Case Report

Intrapulmonary teratoma*

Ricardo Alexandre Faria¹, José Alexandre Bizon², Roberto Saad Junior³, Vicente Dorgan Neto⁴, Marcio Botter⁵, Mauro Ajaj Saieg⁶

Abstract

Case report of a 49-year-old man, presenting chest pain and bloody sputum for six months. Chest X-ray and computed tomography scan showed opacification in the left upper lobe. The bronchoscopy showed bronchial hemorrhage in the lingular bronchial segment. Due to diagnostic and therapeutic needs, this patient underwent a left inframamillary thoracotomy. The anatomopathological analysis of the surgical sample revealed an intrapulmonary teratoma. The patient presented favorable evolution and is now under outpatient follow-up treatment.

Keywords: Teratoma; Lung; Neoplasms.

Introduction

Teratoma can be found in various organs. In decreasing order of location, it is seen in the ovaries, testicles, sacro-cocygeal region, mediastinum, and other sites. (1-4) In the international literature, 43 cases of intrapulmonary teratoma have been described, the first in 1939. (3)

Intrapulmonary teratoma originates from totipotential cells of one or more of the three germinative layers, which can differentiate themselves into any type of tissue. (2,3) There are no specific clinical or radiological characteristics, and the anatomopathological study is the definitive diagnostic method. (1,3,4) Curative treatment consists of complete resection of the tumor. (3,4)

Case report

We report the case of a 49-year-old male Caucasian who was a businessman from Italy and a resident of São Paulo. The patient sought emergency treatment, presenting with bloody sputum and chest pain in the left anterolateral region for six months. The epidemiology was negative for tuberculosis. He was a 20-pack-year smoker. He presented good general health status. The physical examination revealed discrete wheezing in the middle third of the left hemithorax, with no other alterations. The body mass index was 36.3 kg/m².

A chest X-ray revealed nodular opacities in the middle third of the left hemithorax, in lingular projection, meas-

Correspondence to: Ricardo Alexandre Faria. Rua Paracatu, 398, apto. 3B; Pq. Imperial (Saúde), CEP-04302-021, São Paulo, SP, Brasil.

Tel 55 11 2176-7000. E-mail: rickpreto@zipmail.com.br

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^{*} Study carried out at the Faculdade de Ciências Médicas da Santa Casa de São Paulo - FCMSCSP, Santa Casa School of Medical Sciences in São Paulo - São Paulo (SP) Brazil.

^{1.} Third-year Medical Resident in the Thoracic Surgery Department. Faculdade de Ciências Médicas da Santa Casa de São Paulo – FCMSCSP, Santa Casa School of Medical Sciences in São Paulo – São Paulo (SP) Brazil.

^{2.} Fourth-year Medical Resident in the Thoracic Surgery Department. Faculdade de Ciências Médicas da Santa Casa de São Paulo – FCMSCSP – Santa Casa School of Medical Sciences in São Paulo – São Paulo (SP) Brazil.

^{3.} Full Professor in the Thoracic Surgery Department. Faculdade de Ciências Médicas da Santa Casa de São Paulo – FCMSCSP, Santa Casa School of Medical Sciences in São Paulo – São Paulo (SP) Brazil.

^{4.} PhD Professor in the Surgery Department. Faculdade de Ciências Médicas da Santa Casa de São Paulo – FCMSCSP, Santa Casa School of Medical Sciences in São Paulo – São Paulo (SP) Brazil.

^{5.} PhD Professor in the Surgery Department. Faculdade de Ciências Médicas da Santa Casa de São Paulo – FCMSCSP, Santa Casa School of Medical Sciences in São Paulo – São Paulo (SP) Brazil.

^{6.} Graduate Student in the Pathology Department. Faculdade de Medicina da Universidade de São Paulo – FMUSP, University of São Paulo School of Medicine – São Paulo (SP) Brazil.

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uring approximately five centimeters in diameter (Figure 1), confirmed through a tomography scan of the chest (Figure 2). Sputum culture for Koch's bacillus was negative, and the patient presented no reaction on the purified protein derivative test. Bronchoscopy revealed hematogenous remnants in the lingular bronchial segment, without further alterations. A transbronchial biopsy revealed a nonspecific inflammatory process. Pulmonary function test results showed mild obstructive ventilatory disorder, with a forced expiratory volume in one second of 82%.

In view of the imperative diagnostic and therapeutic conditions, surgery was recommended. A left inframamillary thoracotomy was performed, revealing a tumor in the lingular region, and we opted for a segmentectomy. The frozen section biopsy findings were negative for neoplasia, and the sample remnants were sent for anatomopathological analysis. The patient presented favorable evolution and was discharged on postoperative day 3.

