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Endobronchial teratoma: A systematic review of the literature with a case report

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ABSTRACT

INTRODUCTION: Endobronchial teratoma is sporadic disease. The study aims to present a case with endobronchial teratoma with a brief literature review. A 26-year-old male presented with a history of frequent attacks of chest infection for the last two years. Chest examination showed diffuse wheeze all over the left side of the chest. Chest x.ray showed opacification involving all of the left side of the chest with elevated left hemi diaphragm while computed tomography scan confirmed complete collapse of the left lung with consolidations and air bronchogram. Flexible bronchoscopy showed near-total obstruction of the left main bronchus. Under general anesthesia, left pneumonectomy was performed. The result of the histopathological examinataion showed mature teratoma.

CONCLUSION: Endobronchial teratoma is an exceedingly rare type of intrathoracic teratoma that mainly affects males and is usually diagnosed at the 3rd decade of life; lobectomy or pneumonectomy are optimal managements for these cases.

1. Introduction

Teratoma is a rare tumor containing different tissues (teeth, hairs, and others) arising from pluripotent cells of multiple germ cell lines. These tumors are usually benign and are primarily found in the ovaries and testicles (gonadal organs) [1]. Although extragonadal teratoma are highly uncommon, they can still occur in other parts of the body, such as; retroperitoneum, sacrococcygeal region, head and neck. When it occurs extragnadally, it is frequently found in the mediastinum, and with very rare occurrence in the lungs [2]. Intrapulmonary teratomas (IPT) are extremely rare [3]. An equal incidence of IPT has been reported between men and women, that become symptomatic around the 3rd and 4th decades of life [4]. An even rarer incidence inside the lungs, is the endobronchial teratoma (EBT), with only a few reported cases worldwide [5].

The aim of this study is to present a case of endobronchial teratoma with a brief literature review. The report has been written in line with SCARE guidelines [6].

1.1. Patient information

A 26-year-old male student presented with a history of frequent attacks of chest infection for the last two years, responding temporarily to medications. Family, drug, past medical, and past surgical history were negative.

1.2. Clinical findings

a tall, thin patient had generalized weakness and pallor. Chest examination showed diffuse wheeze all over the left side of the chest. Oxygen saturation was 92 % on room air, blood pressure: 110/85 mmHg, pulse rate: 88 beats per minute, respiratory rate: 13 cycles/minute.

1.3. Diagnostic assessment

Hematological tests were within the normal ranges. Chest x.ray showed opacification involving all of the chest's left side with elevated left hemi diaphragm (Fig. 1). Chest computed tomography scan (CT scan) revealed complete collapse of the left lung with consolidations and air bronchogram, the contralateral lung expanded and mediastinum shifted to the ipsilateral side (Fig. 2). Spu-

