Case Report

Endobronchial angioleiomyoma: Diagnostic difficulties of a rare lung neoplasm

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Submission: 15-09-2023
Revised: 06-12-2023
Accepted: 15-04-2024
Published: 25-04-2024

Abstract:

Angioleiomyoma is a benign soft-tissue tumor that rarely develops in the respiratory tract. Here, we report a case of a 51-year-old female with an angioleiomyoma developed in the left lobar bronchial branch and extended to the left principal bronchus, causing nonspecific symptoms, and not visible on the chest X-ray examination. The suspected diagnosis was established by high-resolution computed tomography and confirmed by the histological evaluation of the endoscopically removed lesion.

Keywords:

Angioleiomyoma, bronchial neoplasm, vascular leiomyoma

Among the benign soft-tissue tumors, angioleiomyoma or vascular leiomyoma accounts for ~4.4% of all being soft-tissue tumors and is located anyplace preferring the hypoderm of the inferior limbs: only 2.4% are located at the trunk.[1] Few cases are reported in the lungs.[2] Histologically, the lesion is characterized by well-differentiated smooth muscle cells surrounded by several blood vessels.[3]

Here, we present a case of angioleiomyoma arising from the left lobar bronchial branch and extended to the main left bronchus in a paucisymptomatic patient revealed by chronic dry cough.

Case Report

A 51-year-old woman (nonsmoker), an amateur swimmer, comes to our attention with a chronic dry cough suddenly insurgent 4 months before our first clinical evaluation.

The medical history shows no signs of any sufferance of the pulmonary tract and general symptoms of gastroesophageal reflux disease (GERD).

Previously, the patient was surgically treated for the removal of the entire right ovary and for a partial left oophorectomy, followed by a combined estrogen–progestogen therapy, which continued up to now.

Physical examination of the thorax showed no pathological signs. Common blood tests showed normal range results. Before our clinical evaluation, the patient underwent a spirometer examination, which showed values on average. Because of the paucity of symptoms and the scarcity of signs detectable by radiographic investigations, she was initially treated with antihistamine and corticosteroid, albeit with partial benefit, to finally arrive at a diagnostic evaluation by esophagogastroduodenoscopy, for which under a guide of a gastroenterologist, she received a general GERD medical treatment.

The patient arrived at our evaluation because of the persistence of symptoms resilient to the prescribed pharmacologic therapy. We, thus, decided to evaluate the conditions of the patient pulmonary tract

Access this article online

Quick Response Code:

Website: www.thoracicmedicine.org

DOI: 10.4103/atm.atm_223_23

How to cite this article: Orlando M, Alimandi M, Monteleone G. Endobronchial angioleiomyoma: Diagnostic difficulties of a rare lung neoplasm. Ann Thorac Med 2024;19:172-4.

using high-resolution computed tomography (HRCT) of the lungs. This analysis shows a tubular bronchiectasis in the anterior segment of the left upper lobe occupied by hypodense tissue extended to the main left bronchus [Figure 1].

The fibrobronchoscopy procedure detected the presence of dense material mixed with blood in the main left bronchus, which was lately removed. The histological evaluation showed a tissue composed of melted cells with smooth muscle differentiation, positive for immunohistochemical staining for desmin [Figure 2] and actin.

These characteristics are compatible with the diagnosis of a primary solid-type angioleiomyoma. The endoscopic removal of the primary lesion determined the complete resolution of the respiratory symptoms. The remission of symptoms persists in the 12-month follow-up after the fibrobronchoscopy procedure.

Discussion

Vascular leiomyoma is an uncommon neoplasm potentially developing in any district of the body, and, among leiomyomas, is the most frequent in peripheral soft tissues. It typically appears in women between the fourth and sixth decades,[4] as neoplasms of cutaneous or subcutaneous tissues.

It stands out from other tumor lesions involving the proliferation of smooth muscle cells, for the spike form of cellular elements, for the growth pattern that is linked to the development of neovascularization surrounding tumors, and for a benign clinical course. As reported by McCarthy and Chetty,[5] vascular leiomyomas share an identical immunophenotype with leiomyomas of the deep soft tissues, showing the same positivity for the immunostaining of smooth muscle actin and desmin. Association with a vein wall is the feature to distinguish vascular leiomyomas from leiomyomas of deep soft tissue.