The anatomopathological examination showed that the sample was nodular, weighed 25 g, and measured $5 \times 4.5 \times 3.5$ cm, with a smooth, brownish surface, and that there were pulmonary and adipose tissue adherences. The distal pulmonary margin was normal. Microscopy showed mature epithelial tissues, such as pancreatic, epidermis, sebaceous glands, and mature mesenchymal tissues, such as bone and cartilage (Figure 3). Therefore, we concluded that it was a primary intrapulmonary mature teratoma.

Discussion

Teratomas present slow growth and are typically benign. (3,4) When in the lung, they are most often found in the upper left lobe, as in the case reported. (1-6) Symptomatology varies according to the location and size of the tumor, as well as to the histological components. (3) In this case, the patient reported bloody sputum and thoracic pain which, together with cough, are the most commonly reported symptoms. Other symptoms reported in the literature are trichoptysis, bronchiectasis, fever, abscess, repeated pneumonia, signs of compression of thoracic structures, and fistulas, with or without infection of the tumor. (1-6)

Teratoma is an extremely rare disease that affects both genders and all age brackets equally. (1-6) The size of the tumor has not relationship with malignancy. (1,3,4) On chest X-rays, teratoma can present as lobulated upper lung lobe tumors or intraparenchymal opacities, corresponding to the exam shown above. On tomography scans of the chest, homogeneous or heterogeneous tumors can be observed. (1-3,5,7)

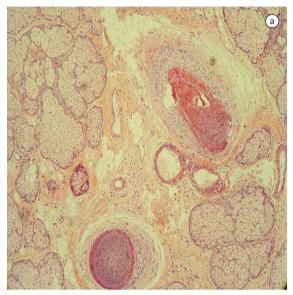
In anatomopathology, these tumors present endodermic components (pancreatic acinar tissue, with or without islets of Langerhans and respiratory epithelium), thus differing those found at extrathoracic sites. Intrapulmonary teratoma originates in cells that can differentiate into any tissue type. In the case reported, microscopy of the resected surgical sample revealed pancreatic tissue, carti-



Figure 1 - Chest X-ray.



Figure 2 - Computed tomography of the chest.



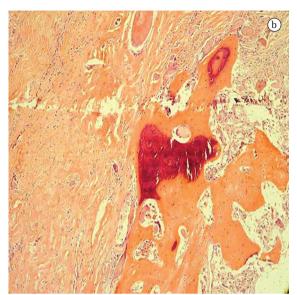


Figure 3 - a) Sebaceous glands and hair follicle; and b) bone parenchyma.

lage, bone, epidermis and sebaceous glands.^(2,3) It has been suggested that intrapulmonary teratoma derives from the thymic tissue of the third branchial arch.⁽²⁻⁶⁾ However, mediastinal teratoma results from the dislocation and early separation of the thymic tissue, during embryogenesis, with capture and migration of thymic primordia during the development of the respiratory system.⁽²⁾

The classification system used in order to categorize teratomas as benign or malignant is that proposed by Gonzales-Crussi in 1982, which has proven practical to date.⁽⁸⁾

- 1) Benign teratomas:
 - a) Mature teratoma

Grade 0 - all components are well differentiated

Grade 1 - non-undifferentiated components do not exceed 10%

b) Immature teratoma

Grade 2 - immature tissues among 10 to 50% of components

Grade 3 - undifferentiated components over 50% and probable metastatic potential.

Benign evolution is still possible.

- 2) Malignant teratomas:
 - a) With germ cell tumor areas Germinoma (seminoma - dysgerminoma) Embryonal carcinoma - Coriocarcinoma Yolk sac tumor

Mixed cell types

b) With non-germ cell tumor areas
Carcinoma - Sarcoma - Malignant embryonal tumor

Mixed cell type

c) Malignant immature teratoma⁽⁸⁾

The prognosis of malignant teratoma is poor. In a study comprising eleven patients with malignant teratoma, seven patients (63.6%) died after a six-month follow-up period. None of these patients underwent surgery, or the tumor was nonresectable by thoracotomy. Metastases were reported in three of these patients.⁽⁴⁾

In patients with benign teratoma, the recommended procedure is complete resection of the tumor, upon which the patient is considered totally cured. Patients who did not undergo surgery can present massive hemoptysis or increased tumor growth and can die. In the case reported, the patient evolved without complications and did not require any further therapeutic measures, as has been reported in the literature, and, after two years of outpatient follow-up treatment, presented no signs of relapse.

In conclusion, intrapulmonary teratoma is a rare tumor, and surgery is the definitive treatment. Definitive confirmation of the diagnosis is made through anatomopathological examination, and prognosis is good when the condition is properly treated.

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