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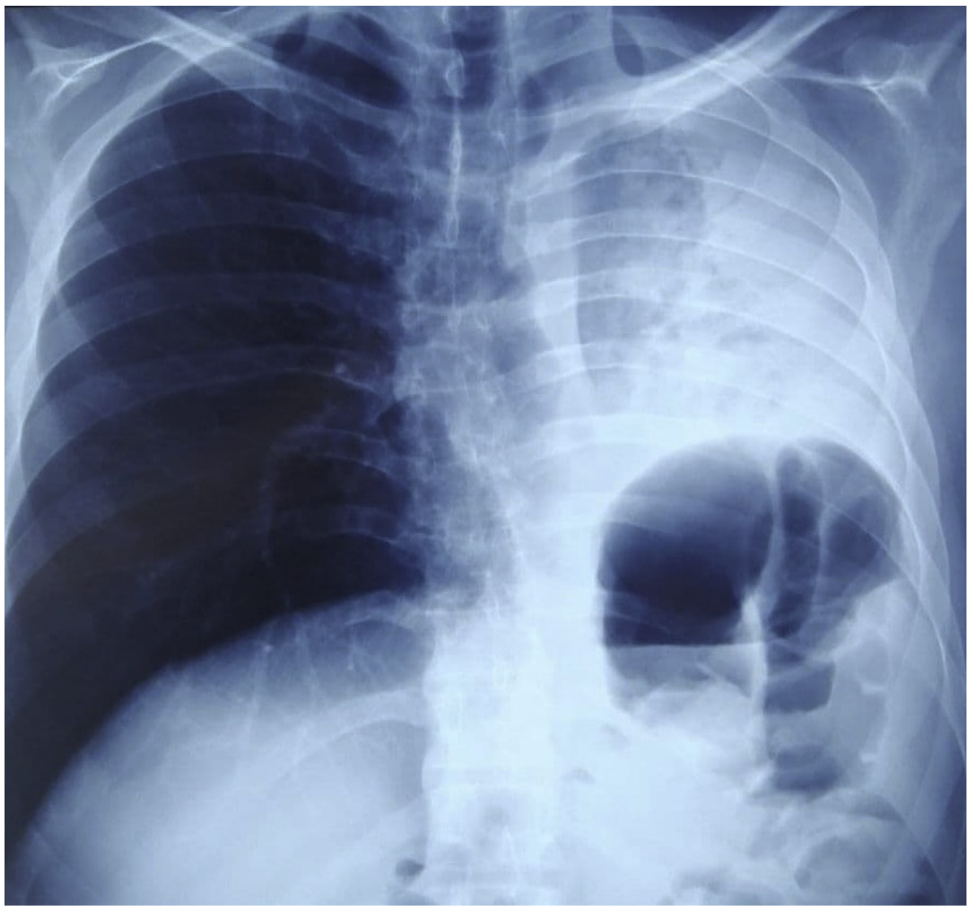

Fig. 1. Postero-anterior plain chest x.ray showing left side opacification with elevated left hemidiaphragm and left-ward mediastinal shift.

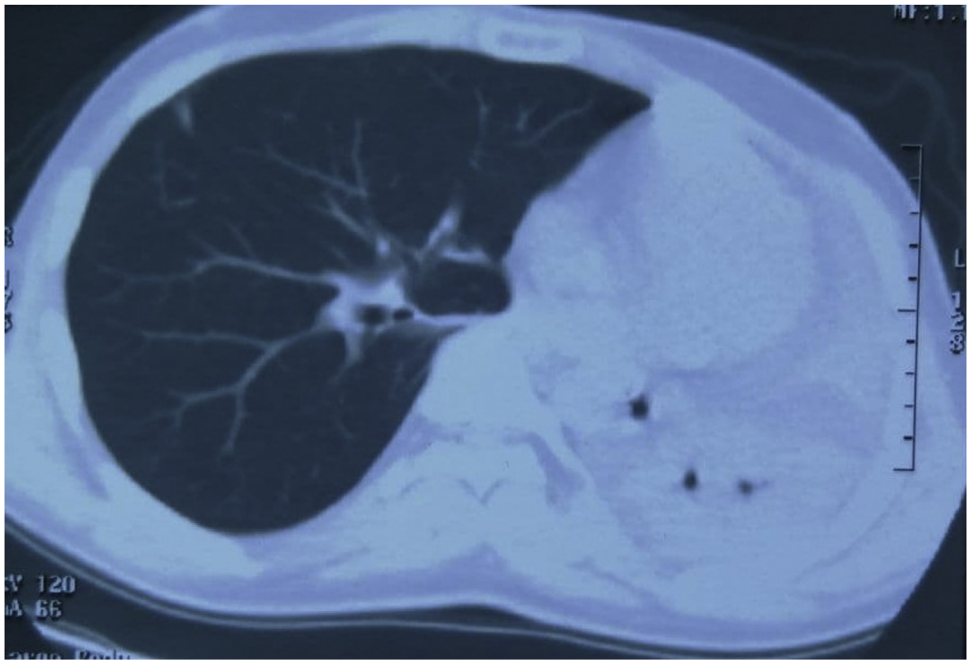

Fig. 2. Axial section chest computed tomography scan showing left side collapse and consolidation with destroyed left lung.

Table 1
XXX.

