Management of infected recurrent mediastinal bronchogenic cyst with tuberculous mediastinal lymphadenitis

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Abstract

We describe a case of a 20-year-old woman with recurrent bronchogenic cyst. The cyst made its debut at the age of six, which was excised surgically. Preoperative investigations revealed a mass in the right parasternal region extending from around the pericardium, present between the second and fourth ribs and abutting the right mediastinal structures. Dense adhesions were noted and released selectively to avoid iatrogenic injury. Complete evacuation and excision of the cyst from around the pericardium and phrenic nerve with marsupialization of the sac were performed. The patient's post-operative course was uneventful.

Keywords Bronchogenic cyst · Recurrent bronchogenic cyst · Infected bronchogenic cyst

Introduction

Bronchogenic cysts are a rare congenital anomaly, often a consequence of abnormalities in the developing tracheobronchial tree during the fetal development. Abnormal budding of the tracheobronchial tree is known to be the initiating cause of cystic formation, whose location is principally determined by the stage of budding. The cyst is likely to present in the mediastinum in early stages of malformation, which is most commonly observed, otherwise in the lung parenchyma at later stages [1].

Case report

A 20-year old female with a history of mediastinal cyst (details unavailable) excision at the age of 6 years through right anterolateral thoracotomy with a posterior extension presented

to us with complaints of atypical chest pain in the last 4 months. Chest X-ray showed large round opacity measuring 6.7×9.5 cm in the right parasternal region with regular borders, extending from the second to the fourth rib, abutting right atrium and superior caval vein. The lung fields were clean and there were no associated lung collapse or rib erosions (Fig. 1). Computed tomography revealed a large right-sided pericardial cyst (Fig. 1). She was taken up for surgery after routine preoperative evaluation.

The mass presenting near the midline prompted access to the mediastinum through a median sternotomy. Multiple enlarged mediastinal lymph nodes seen were excised for histopathological evaluation. A large thick-walled cyst measuring $8 \times 6 \times 6$ cm in size was found adherent to the right side of the pericardium posterior to and adhered to the phrenic nerve (Fig. 2). It was densely adhering to the right upper and middle lobe of the lung. Adhesions were released from the lower border and from the thoracic surface; however, dense adhesions with the middle and upper lobes could not be released, unless the lung tissue was peeled. Hence, the cyst was drained (Fig. 3) and the emanating pus was collected and sent for culture. Upon aerating the lungs, no communications were noted with the bronchus. A segment of the sac wall was sent for histopathological evaluation. Marsupialization of the sac wall was done (Fig. 4). The patient was shifted and extubated in the intensive care unit on the day of surgery and shifted to step down care a day later with uneventful recovery.

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Fig. 1 Bronchogenic cyst impression on chest X-ray and computed tomography slice

Results

The microbiology report of the pus inferred growth of *Stenotrophomonas maltophilia* which is an aerobic, non-

fermentative, gram-negative bacterium, for which, the patient was administered meropenem as per sensitivity pattern for a fortnight. Histopathologic examination revealed bronchogenic cyst lined by pseudo-stratified ciliated, columnar

Fig. 2 Intraoperative image showing bronchogenic cyst adherent to the right upper and middle lobe of the lung

Fig. 3 Intraoperative image of pus aspiration and incision drainage of the cyst

epithelium. The histopathology of the enlarged lymph nodes showed *tuberculous lymphadenitis*, which was treated with anti-tuberculosis therapy encompassing ethambutol hydrochloride 800 mg, isoniazid 300 mg, pyrazinamide 750 mg, and rifampicin 450 mg.

The patient was discharged a fortnight after surgery.

Discussion

Bronchogenic cysts are a rare congenital anomaly representing 12–30% of cysts in the mediastinum, while its subset of bronchogenic cyst accounts for 6–15% of primary mediastinal masses, often a consequence of abnormalities in the developing tracheobronchial tree during the fetal development. Abnormal budding of the tracheobronchial tree is known to be the initiating cause of cystic formation, whose location is principally determined by the stage of budding. The cyst is likely to present in the mediastinum

in early stages of malformation, which is most commonly observed, otherwise in the lung parenchyma at later stages [1].

The cyst presentations are even rare in adults and in most presentations, the cysts are asymptomatic and found incidentally. However, the cyst may cause compression upon the developing tracheobronchial tree in the pediatric group, soliciting early medical aid. The presentation includes cough, chest pain, fever, breathlessness, or atypical chest pain, as seen in our case. Although most patients are asymptomatic for a long time, they may present at a later age for what might be cyst-related sequelae, which include compression of the adjacent structures due to cyst enlargement, infection, rupture, hemorrhage, or malignant transformation [2, 3].

Cysts located in the lung are more prone to infection, as against their mediastinal counterparts owing to their exposition/communication to the tracheobronchial tree [1].

The unique and rareness of this presentation go by the mediastinal bronchogenic cyst in our patient, with no communication to the adjacent structures, yet turning infections possibly through a hematogenous route. Organisms such as *pneumococcus*, *peptococcus*, *E. coli*, *H. influenzae*, *Mycobacterium kansasii*, and *avium complex* have been reported to cause infections in the domain of bronchogenic cysts [1].

The cyst appears ovoid or spherical in chest roentgenograms and computerized tomography with clear margins and air fluid levels that can be noted in the former should the cyst have any tracheobronchial connections. Examination of the cut sections under microscope, the cyst wall has a lining of pseudo-stratified ciliated, columnar epithelium with some fibroconnective tissue which includes cartilage, glands, and smooth muscles [4, 5].

Surgical intervention can be made appropriately by either modes: thoracotomy, sternotomy, or with video-assisted surgery, relative to the location of the lesion. The management of bronchogenic cysts is disputed for the existence of multimodal treatment options, ranging from simpler regular follow-up to

Fig. 4 Intraoperative image of marsupialization of bronchogenic cyst wall

percutaneous intervention or surgical excision. Follow-up studies of the asymptomatic patients suggest the eventual necessitated surgical intervention, as sequelae of the cyst, mentioned above, make the patient symptomatic [6]. Percutaneous intervention for the cyst does not yield good results in the long term since it carries a high rate of recurrences and morbidity; hence, complete surgical extirpation is the preferred option for the expeditious treatment [7].

With the high incidence of tuberculosis in Oriental countries, bronchogenic cysts although rare [8, 9] may be incidentally reported as a consequence of concurrent tuberculosis or misdiagnosed as tuberculosis. Should the microbial analysis from the associated nine regional lymph nodes prove gram-positive for *Mycobacterium tuberculosis*, the patient is treated prophylactically.

Conclusion

Recurrent bronchogenic cysts have been reported sporadically in literature [10–12] and carry significant morbidity for the patient. Hence, complete surgical excision and extirpation should be the proffered mode of management, laying a good foundation for achieving not only accurate diagnosis and treatment but also good long-term outcomes.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval Obtained.

Informed consent Informed consent was obtained from all individual participants included in the study.

No animals were harmed in the context of this study.

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