

Endobronchial Teratoma Treated Via Upper Right Upper Lobe Sleeve Resection - A Case Report

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Abstract

Endobronchial teratoma is very uncommon neoplasia in clinical practice. To date, only three cases of primary endobronchial teratoma were reported in the literature among the 50 cases of primary pulmonary teratomas. Here we describe a case of a mature primary endobronchial teratoma located at the bifurcation of the upper lobe bronchus and the intermediate bronchus, macroscopically simulating an endobronchial carcinoid tumor. Its final diagnosis was only possible by a complete study of the permanent section after the surgical resection of the tumor. The treatment option consisted of a right upper lobe sleeve lobectomy and bronchoplasty. The patient has already seven-years of postoperative follow-up with no evidence of recurrence.

Keywords: Lung; Endobronchial Tumors; Benign Lung Tumors; Germ Cell Tumors; Endobronchial Teratoma

Introduction

Endobronchial teratoma is very uncommon neoplasia in clinical practice. Detailed search in medical literature databanks (Pub Med, Scielo, Embase, and Google Scholar) has detected, to date, only 3 cases of endobronchial teratoma among around 50 cases of pulmonary teratoma [1-3]. The search criteria used were associations of the following terms: teratoma, germ cell tumor, mediastinum, thorax, lung, and endobronchial.

The most common form found in the thorax is the mediastinal teratoma in its cystic, homogenous form [4]. We present a case of solid endobronchial teratoma, with a clinical presentation of long-term pneumonia, which was successfully treated with surgery.

Case Report

A sixty-three-year-old non-smoking male sales representative patient complained during the consultation of a dry irritating cough and fever. His symptoms had started approximately 25 days before the consultation. A radiological examination of the thorax showed an image that suggested pneumonia of the right lower lobe. After two weeks of antibiotics, the X-ray examination was repeated, with any radiologi-

cal changes, despite a noticeable clinical improvement. A CAT scan of the thorax was performed, revealing an endobronchial lesion with a smooth surface and areas of fat density (-65 UH) in its inner portion (Figure 1A). The diagnostic hypothesis, at that time, was of a possible hamartoma or endobronchial lipoma. A bronchoscopy was performed, showing a submucosal tumor in the secondary carina between the right upper lobe bronchus and the intermediate bronchus. The endoscopic image suggested an endobronchial carcinoid tumor (Figure 1B), with rough, red, and *strawberry-shaped* appearance. During the biopsy there was significant bleeding, which was controlled with cold saline solution and the biopsies histopathological study were inconclusive. The patient presented normal respiratory function, with FEV1 post right pneumonectomy estimated at 1.60 liters, and surgery was indicated. We started the surgery via muscle-sparing right lateral thoracotomy, with the resection of the carina between the upper lobe and the intermediate bronchus containing the tumor (Figure 2A), without resecting lung parenchyma, aiming to do a complete reconstruction of the right lung. The on-site pathologist made an assessment of the lesion and concluded that it was neither a lipoma or a carcinoid; however, intraoperatively, it was not possible to establish its histological origin or malignancy level. We continued with the surgery, carrying out a right upper lobe sleeve lobectomy with anastomoses of the intermediate bronchus to the trachea. The lesion was yellowish, slightly soft in its consistency, and its macroscopic appearance was of carcinoid or atypical lipoma since it was tougher in touch than an ordinary lipoma. The final histopathology showed that the tumor was composed of cellular and myxoid conjunctive tissue, serous and mucous glands, sympathetic neural structures, glandular structures with the appearance of a salivary gland and epidermis, with no characteristics of malignancy (Figure 2B). Thus, it contained tissue from the three embryonic germ layers (ectoderm, mesoderm, and endoderm), confirming the final diagnosis of a mature endobronchial teratoma. The patient recovered uneventfully during the immediate postoperative period and has completed seven-years of follow-up with no symptoms.

