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Marta López Porras,[a,*] Ana Isabel Blanco Orozco,[a] and María de la Cruz Marchena Parra[b]

[a]*Servicio de Cirugía Torácica, Hospital Universitario Virgen del Rocío, Sevilla, Spain*
[b]*Servicio de Anatomía Patológica, Hospital Universitario Virgen del Rocío, Sevilla, Spain*

*Corresponding author.
E-mail address: martalopezporras@hotmail.com (M. López Porras).

Recurrent Lithoptysis in a Patient With Bronchiectasis

Litoptisis recurrente en paciente con bronquiectasias

To the Editor:

Lithoptysis, referring to stone expectoration, comes from the Greek *lithos* (stone). Although stone expectoration has been known since the time of Aristotle in the 4th century BC, it is rare, associated with an equally rare entity, broncholithiasis, or the presence of calcified material inside or near the bronchial tree. Such calcification can erode the mucosa and cause symptoms.

We report the case of a 39-year-old woman, an ex-smoker, with a history of typhoid fever. She reported having had cough and purulent sputum since childhood. For the last 10 years she had experienced wheezing, not primarily at night or associated with exercise. She also reported coughing up whitish, foul smelling, calcified material on 3 occasions. During the physical examination, end-expiratory wheezing in the left hemithorax was noted. Blood tests; arterial blood gas analysis; chest radiographs; a protein study; determinations of immunoglobulin G subclasses, immunoglobulin E, and α_1-antitrypsin levels; and bone metabolism, sweat, and mucociliary transport tests all gave normal results. A Mantoux test, sputum microbiology, and skin tests with respiratory allergens were negative. Lung function tests were normal. High resolution computed tomography of the chest showed cylindric bronchiectasis in the lingula and left lower lobe, with no intrathoracic calcifications. Fiberoptic bronchoscopy revealed indirect evidence of bronchiectasis but no visible broncholiths. Microbiology and cytology of bronchial aspirates were negative.

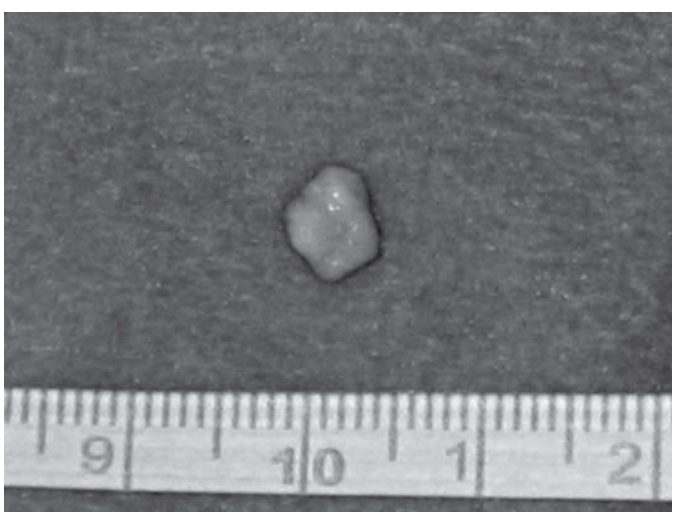

Figure. Yellowish-white coral-like calculus, 5-7 mm, expectoration by yhe patient.

Postinfectious bronchiectasis was diagnosed. During a follow-up visit the patient reported lithoptysis with coughing. She brought a yellowish-white, foul-smelling, coral-like piece of 5 to 7 mm in diameter (Figure). Mineral analysis showed it to contain calcium oxalate (85%) and calcium phosphate (15%). Cultures for bacteria, fungi, and mycobacteria were negative. The pathologist reported finding signs of mucus and cell detritus. Since the patient's condition subsequently improved, a wait-and-see approach to follow-up was taken.

To our knowledge, there are only 4 cases of stone expectoration described in the Spanish medical literature.[2-5] The characteristics of these cases are shown in the Table. Present in 15% of patients with broncholithiasis,[4] lithoptysis may run a chronic, recurrent clinical course, with calculi coughed up with purulent expectoration or blood.[2] The size of the calculi may range from less than 1 mm (described as sandy expectoration)[4] to 135 g.[1] Stones may range in number from 1[5] to, more commonly, several.[2-4] This phenomenon is associated with tuberculosis, primary ciliary dyskinesia, and pneumoconiosis.[2]

