Case Report

A rare cause of airway obstruction: Mediastinal cyst secondarily infected with *Mycobacterium tuberculosis*

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ABSTRACT

Mediastinal cysts are mostly congenital, but rarely, infections and malignancies can cause cystic degeneration of enlarged mediastinal lymph nodes. Diagnosis is challenging as the presenting symptoms are nonspecific. Surgical resection is the reference modality both for diagnosis and management. Secondary infection of mediastinal bronchogenic cyst with *Mycobacterium tuberculosis* is rare. Herein, we describe a young male who was managed as bronchial asthma with inhalational bronchodilators and glucocorticoids. Computed tomography revealed a cystic lesion in the subcarinal region. Endobronchial ultrasound-guided transbronchial needle aspiration was done to perform diagnostic and therapeutic aspiration of the cyst that showed infection with *M. tuberculosis*. A subsequent surgical resection confirmed the cystic lesion to be a bronchogenic cyst.

KEY WORDS: Bronchogenic cyst, cyst, foregut cyst, mediastinum, tuberculosis

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INTRODUCTION

Mediastinal cysts are uncommon and account for 10%–18% of all mediastinal abnormalities.[1] Most of the mediastinal cysts are congenital, with bronchogenic cysts being the most common.[1] Other causes of mediastinal cyst include cystic degeneration of mediastinal lymph nodes due to malignancy or infection. The diagnosis of mediastinal cyst remains challenging due to nonspecific nature of the symptoms.[1,2] Most commonly, patients remain asymptomatic and become symptomatic if there is a rapid increase in the size of the cyst either spontaneously or due to secondary infection. The secondary infection is usually bacterial, although uncommonly *Mycobacterium tuberculosis* or nontuberculous mycobacteria can also infect the cyst. In such a scenario, it becomes difficult to determine the exact etiology of the cyst, whether congenital or cystic degeneration of the mediastinal lymph node. Herein, we report a case with a mediastinal cyst that proved to be a bronchogenic cyst secondarily infected with tuberculosis.

CASE REPORT

A 45-year-old gentleman presented with progressive breathlessness and noisy breathing of 2 months' duration. There was no fever, cough, chest pain, anorexia, or weight loss. He had no prior history of asthma, atopy, and smoking.

Access this article online

Quick Response Code:

Website: www.lungindia.com

DOI: 10.4103/lungindia.lungindia_445_17

How to cite this article: Chhabria BA, Agarwal R, Garg M, Gupta N, Bal A, Dhooria S, *et al*. A rare cause of airway obstruction: Mediastinal cyst secondarily infected with *Mycobacterium tuberculosis*. Lung India 2018;35:421-4.

On auscultation, there was bilateral diffuse expiratory wheeze. Spirometry revealed obstructive ventilatory defect with no significant bronchodilator reversibility [Table 1]. He was diagnosed as a case of bronchial asthma and was prescribed a combination of inhaled formoterol and budesonide. However, there was no relief in his symptoms after 6 weeks despite good compliance and correct inhaler technique.

On reevaluation, the chest radiograph revealed subtle mediastinal widening [Figure 1a]. Contrast-enhanced computed tomography (CECT) of thorax revealed a cystic lesion in the subcarinal location that was causing extraluminal compression of the airways at the level of carina and both main bronchi [Figure 1b]. Flexible bronchoscopy revealed a splayed carina, with dynamic airway collapse on expiration and narrowing of bilateral main stem bronchi. With a clinical suspicion of a congenital bronchogenic cyst, the patient was subjected to endobronchial ultrasound-guided transbronchial needle aspiration (EBUS-TBNA), and 50 mL of turbid pus-like material was aspirated. The aspirated material was sent for analysis, including Gram stain, bacterial culture, mycobacterial cultures, and cytological examination. There was significant resolution of symptoms and normalization of the spirometric lung functions following therapeutic aspiration of the cyst [Table 1]. CECT thorax showed a reduction in size of the cyst [Figure 1c]. Stain for acid-fast bacilli was positive [Figure 2] from the fluid with no evidence of bronchial epithelial cells. A diagnosis of tuberculous lymphadenitis with cystic degeneration was considered, and the patient was treated with anti-tuberculosis therapy for 6 months. After completing treatment, a follow-up CECT thorax showed significant resolution as compared to the baseline [Figure 1c]; however, the lesion did not disappear completely. The patient underwent surgical excision of the mediastinal cyst. The histopathological examination demonstrated ciliated columnar lining epithelium [Figure 3a] and surrounding mediastinal lymph nodes showing granulomatous inflammation [Figure 3b] that confirmed the diagnosis of bronchogenic cyst.