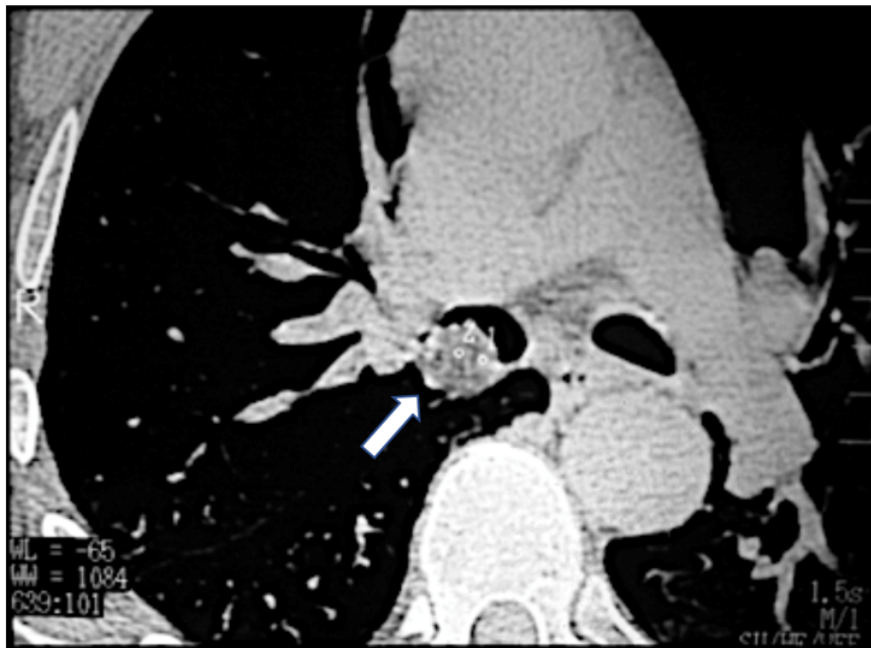


Figure 1A: CT scan of the thorax with mediastinal window showing a lesion in the intermediate bronchus obstructing more than 50% of its lumen (arrow), with low tomographic density of -65UH (fat density).

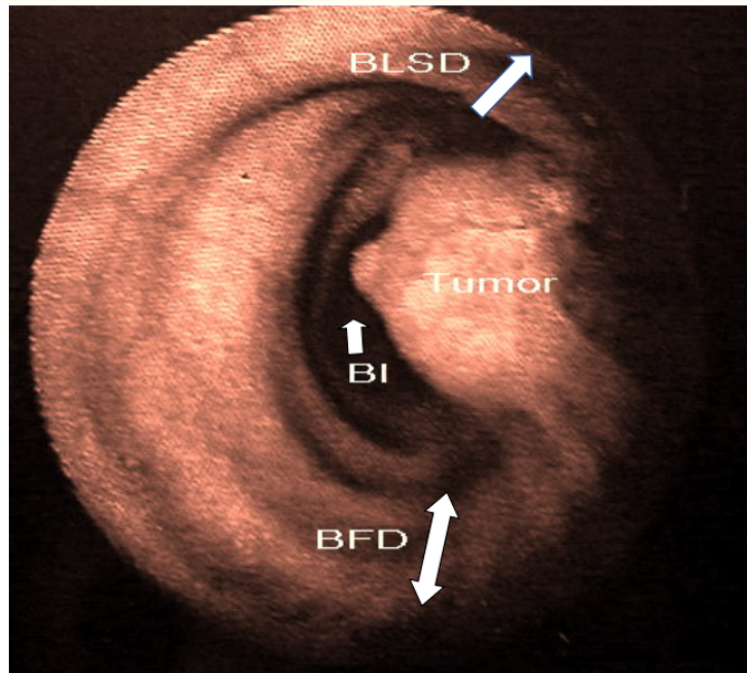


Figure 1B: Endoscopic aspect of the submucosal endobronchial lesion (Tumor), showing its location in the carina between the right upper lobe bronchus (BLSD) and the intermediate bronchus (BI). BFD shows the position and direction of the right main bronchus.

