

## Letters to the Editor

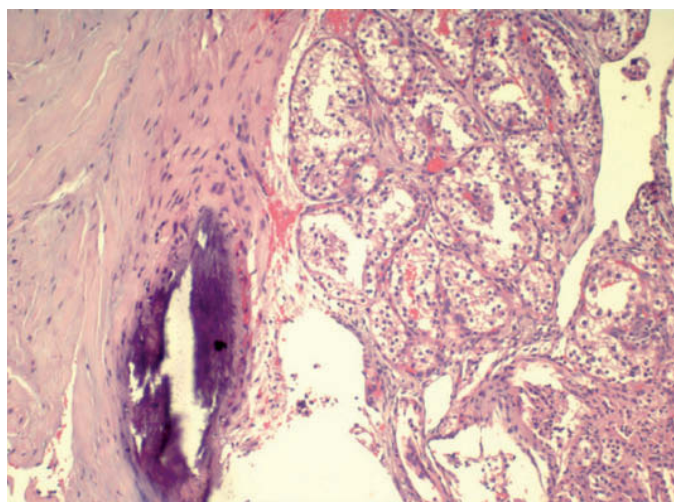
### Intrasternal Parathyroid Adenoma

#### *Adenoma paratiroideo intraesternal*

##### To the Editor:

The 4 parathyroid glands arise embryologically from the third and fourth branchial pouches. During development, they descend with the thymus, a migration that leaves accessory glands in its path.<sup>1</sup> Parathyroid adenomas occur in an ectopic location in 5% to 10% of cases and are most commonly found in the anterosuperior mediastinum. We report the case of a woman with a parathyroid adenoma in the sternum whose only symptom was local pain.<sup>2</sup>

The patient was a 77-year-old woman who had undergone amygdalotomy, was allergic to penicillin and phenylbutazone, and presented arterial hypertension, dyslipidemia, and a duodenal ulcer. For the last year, the patient had been suffering presternal pain radiating to both submammary regions. Physical examination only revealed local pain in the middle third of the sternum and the laboratory tests were normal. A computed tomography scan and magnetic resonance imaging revealed permeative cortical erosion with bone expansion in the lower two-thirds of the sternum. A tumor was suspected, so the patient underwent midline sternotomy with biopsy and removal of the lesion. Macroscopic inspection revealed an osteolytic lesion with destruction of the sternum and profuse bleeding that was hard to control. The pathology report indicated parathyroid adenoma, with histochemical findings consistent with such a tumor. Postoperative recovery was uneventful.



**Figure.** Histological image: parathyroid gland in relation to the bone (periosteum).

The prevalence of parathyroid adenoma is 1%, with incidence peaking between 20 and 50 years. It is more common in women (a ratio of 3:1)<sup>1</sup> and has been described in the following sites: tracheoesophageal groove (27%), anterior mediastinum/thymus (18%), superior parathyroid gland (13%), inferior parathyroid gland (12%), intrathyroid (10%), carotid sinus (3.7%), undescended parathyroid gland (8.4%), and others (7.9%).<sup>2</sup> There are usually no symptoms and diagnosis is generally based on a chance finding of persistent hypercalcemia in laboratory tests. In the present case, the patient had no laboratory abnormalities that might lead to suspicion of parathyroid adenoma. Additional tests performed when this process is suspected include a computed tomography scan and technetium Tc 99m sestamibi scintigraphy, which together achieve 100% sensitivity and a positive predictive value of 97.4%.<sup>2,3</sup> A firm diagnosis is obtained by pathological assessment. Macroscopically, parathyroid adenomas are oval shaped and surrounded by a capsule of brown connective tissue. Microscopically, they are characterized by an abundance of cellular material. They can contain any type of parathyroid cell although principal cells predominate. The cells have a variable nuclear size, large hyperchromatic nucleoli, and infrequent mitoses that do not indicate malignancy. The Figure shows images of histological studies of the present case in which cells specific to parathyroid adenomas are clearly visible, together with bone cells from the sternum.

Treatment will depend on the presence of symptoms and compliance with the criteria of the World Health Organization.<sup>1</sup> Since removal of the affected gland is the only curative treatment, surgery is clearly indicated. Depending on location, the most frequently employed surgical approaches include cervicotomy, sternotomy, thoracotomy, and video-assisted thoracoscopy.<sup>4</sup>

After an exhaustive literature search in Medline, we found no cases in the location we report. There have been reports in the literature of a so-called "brown tumor" that mimicked a parathyroid adenoma in patients with hyperparathyroidism.<sup>5,6</sup> However, additional tests (thallium 201 and Tc 99m sestamibi scintigraphy), but not the pathology report, indicated parathyroid gland adenoma.

