

Chronic Idiopathic Lithoptysis

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Broncholiths, which usually arise from calcified peribronchial lymph nodes, can be found by radiography or bronchoscopy. We describe the case of a 19-year-old man who had experienced lithoptysis of bronchial hydroxyapatite calculi for over 6 months and who reported having sandy expectoration since childhood. Exhaustive clinical, radiographic, and endoscopic diagnostic studies detected no calcified lesions in the thorax that could explain the origin of the broncholiths. Therefore, we propose that broncholiths may form by mechanisms similar to those involved in calculi formation in other locations.

Key words: *Broncholithiasis. Broncholiths. Expectoration. Hydroxyapatite. Lithoptysis.*

Litoptisis crónica idiopática

La litoptisis (expectoración de piedras) es un hallazgo clínico muy infrecuente. Los broncolitos, secundarios en general a ganglios linfáticos peribronquiales calcificados, se pueden hallar en las exploraciones radiológicas o en la broncoscopia. Describimos el caso de un varón de 19 años con litoptisis, durante más de 6 meses, de cálculos bronquiales de hidroxipatita que refería expectoración de arenilla desde la infancia. A pesar de un extenso estudio diagnóstico clínico, radiológico y endoscópico, no se detectaron lesiones calcificadas en el tórax que pudieran explicar el origen de los broncolitos, por lo que se propone un mecanismo de formación similar al de los cálculos de otras localizaciones.

Palabras clave: *Broncolitiasis. Broncolito. Expectación. Hidroxipatita. Litoptisis.*

Introduction

Lithoptysis (literally, spitting of stones) is characterized by the expectoration of bronchial calculi (broncholiths) that are defined as calcified material in the lumen of the tracheobronchial tree.^{1,2} Intrathoracic calcifications are not uncommon whereas lithoptysis, though described by Aristotle and Galen,³ is very rare. Broncholiths are generally thought to be the result of a postinflammatory process in which a calcified peribronchial lymph node erodes in the airways. The most common causes are tuberculosis and histoplasmosis, but broncholithiasis can also arise as a result of other situations, such as aspiration of bone tissue, calcification *in situ* of an aspired foreign body, erosion and extrusion of calcified bronchial cartilage, and silicosis.^{4,5} Rarely have mineralogical analyses been carried out on such calculi, which consist of calcium and phosphorus.

Our report of a long-standing case of lithoptysis includes a description of the mineralogical findings.

Cause could not be established despite exhaustive diagnostic testing. We also discuss the mechanisms that would explain the formation of such intrabronchial calculi.

Case Description

A 19-year old man came to our hospital complaining of a 6-month history of expectoration of "stones" with sputum. He was a nonsmoker and had no history of respiratory risk factors. Since childhood, he had had a cough with scant thick mucous that could be broken down into a gritty substance between his fingers. When he was 13, the expectoration became more fluid and abundant, and was produced especially after physical exertion. Radiographic images of the chest and paranasal sinuses were normal, and a general assessment and spirometry revealed no impairment. The symptoms persisted intermittently with less intensity for years, generally occurring after physical exercise and lasting 2 or 3 days. The mucous sometimes contained small, dry, and very dense formations that were described as sandy. Bronchodilator treatment was prescribed but later abandoned by the patient as no improvement was evident. Upon arrival at our clinic the patient presented with moderate expectoration. Mucous containing tiny "stones" was produced following forced deep breaths. Several irregular spheroid-shaped green calculi were obtained (Figure 1) after an episode and were later analyzed. Physical examination of the patient was unremarkable. Complete blood count and serum

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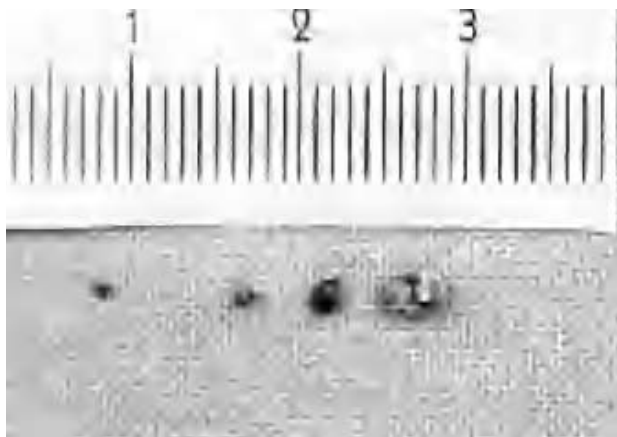


Figure 1. Broncholiths obtained from a single expectoration maneuver.

biochemistry, including immunoglobulin tests, autoimmune serology, and alpha-1 antitrypsin were also normal, as were urine analysis and a sweat test. Results from both the tuberculin test and the respiratory allergen tests were negative. Values obtained for respiratory function tests (spirometry, static lung volumes, and carbon monoxide transfer) were within the normal range. No abnormalities were detected in the chest x-ray, and bronchoscopy revealed no endobronchial lesions or broncholiths although a dark and solid round formation of less than 1 mm was observed in the bronchial aspirate. Microbiological and cytological studies of the bronchial aspirate were negative. A spiral computed tomography (CT) scan with 1 mm slices detected no bone density lesions, bronchiectasis, or other alterations in the lung or the mediastinum. Mineralogical analysis of the broncholiths (stereoscopic and scanning electron microscopy and energy-dispersive x-ray microanalysis) showed hydroxyapatite to be the main component. Organic material was also present (Figure 2). The structural pattern was the same as that observed in salivary gland stones and in the hydroxyapatite kidney stones unrelated to infection. In the following months, the amount of expectoration, which had been very thick and sandy, progressively decreased, and no other symptoms appeared.