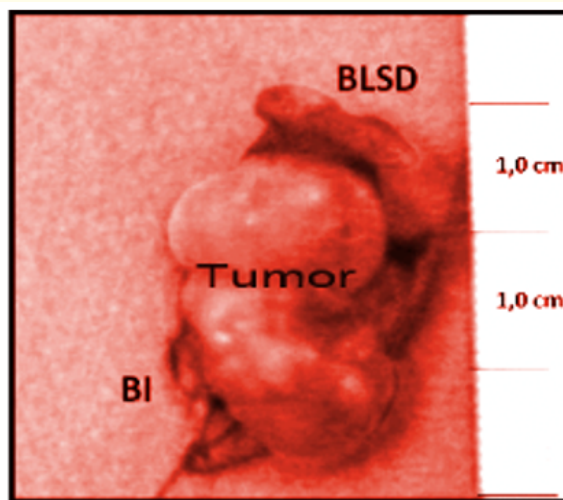


Figure 2A: First surgical piece of the locally resected tumor including only part of the RULB and part of the IB without any parenchymal resection (BLSD: Right Upper Lobe Bronchus; BI: Intermediate Bronchus).

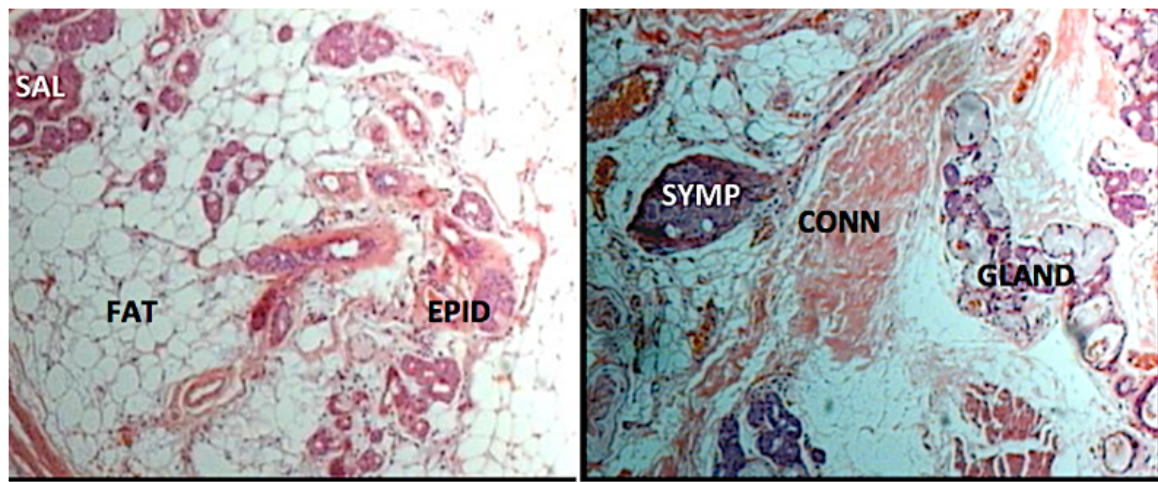


Figure 2B: Two anatomopathological slides (HE - 40X) showing presence of fat tissue (FAT), well-formed sweat glands (GLAND), epidermis (EPID), sympathetic nerve (SYMP), salivary glands (SAL) and connective tissue (CONN).

Discussion and Conclusion

Teratomas have been described as real tumors composed of foreign tissues in part or throughout the organ in which they were found. They are classified as non-seminomatous germ cell tumors and represent a heterogeneous group of tumors that has remaining components of all three embryonic germ layers. Four grades are described, based on the different levels of histological differentiation [5]:

- a. Mature teratoma, grade 0 or differentiated - benign,
- b. Immature teratoma, grade 1 - probably benign,
- c. Immature teratoma, grade 2 - probably malignant,
- d. Immature teratoma, grade 3 - malignant (teratocarcinoma).