Authors/ year of publications	Age(year)/Sex	Site	Presentation	Management
Bateson/1968	Unknown/Male	Left anterior segmental bronchus	left pleuritic pain, productive cough, and recurrent haemoptysis	Pneumonectomy
Jamieson/1982	22/Female	Left anterior segmental bronchus	pleuritic chest pain, cough, and haemoptysis.	Lobectomy
Haddad/2020	63/Male	Between the Right main bronchus and Bronchus Intermedius	dry irritating cough and fever	Right upper lobe sleeve lobectomy
Agarwal/2007	28/Male	Left main bronchus	Cough, expectoration, hemoptysis, and trichoptysis	Pneumonectomy
Victor/1982	20/Male	Right anterior segmental bronchus	expectoration, haemoptysis, and fever	Lobectomy
Asano/2000	23/Male	Left main bronchus	left chest pain, fever, general malaise, cough and yellow sputum	Lobectomy
Petrunina/1975	Irretrievable	Irretrievable	Irretrievable	Irretrievable
Kravetz/1976	Irretrievable	Irretrievable	Irretrievable	Irretrievable

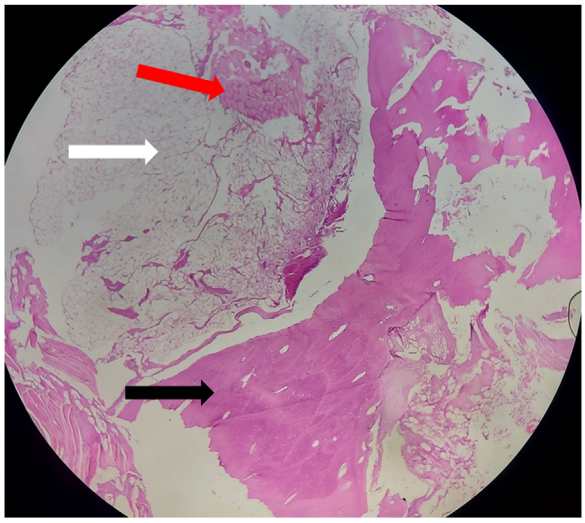

Fig. 3. Histopathological examination of the specimen showing mature bone cells (black arrow) with muscle (red arrow) and adipose (white arrow) tissue.

tum cytology was negative for malignancy. Flexible bronchoscopy under local anesthesia showed near-total obstruction of the left main bronchus a few millimeters distal to the carina by a benign-looking mass. Biopsy showed normal bronchial mucosa.

1.4. Therapeutic intervention

the patient was prepared for left pneumonectomy under general anesthesia. In the right lateral position, through the classical posterolateral incision, left pneumonectomy was performed. The result of the histopathological examination of the specimen showed thick and broad tuberculae of mature bone with associated marrow fat and hematopoietic tissue and a few bundles of normal skeletal muscle interspersed with several large lobules of mature adipose tissue, pictures consistent with mature teratoma (Fig. 3).

1.5. Follow up

the post-operative period was uneventful. The patient was discharged home 3 days after the procedure. The patient was normal and the scare was healthy six months after the intervention.

2. Discussion

Although extragonadal teratomas have a rare occurrence, they can still be found in different parts of the body, most frequently in the anterior mediastinum in adults and sacrococcygeal region in children [7]. Teratomas are mostly considered as slow-growing benign tumors with differentiated tissues, which have a low potency for malignant conversion [2]. IPT is an exceedingly rare type of teratoma; as such, since Mohr first described it in 1839 until 2021 less than a hundred cases have been reported in the literatures [3]. Teratoma occupying all or part of the bronchi has an even rarer occurrence, with only eight cases reported worldwide (Table 1) [5,8–14].

It has been speculated that IPTs arise from the thymic tissue of the third pharyngeal pouch during embryonic development and are usually diagnosed at a later stage of life due to their slow-growing nature [15]. IPTs often affect the upper lobes for reasons yet to be identified [1]. Symptoms most frequently reported include chest pain, cough, fever, dyspnea, hemoptysis, bronchiectasis, and rarely hair expectoration (Trichoptysis) as the result of bronchial communication with the teratoma, which can be the most IPT specific symptom. [16]. The symptoms depend on multiple factors, such as; location, tissue type, and mass size ranging from a few centimeters and up to 30 cm [17,18]. In the systematic review, amongst the 8 cases of endobronchial teratoma (EBT), it has been found that EBTs are mostly diagnosed at the 3 decade of life in the left upper lobe bronchus with a higher occurrence in males, just like the current case. Patients with EBT often present with chest pain, cough, and hemoptysis with occasional trichoptysis for a long period [8,11]. The current case has a cough, dyspnea, weight loss, and trichoptysis for five years.