Lithoptysis is among the clinical signs of broncholithiasis, which occurs commonly in the fifth and sixth decades of life and involves mainly the right bronchial tree. Implicated in the pathogenesis of broncholithiasis are such phenomena as dystrophic calcifications due to heterogeneous nucleates (inducers of crystallization),[4] local alkalinization, and bronchial erosion by granulomas or calcified foreign bodies.[1,6] Primarily associated with tuberculosis in Europe and histoplasmosis in North America, broncholith formation has been described in relation to other infections, such as actinomycosis, cryptococcosis, nocardiosis, aspergillosis, and coccidiodomicosis.[1] It has also been reported with pneumoconiosis, pulmonary neoplasms, and primary ciliary dyskinesia.[1,6] The calculi are composed of salts-M-dosh calcium, phosphate, or oxalate-M-dosh and organic material.[4] The pathophysiology can be broken down into mechanisms that are local (bronchial irritation, erosion, and distortion), regional (bronchial obstruction, retention of secretions, and infection of the distal bronchial region), and distant (fistulization and migration to mediastinal structures).[3-5] The most common manifestations of broncholithiasis are hemoptysis and long-standing dry cough. In 11% to 61% of cases, purulent expectoration and fever associated with lung infections and abscesses may be present. Wheezing owing to mechanical obstruction of the airways can be heard on examination.[6] A diagnosis requires evidence of intrabronchial or peribronchial calcified material. Fiberoptic bronchoscopy can visualize broncholiths 28% to 56% of the time. This relatively low sensitivity is attributed to peribronchial adenopathies and bronchial distortion. The most common radiologic findings are calcified lymph nodes, though there may also be atelectasis, mucus plugs, air trapping, or bronchiectasis.[1]

Findings of bronchiectasis have led to the hypothesis that there is an association between the 2 conditions. Various authors have suggested that the bronchial obstruction generated by broncholiths favors infection of the distal bronchial tree and, therefore, the

Cases of Lithoptysis in Spanish Medical Literature

	Verea-Hernando et al[2]	Carvajal Balaguera et al[3]	García Pachón et al[4]	Roig Vázquez et al[5]	Present case
Age, y	47	57	19	34	39
Sex	Man	Woman	Man	Man	Woman
Medical history	Smoking, pneumoconiosis, tuberculosis, chronic renal failure	Smoking, renal duplication, ovarian cyst, fibroadenoma of the breast	Mucoid sputum	PCD, idiopathic pleural effusion, BHR, tuberculosis	Smoking, typhoid fever
Associated clinical Manifestations	Purulent sputum	Cough aggravated by swallowing, blood-tinged sputum, reduced dietary intake	Expectoration	Cough	Cough, purulent expectoration, wheezing audible to the patient, dyspnea
Duration of lithoptysis	Several previous hospitalizations with this sign	2 mo –	Since early childhood	A single episode	10 y
Macroscopic appearance	Multiple grayish nodules, 2-3 mm		Sandy particles and greenish spheroid calcul	Whitish, irregular-shaped, coral-like calculi with a hard surface	Yellowish-white, foul-smelling, coral-like calculi, 5-7 mm
Compositions	Calcium phosphate, calcium carbonate, iron, copper, zinc	–	Calcium phosphate, organic material	Calcium oxalate (70%), calcium phosphate (30%)	Calcium oxalate (85%), calcium phosphate (15%), mucus, cell detritu
Imaging techniques	Radiograph: calcified, enlarged hilar and mediastinal lymph nodes	Radiograph and CT: calcified, enlarged paratracheal, and tracheobronchial nodes	Radiograph and CT: no abnormal findings	CT: bilateral bronchiectasis and broncholithiasis	Radiograph: normal. CT: bronchiectasis in lingula and LLL
Bronchoscopy	–	Endobronchial granuloma	Round, dark formation <1 mm in BA	Normal	Indirect signs of bronchiectasis
Approach	Wait and see	Surgical repair of bronchoesophageal fistula	Wait and see	Wait and see	Wait and see

Abbreviations: BA, bronchial aspirates; BHR, bronchial hyperreactivity; CRI, chronic respiratory insufficiency; CT, computed tomography; LLL, left lower lobe; PCD, primary ciliary dyskinesia.

formation of bronchiectases.[1] Others have argued the opposite, that the repeated infections in bronchiectasis can play a role in the genesis of broncholiths and, consequently, cause lithoptysis.[5] Our patient's history of typhoid fever seems to explain her localized bronchiectasis, for which reason we defend the relevance of the second theory to this case.

A wait-and-see approach is occasionally adopted for the treatment of broncholithiasis owing to the self-limiting nature of the condition.[2,4,5] Fiberoptic bronchoscopy enables calculi to be extracted, and the technique can be enhanced with endobronchial laser therapy. Surgical treatment is reserved for cases in which there is a high risk of hemorrhage or an associated fistula.[4] We decided on a conservative approach because of our patient's spontaneous improvement and the fact that no broncholithiasis was observed. The prognosis is usually favorable given that symptoms often disappear with expectoration of the calculi and there remains the possibility of effective treatment.[6]

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José Norberto Sancho,* Eusebio Chiner, and Ana Camarasa

Secció de Pneumologia, Hospital Universitari Sant Joan d'Alacant, Sant Joan d'Alacant, Alacant, Spain

*Corresponding author.
E-mail address: chiner_eus@gva.es (E. Chiner).