### References

- Potts JT. Enfermedades de las glándulas paratiroides y otros procesos hipercalcémicos e hipocalcémicos. In: Braunwald E, Kasper DL, Fauci A. Harrison. Principios de medicina interna. Vol. 2. 15th ed. Madrid: McGraw-Hill; 2005.
- Jaskowiak N, Norton J, Alexander HR, Doppman JL, Shawker T, Skarulis M, et al. A prospective trial evaluating a standard approach to reoperation for missed parathyroid adenoma. *Ann Surg.* 1996;224:308-20.
- Ng P, Lenzo NP, McCarthy MC, Thompson I, Leedman PJ. Ectopic parathyroid adenoma localized with sestamibi SPECT and image-fused computed tomography. *Med J Aust.* 2003;179:485-7.
- Amar L, Guignat L, Tissier F, Richard B, Vignaux O, Fulla Y, et al. Video-assisted thoracoscopic surgery as a first-line treatment for mediastinal parathyroid adenomas: strategic value of imaging. *Eur J Endocrinol.* 2004;150:141-7.

5. Kumar R, Gupta R, Malhotra A. Brown tumor of the sternum: a potential source of false-positive Tl-201 and Tc-99m subtraction imaging in the mediastinum. Clin Nucl Med. 2000;25:44-7.
6. Kao CL, Chang JP, Lin JW, Lin CC. Brown tumor of the sternum. Ann Thorac Surg. 2002;73:1651-53.

Marta López Porras,<sup>a,\*</sup> Ana Isabel Blanco Orozco,<sup>a</sup>  
and María de la Cruz Marchena Parra<sup>b</sup>

<sup>a</sup>Servicio de Cirugía Torácica, Hospital Universitario Virgen del Rocío,  
Sevilla, Spain

<sup>b</sup>Servicio de Anatomía Patológica, Hospital Universitario  
Virgen del Rocío, Sevilla, Spain

\*Corresponding author.

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

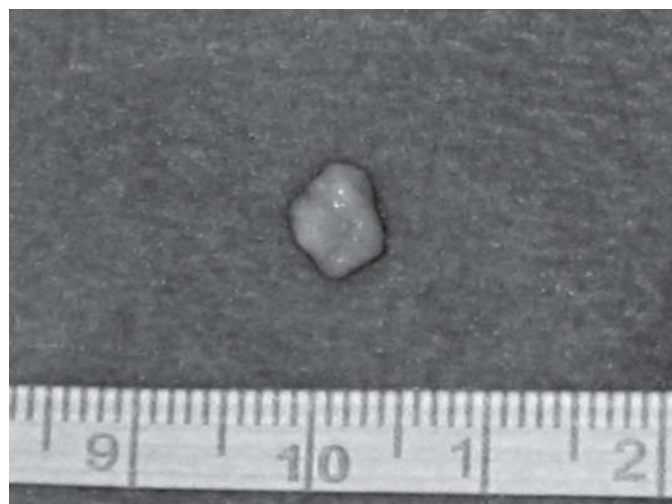
## Recurrent Lithoptysis in a Patient With Bronchiectasis

### *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  $\alpha_1$ -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 cylindrical 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 the 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.<sup>2-5</sup> The characteristics of these cases are shown in the Table. Present in 15% of patients with broncholithiasis,<sup>4</sup> lithoptysis may run a chronic, recurrent clinical course, with calculi coughed up with purulent expectoration or blood.<sup>2</sup> The size of the calculi may range from less than 1 mm (described as sandy expectoration)<sup>4</sup> to 135 g.<sup>1</sup> Stones may range in number from 1<sup>5</sup> to, more commonly, several.<sup>2-4</sup> This phenomenon is associated with tuberculosis, primary ciliary dyskinesia, and pneumoconiosis.<sup>2</sup>

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),<sup>4</sup> local alkalinization, and bronchial erosion by granulomas or calcified foreign bodies.<sup>1,6</sup> 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 coccidioidomycosis.<sup>1</sup> It has also been reported with pneumoconiosis, pulmonary neoplasms, and primary ciliary dyskinesia.<sup>1,6</sup> The calculi are composed of salts—M-dosh calcium, phosphate, or oxalate—M-dosh and organic material.<sup>4</sup> 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).<sup>3-5</sup> 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.<sup>5</sup> 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.<sup>1</sup>

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