Teratomas can be found in any part of the body; however, they are more frequently found in the ovaries, testis, sacrococcygeal region, and mediastinum. All other locations are very rare. For that reason, a detailed physical and imaging examination must be conducted to rule out the presence of a germ cell tumor hidden in an extrapulmonary site [4,6-8].

In the thorax, the anterior mediastinum is the most common extragonadal site of primary tumors derived from multipotential primitive germ cell [9] and is usually derived from the thymic tissue left behind from the third pharyngeal pouch during the displacement or separation of tissue portion from the thymus during embryogenesis.

Pulmonary teratomas are rare and occur more frequently in women. They are usually found in the left upper lobe (over 65%) with adhesions to the anterior mediastinum. It is important to determine the connection with the bronchial mucosa to check for primary pulmonary origin *because of the association to infection, adhesion, and subsequent rupture*.

Mediastinal teratomas are asymptomatic in 53% of the cases, being incidentally discovered during a radiographic examination. Occurring symptoms are mostly related to compression of adjacent structures by the teratoma [1].

The most frequent symptoms of pulmonary teratoma are thoracic pain, hemoptysis, cough, and expectoration. A pathognomonic symptom of mediastinal and pulmonary teratomas is trichophytosis (hair expectoration), which remains exceptional and has been reported only in eight cases in the English and the Japanese literature from 1839 to 1996 [10] or 44 cases in the English literature until 2007 [11]. Mature teratomas are the most common histologic type [12] composed of multiple tissues representing elements of the ectoderm, mesoderm, and endoderm. They often contain fat, discrete or well-formed calcification, or teeth.

To the present date, around fifty cases of pulmonary teratoma have been described. However, up to now, no more than three cases of endobronchial teratoma have been published. In this case, the impossibility of defining the lesion's character (diagnosis) in the pre and the perioperative period justified the aggressive treatment. In some cases, the radiological studies could evidence an intrapulmonary lobulated mass with calcification; also, a peripheral radiolucent area indicating trapped air, suggests a connection between the tumor and bronchial tree, helping to distinguish between intrapulmonary and mediastinal teratomas [13].

The differential diagnosis of thoracic teratomas includes neurogenic tumors, liposarcoma, neuro-enteric cyst, extramedullary hematopoiesis, and extra-lobar pulmonary sequestration [14]. Endobronchial and pulmonary teratoma are other thoracic lesions that might contain fat. In an excellent review of thoracic lesions containing fat, *Gaerte, et al.* [15] only mention lipoma and hamartoma as endobronchial lesions.

Determining the nature and location of mediastinal masses is very important for the correct treatment of the disease. Teratomas demonstrate benign clinical behavior. However, they may recur with an immature, potentially malignant teratoma with a worse prognosis [16]. In the absence of other criteria, the presence of primitive embryonic tissue components classifies a lesion as an immature teratoma and potentially malignant. That is why the accurate assessment of these lesions requires a trained pathologist who has extensive experience in human germ cell neoplasia [17].

With all the advances of endoscopic instrumentation, thoracoscopic and robotic surgery are well accepted as an alternative to removing mediastinal tumors [18]. Surgical resection is always mandatory with favorable prognosis in benign lesions. Minimally invasive surgery has several advantages over open surgery. The surgery has to be done as soon as possible because it will undoubtedly prevent complications [19] as the tumor can grow, compressing adjacent structures or sometimes can even undergo malignant transformation [20].

In this case report, we present an exceedingly rare entity (endobronchial teratoma) that should also be considered as a differential diagnosis of benign endobronchial tumors.

Conflicts of Interest

All the authors declare not to have conflict of interest in this publication.

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