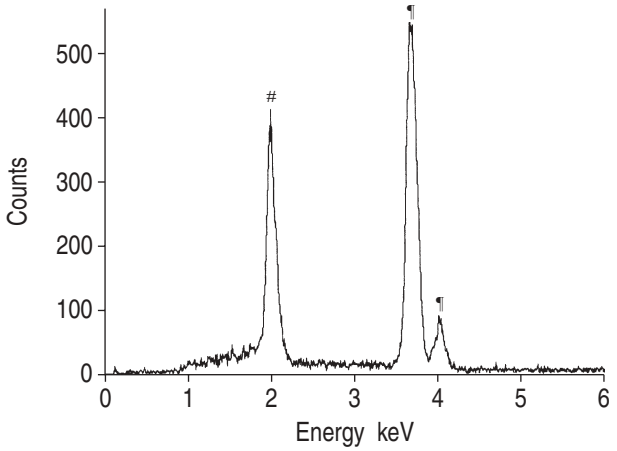

Fig. 4. – Graph showing the energy-dispersive spectrometry, semiquantitative chemical analysis of the expectorated material, characterising the presence of calcium (¶) and phosphorus (#) as constitutive elements of broncholiths. keV: kilo-electron volt.

ANDERSEN [7] in their study of nine cases of broncholithiasis.

SARTORELLI [8] was the first to describe broncholithiasis in patients with silicosis in 1957. Eggshell calcification of the lymph nodes is a common finding in patients with this pneumoconiosis, but these calcified lymph nodes rarely erode into the bronchial lumen. This physiopathological mechanism was reported in cases where the association of silicosis and broncholithiasis was present, leading to bronchial obstruction [4], broncho-oesophageal fistula [5] and haemoptysis [3].

An interesting aspect of the present case is the role played by the parenchymal lesions in the genesis of broncholithiasis. The authors hypothesise that the silicotic large opacities became calcified, and were then subsequently disaggregated in multiple fragments that were expectorated by the patient. These broncholiths may have obstructed small bronchi, leading to air trapping and the formation of bullae, through valvular mechanisms. The remaining lesions were then pushed towards the middle zones of the lungs by the bullae.

Mineralogical analysis revealed that the expelled substance was composed of calcium phosphate. In fact, the alkalisation of the pulmonary environment, with precipitation of calcium and phosphate as well as the erosion of calcified granulomas by respiratory movements, appears to be one of the most important mechanisms related to the formation of broncholiths [1, 6].

The clinical presentation of broncholithiasis depends primarily on its location and its obstruction level. Some patients may be asymptomatic. The main symptom is a dry cough with eventual expectoration. Haemoptysis may vary from mild to severe. Fever and chills are often related to infectious complications. Wheezing may occasionally occur due to mechanical obstruction of the airways. According to the 15-case series studied by CONCES *et al.* [9], lithoptysis is not frequent, but the present authors' patient presented recurrent lithoptysis with expectoration of multiple broncholiths.

The most important radiological manifestations are signs of bronchial obstruction and changing position or disappearance of a calcified focus on repeat radiographs [10], this being a significant aspect in the present case. CT allows a better visualisation of the lesions and it is especially useful for establishing the relationship between the calcification and the bronchi [9]. Since bronchoscopy was normal, the CT scans provided essential information for the diagnosis of broncholithiasis in this patient, by clearly showing broncholiths closely related to the involved bronchi.

The prognosis of broncholithiasis is usually favourable and its treatment is restricted to clinical follow-up. Some invasive procedures may be necessary, depending on location, size and associated complications. In addition to its diagnostic role in central lesions, bronchoscopy may be useful for the removal of broncholiths. Surgery might be indicated in some selected cases and when there is risk of bleeding [6].

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