Discussion

A review of the Spanish literature yielded a single case of lithoptysis⁶ among 3 reported cases of broncholithiasis.⁶⁻⁸ However, it is possible that both broncholithiasis and lithoptysis occur more frequently than believed. In a series of 25 patients in whom bronchoscopy was performed for refractory chronic cough, broncholithiasis was detected in 2 cases,⁹ and it is known that the majority of patients with broncholithiasis do not report their lithoptysis unless they are explicitly asked.^{1,10} The signs and symptoms of broncholithiasis arise from the bronchial erosion, irritation, and distortion caused by the broncholiths. These formations may also lead to chronic cough—occasionally with purulent expectoration, hemoptysis,

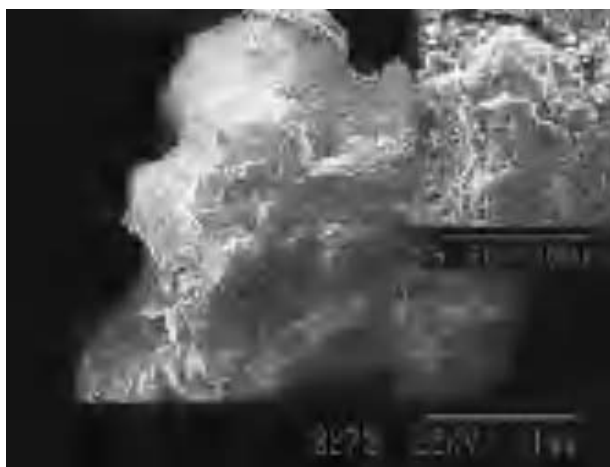


Figure 2. Scanning electron microscope image of a broncholith. A higher magnification in the upper frame shows the presence of both hydroxyapatite and organic material.

lithoptysis, recurrent pneumonia, and fistulas between the bronchus and the adjacent mediastinal structures. In a series of 95 patients with bronchoscopically visible broncholiths, the most common symptoms reported were cough and hemoptysis.¹⁰ In 2 exhaustive series, only 15% of patients with broncholithiasis reported lithoptysis.^{1,10} Expectored broncholiths vary in size from “grains of sand” up to a calculus of 135 g reported by Samson and Rossoff.¹¹ Interestingly, several reported case series include patients who had been previously diagnosed with asthma after presenting with cough, wheezing, and purulent expectoration.¹²⁻¹⁴ Bronchoscopy is effective in the treatment of broncholithiasis. Loose broncholiths and many that are partially adhered to the bronchial wall can be extracted by this method¹⁰ although in some cases laser treatment may be necessary¹⁵ and in exceptional cases, surgery.¹⁶

The source of broncholiths is not usually difficult to find in a chest x-ray or CT scan and can normally be seen directly in bronchoscopy. CT scanning, especially with very thin slices (like those used in the case we report), can easily detect broncholiths.^{4,17,18} Our patient had no history of tuberculosis and did not react to the tuberculin test. No broncholiths were observed during bronchoscopy, although “sand” was obtained in the bronchial aspirate, and no calcifications were detected in the CT scan (1 mm sections) along the entire chest. As a result, we could not establish a point of intrathoracic calcification due to an earlier disease process that could explain the source of the broncholiths.

The childhood history of gritty expectoration together with the calculi expelled over several months and no evidence of an intrathoracic calcification is extremely rare; however, a mechanism of calculi formation similar to that of urolith or sialolith production could explain the clinical findings.^{19,20} Such formations are structurally similar to the broncholiths of our patient. Calculi production in organic fluids is the result of the physical and chemical conditions present. Body fluids (saliva,

urine, blood, or interstitial fluid) are usually in the state of supersaturation and therefore constitute an unstable system favorable to the formation of crystals. While crystallization is impeded by inhibitors and by fluid replacement, greater supersaturation and the presence of crystallization promoters (heterogeneous nucleants) will favor the process. Blood and interstitial fluids are supersaturated with calcium phosphates, or hydroxyapatite, in which case cellular debris and other waste products (such as bacterial debris) may act as very effective heterogeneous nucleants.^{19,20}

The few previous studies about broncholith composition merely identify the presence of calcium and phosphorus. In the case we report, a complex mineralogical study (stereoscopic and scanning electron microscope and energy-dispersive x-ray microanalysis) was carried out to provide information in greater depth for the first time. Our analysis revealed that the composition and microstructure of the broncholiths in our patient were completely analogous to those of sialoliths and of noninfective hydroxyapatite renal calculi. Therefore, the formation mechanisms of these concretions must be very similar and involve factors related to a deficit of crystallization inhibitors, presence of organic material, and fluid stasis.^{19,20} Gritty formations in the bronchial tree require prolonged inhibitor deficiency, long-term presence of intraluminal fluid (due to excess filtration or reabsorption defect), and the presence of abundant organic material (in this case, mucoproteins). The hydrodynamics of such a system trigger the development of small particles that, if retained, may grow into a large broncholith. When a broncholith arises from lymph node calcification, the large amount of unabsorbed organic material and the reduced flow of fluids may be the main factors responsible for calculi formation although crystallization inhibitor deficiency in lymphatic fluid may also be implicated. The absence of intrathoracic calcifications in our patient as well as the negative microbiological tests would suggest that the mechanisms of production of such broncholiths is analogous to those operating to create the better known types of calculi.

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