Table 1: Spirometry suggested severe obstruction which resolved following therapeutic aspiration

Parameters	At presentation	After therapeutic aspiration
FVC in L	3.52 (90)	3.82 (98)
FEV_1 in L	1.17 (38)	3.05 (98)
FEV_1/FVC	33.2	79.8
PEFR L/min	219 (43)	450.6 (89)
FEF 25-75 L/min	68.4 (33)	181 (88)

The percentage of predicted values of the spirometric variable is mentioned in parentheses. FEV_1: Forced expiratory volume in the first second, FVC: Forced vital capacity, PEFR: Peak expiratory flow rate, FEF 25–75: Forced expiratory flow rate between 25%–75% of FVC

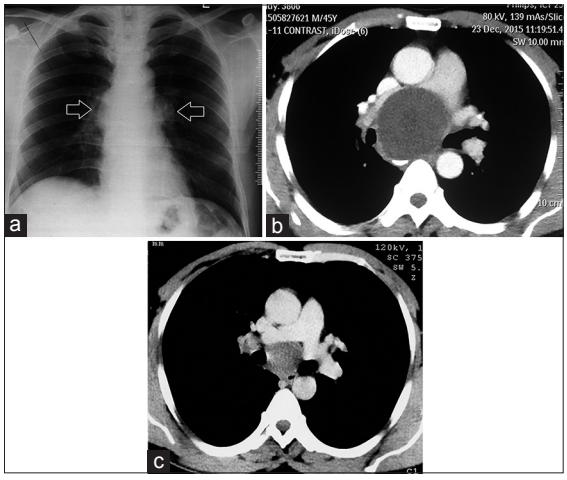

Figure 1: Chest radiograph demonstrating mediastinal widening (a); contrast-enhanced computed tomography of thorax showing a homogenous circumscribed hypodense lesion in the subcarinal region (b); contrast-enhanced computed tomography thorax at 6 months demonstrating a reduction in the size of the cyst after therapeutic aspiration with endobronchial ultrasound and antituberculosis therapy (c)

DISCUSSION

The index case highlights few important points. First, it can be difficult to distinguish a congenital mediastinal cyst from cystic degeneration of mediastinal lymph nodes. Second, an obstructive pattern on spirometry is not always asthma or chronic obstructive pulmonary disease.

Mediastinal cysts can have varied etiologies. Congenital foregut cysts are the most common, with bronchogenic cysts constituting 50% of all mediastinal cysts in adults.[1] Majority of these cysts localize in the anterior or middle mediastinum. When the anomaly associated with cyst formation is delayed, the cysts can also localize to the lung parenchyma and become susceptible to secondary infections.[1,2] About two-third of patients with mediastinal cyst remain asymptomatic; however, they may develop symptoms if the cyst increases in size or becomes

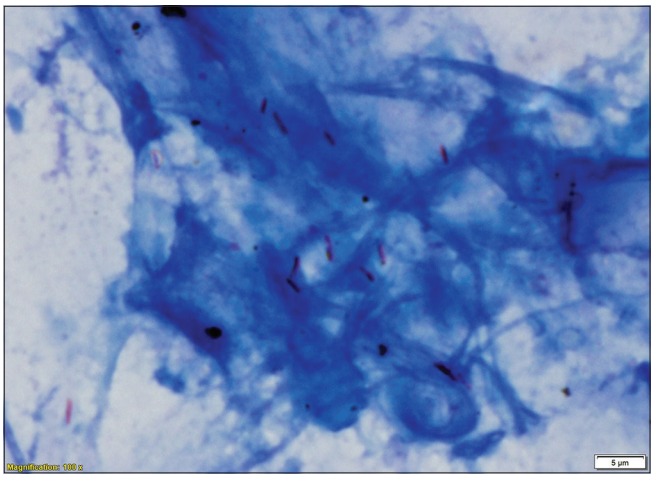

Figure 2: Photomicrograph of the cytology specimen obtained after aspirating the cystic contents using endobronchial ultrasound-guided transbronchial needle aspiration demonstrating multiple acid-fast bacilli in the background of necrosis

secondarily infected. Infection of cysts and related symptoms such as fever and chest pain are more common with cysts situated in the lungs in comparison to those in the mediastinum.[3,4] In addition, obstructive symptoms similar to the index case are usually reported in children owing to the narrow and pliable airway.[5] However, adults may present with symptoms of airway obstruction if the cyst is large and is situated around the tracheal bifurcation or is retrotracheal.[3] In symptomatic individuals with suspicious chest radiographs, CECT thorax or magnetic resonance imaging of thorax can confirm the presence of cysts and can delineate anatomy. EBUS-guided TBNA serves as a diagnostic as well as a therapeutic modality. The diagnosis of bronchogenic cyst can be confirmed by cytological examination of aspirated material that reveals the presence of mucus or bronchial epithelial cells without polymorphonuclear leukocytes or lymphocytes or malignant cells.[6] In the presence of a secondary infection, it is not possible to confirm or refute a diagnosis of a bronchogenic cyst only by aspiration. The persistence of the cystic lesion in the index case after treating for tuberculosis led us to suspect an underlying bronchogenic cyst. Histological examination of the surgically resected cyst confirmed the diagnosis.