The diagnosis of angioleiomyoma is possible only by histological examination. A histomorphological classification divides angioleiomyoma into three different histological subtypes: (a) cavernous-type tumor with widened vascular channels and a few muscle bands, (b) venous-type tumor with vases surrounded by thick walls, and (c) solid-type tumor (the tumor histotype described in our case) characterized by the presence of strictly intertwined muscle bundles surrounding the vascular channels.[3] The last is the most frequent histotype in women.

The occurrence of angioleiomyoma in the lungs is exceptional. To the best of our knowledge, this is the second report in the literature describing the occurrence of a vascular leiomyoma in a bronchus. While in that case, the neoplasm became clinically manifested with self-progressive severe dyspnea,[6] in our patient, the only manifestation of the illness was a dry cough. In the last clinical situation, and after the exclusion of life-threatening situations, elements for the diagnosis include accurate anamnestic history, chest X-ray examination, and previous general and empirical treatment aimed to treat common causes of chronic cough.[7]

A timely diagnosis of endobronchial vascular leiomyoma appears to be difficult and hampered or retarded because it is rare and accompanied by nonspecific and sometimes misleading symptoms.

To this extent, the sensitivity of the radiographic examination for malignant neoplasms of the lungs is between 79% and 82%.[8] The reported case had a negative chest X-ray examination, and some overlapping signs compatible with the GERD diagnosis.

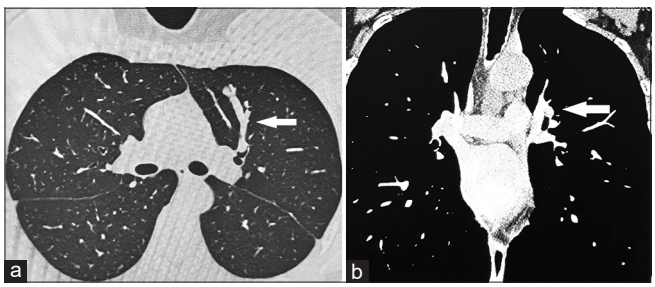

Figure 1: (a and b) High-resolution computed tomography imaging scan showing tubular bronchiectasis in the anterior segment of the left upper lobe occupied by hypodense tissue material extended to the main bronchus (arrows)

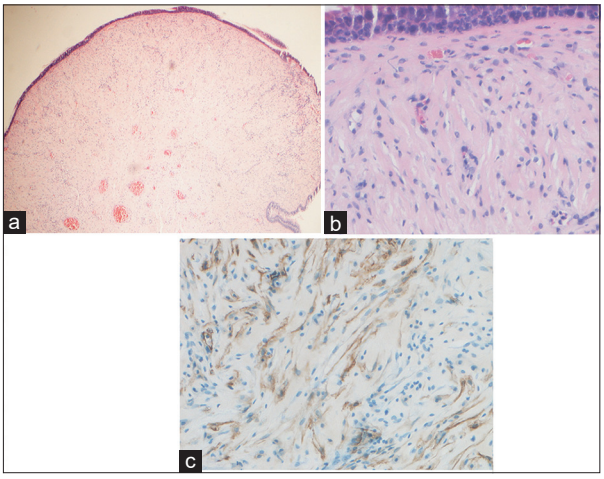

Figure 2: (a) H and E, ×25 lower-power histologic examination shows a sharply marginated lesion with vascular channels surrounded by ciliated epithelium. (b) H and E, ×200 showing a hypocellular proliferation consisting of spindle cells with differentiation of smooth muscle cells, thin-walled vessels, and absence of mitosis. (c) Desmin immunostain ×200 highlighting positivity for smooth muscle desmin

Furthermore, as stated by Katz *et al*.,[9] it may be difficult to establish that GERD is the cause of extraesophageal problems; GERD therapy is normally administered for at least 8 weeks, a period during which the patient accuses respiratory distress significantly impairing his/her quality of life.

The history of the case presented here suggests a revaluation of the instrumental and imaging examinations indicated in the persistence of nonspecific respiratory symptomatology, mainly chronic cough without evidence of lung disease, and a reflection on the role of HRCT in the early phase of the clinical course of these patients.

It is worth mentioning that some authors postulated that local and general conditions, such as venous stasis, local infection, trauma, arteriovenous malformation, and estrogen hormone changes, may play a role in the pathogenesis of angioleiomyoma.[10] We hypothesize that in the reported case, the long-lasting estrogen–progestogen therapy might have influenced the development of the lesion.

In some cases, postsurgical recurrence or rapid regrowth is possible, especially when the development district makes a complete excision difficult; for this reason, it is necessary to program an instrumental clinical follow-up for at least for 10 years from the fibrobronchoscopic resection procedure.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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