Preoperative diagnosis of EBT can be pretty challenging as the symptoms are vague, trichoptysis might be the only symptom that can be diagnostic. However, it is a rare finding. Laboratory tests are usually within the normal range. The appearance of chest X-Ray findings is often of non-diagnostic value, sometime chest X-ray and CT scan show complications of bronchial obstruction [2]. Jamieson and the associate reported a 5-year-old female child presented with constitutional symptoms; preliminarily, she was diagnosed as a pulmonary tuberculosis case and treated accordingly for several months. Later, it became clear that it was a case of EBT as the hilar mass persisted after a complete cycle of anti-tuberculosis medications [10]. Haddad and colleagues managed their patient as a case of pneumonia who did not respond to medication; chest CT scan showed endobronchial mass mimicking hamartoma, bronchoscopy was inconclusive, intraoperative findings were consistent with a carcinoid tumor, later histopathological examination of the specimen revealed EBT [5]. The present case had destructed left lung revealed by chest X-ray and confirmed by CT scan. Hence complete diagnosis can only be made postoperatively via histopathological examination of the specimen [2].

Chest CT scan might give a hint regarding the diagnosis. It generally shows a smooth-walled mass which can contain soft tissues, fluids, fats, and calcifications, the mass is distinguished from mediastinal teratomas by peripheral translucency and the presence of air in the mass if it communicates with the bronchi. [19] The reviewed

studies of EBT showed that CT scan and fiberoptic bronchoscopy could be the optimal methods in the preoperative work up of the cases with suspected endobronchial teratoma [8,9]. The examination of the current case discovered crepitation and wheeze on the left side of the chest. Further investigations, including CT scan disclosed completely destroyed left lung with bronchiectasis and fibrotic change, and the result of histopathological examination of the mass confirmed the diagnosis of teratoma with prominent bone tissue.

The treatment of choice for both IPT and endobronchial teratoma is total excision to prevent life threatening complications and malignant transformation. Teratoma might contain pancreatic tissue secreting digestive enzymes which lead to erosion of the tumor wall and rapture into the nearby airways, this would be suggested when the patients have hemoptysis. IPT might be managed by segmentectomy or wedge resection while EBT necessitates either lobectomy or pneumonectomy [16,20]. The current case underwent pneumonectomy under general anesthesia.

Mature IPTs and EBT have a good prognosis with low-probability of malignancy, and have generally a good clinical outcome with a low reoccurrence rate [4,5,8,9,21]. This patient was hospitalized for three days postoperatively, then discharged in good health.

In conclusion, endobronchial teratoma is an exceedingly rare type of intrathoracic teratoma that mostly affects males and is usually diagnosed at the 3rd decade of life; lobectomy or pneumectomy are optimal managements for these cases.

Declaration of Competing Interest

There is no conflict to be declared.

Sources of funding

No source to be stated.

Ethical approval

Approval is not necessary for case report in our locality.

Consent

Written informed consent was obtained from the patient to publish this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author's contribution

Fahmi H. Kakamad: Surgeon performing the operation, follow up the patient, writing the manuscript and final approval of the manuscript.

Hawbash M. Rahim, Bestoon Kh. Salih: assisting in the writing the manuscript, final approval of the manuscript.

Karokh H. Salih: pathologist diagnosing the condition, final approval of the manuscript.

Abdulwahid M. salih, Shadi Hamid sidiq, Diyar A.Omar, Suhaib H. kakamad, Rawezh Q. Salih, Shvan H. Mohammed: major contribution to the idea, revision and final revision of the manuscript.

Registration of research studies

According to previous recommendation, registration is not required for case report.

Guarantor

Fahmi Hussein Kakamad is the Guarantor of submission.

Provenance and peer review

Not commissioned, externally peer-reviewed.

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