Tuberculosis presenting as a primary mediastinal cystic lesion is extremely rare with only few reports [Table 2]. The possible mechanism of cyst formation due to tuberculosis *per se* involves conglomeration and extensive necrosis of affected tubercular lymph nodes or contiguous spread from bony structures (vertebrae).[7] To the best of our knowledge, secondary tuberculous infection of a mediastinal bronchogenic cyst has not been reported previously, although tuberculous involvement of intraparenchymal bronchogenic cysts has been reported [Table 3]. While intrapulmonary bronchogenic cyst can get secondarily infected with tuberculosis by contiguous spread from the lung parenchyma, in the case of a mediastinal cyst, the source of infection could be the surrounding mediastinal lymph nodes. In the index case, the surgical specimen revealed enlarged lymph nodes around the cysts which showed the presence of epithelioid granulomas on histological examination, thus indicating the potential source.

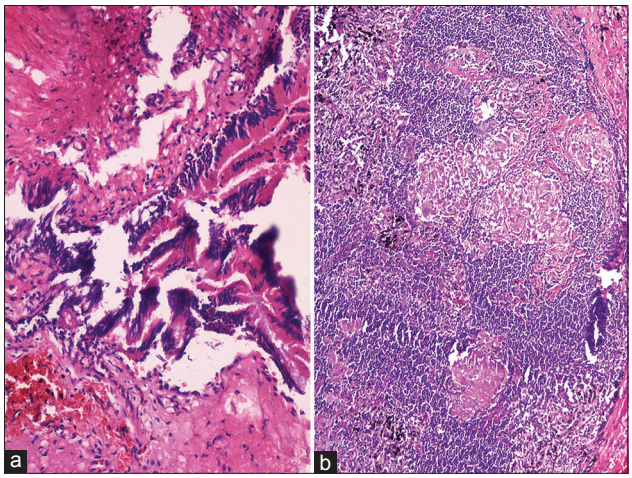

Figure 3: Photomicrographs from mediastinal mass showing bronchogenic cyst with ciliated columnar lining epithelium (a) and surrounding mediastinal lymph nodes showing granulomatous inflammation (b)

Table 2: Summary of previously reported cases of tuberculosis presenting as cystic lesions in the mediastinum

Author/year	Age (years)/sex	Location of cyst	Mode of diagnosis	Final diagnosis
Verma and Narayan[8] 2008	21/female	Anterior mediastinum	Guided percutaneous FNA	*Mycobacterium tuberculosis*
Sahin Yildiz[9] 2011	30/female	Middle mediastinum	Thoracotomy Surgical excision	*Mycobacterium tuberculosis*
Varik *et al*.,[10] 2012	0.4/male	Posterior mediastinum	Thoracotomy Surgical excision	*Mycobacterium tuberculosis*
Dhooria *et al*.,[7] 2015	14/male	Posterior mediastinum	EBUS-TBNA	*Mycobacterium tuberculosis*

FNA: Fine needle aspiration, EBUS-TBNA: Endobronchial ultrasound-guided transbronchial needle aspiration

Table 3: Summary of previously reported cases of bronchogenic cysts with secondary tubercular infection

Author	Number of patients	Patient profile	Location of cyst	Mode of diagnosis	Final diagnosis
Andrews *et al*.[11]	3	NA	Intraparenchymal	Bronchogram followed by surgical excision	*Mycobacterium tuberculosis*
Houser *et al*.[12]	1	9 years/female	Intraparenchymal (right lower lobe)	Right lower lobectomy	*Mycobacterium tuberculosis*
Lin *et al*.[13]	1	39 years/female	Intraparenchymal (right lower lobe)	VATS-assisted resection	*Mycobacterium avium*
Liman *et al*.[14]	2	a) 20 years/male b) 34 years/female	a) Intraparenchymal (right lower lobe) b) Horizontal fissure	Thoracotomy Surgical excision	*Mycobacterium tuberculosis*
Frye and Decou[15]	1	13 years/male	Intraparenchymal (right lower lobe)	Right lower lobectomy	*Atypical mycobacteria*
Chu *et al*.[16]	1	34 years/female	Intraparenchymal (left upper lobe)	Thoracotomy Surgical excision	*Mycobacterium tuberculosis*
Kwon *et al*.[17]	1	21 years/male	Intraparenchymal (right upper lobe)	Right upper lobectomy	*Mycobacterium avium*

NA: Not available, VATS: Video-assisted thoracoscopic surgery

CONCLUSION

Mediastinal cysts can masquerade obstructive airway disease. Mediastinal cysts associated with tuberculosis are rare; they could represent either a necrotic lymph node or a bronchogenic cyst with secondary infection. Surgical excision is definitive both in diagnosis and management of a bronchogenic cyst.

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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