

Bronchogenic Cyst Presenting as Mediastinal Mass with Pleural Effusion and Unusual Bronchoscopic Findings: A Case Report and Review of Literature

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Abstract

Bronchogenic cyst is a benign congenital developmental abnormality. It is most commonly located in middle mediastinum. Its association with pleural effusion is rare. Sometimes it may present with a fixed compression over trachea or oesophagus. We report the rare occurrence of bronchogenic cyst in antero-superior mediastinum mimicking a mass lesion with right-sided pleural effusion and tracheal compression only during expiration in a 39-year-old male. [Indian J Chest Dis Allied Sci 2017;59:81-85]

Key words: Bronchogenic cyst, Bronchoscopy, Surgery, Pleural effusion.

Introduction

Bronchogenic cyst is a benign congenital developmental abnormality of the embryonic foregut. It is formed as a result of an accessory lung bud becoming isolated from the rest of the tracheo-bronchial tree. Bronchogenic cyst can occur in the mediastinum or may be intra-pulmonary. It is usually a solitary extra-pulmonary cyst found most commonly in the middle mediastinum.¹ Radiographically these appear as spherical or oval masses with smooth outlines. On computed tomography (CT), fluid in these cysts has an average density of zero Hounsfield units. However, the CT density may sometimes be higher, comparable to that of soft tissue.² These are rarely associated with pleural effusion.³ The usual bronchoscopic findings is compression of trachea and/or oesophagus.^{4,5} We report the rare occurrence of bronchogenic cyst mimicking a solid tumour with pleural effusion, with unusual bronchoscopic findings in the form of tracheal compression only during expiration.

Case Report

A 39-year-old male, non-smoker, was admitted with chief complaints of cough with mucopurulent expectoration for the last 15 days and low-grade fever for the last 10 days. He also gave history of having similar episodes repeatedly since childhood which improved with treatment. He had taken anti-

tuberculosis treatment (ATT) for six months in 2003 based on chest radiograph finding suggestive of right-sided paratracheal lymphadenopathy. However, the patient did not improve and signs and symptoms suggestive of lower respiratory tract infection had persisted.

The general physical examination was unremarkable. Respiratory system examination revealed pleural effusion on the right side. Chest radiograph (postero-anterior view) showed a homogeneous opacity in the right paratracheal region of approximately 8cm × 8cm in size with mild pleural effusion on the right side (Figure 1).

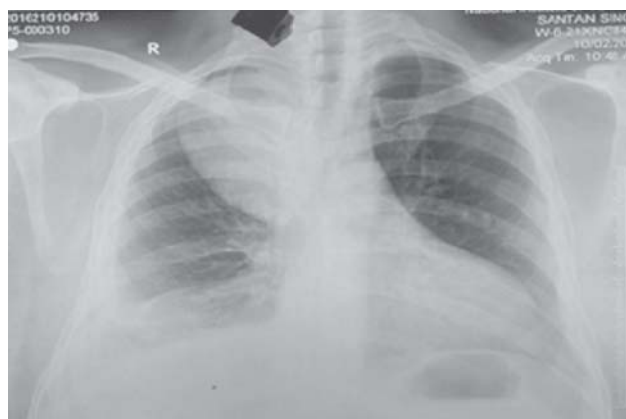


Figure 1. Chest radiograph (postero-anterior view) showing a homogeneous opacity in the right paratracheal region (8cm × 8cm) with mild pleural effusion on the right side.

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collection of histiocytic cells and foamy macrophages. Section from fatty mass revealed clumps of adipocytes. Fluid cytology showed blood and few ciliated columnar cells. No malignant cells were seen. A final diagnosis of bronchogenic cyst was made.

Summary of published case reports and case series of bronchoscopic findings in patients with bronchogenic cyst is presented in the table below.

Discussion

The term 'bronchogenic cyst' is often used synonymously with 'bronchial cyst' for foregut duplications that are clearly related to the airways, especially the trachea and the main bronchi. It is the most common form of mediastinal cysts and accounts for 36% of mediastinal cysts in all ages and 53% in paediatric age group.⁶ The incidence of mediastinal cysts is equal between both the genders whereas intra-pulmonary cysts are reported to have a male predilection.⁷

Table. Some of the case reports and case series documenting bronchoscopic findings in patients with bronchogenic cysts

Author	Type of Study	Year of Publication	No. of Cases	No. of Cases for Which FOB was Done	Modality Used for Diagnosis	Bronchoscopic Findings
Sarper <i>et al</i> ²	Case Series	2003	22	22	Histopathology	5 had extrinsic compression of the tracheobronchial tree 2 had purulent material seen coming from the upper lobe 1 had direct communication from a residual bronchogenic cyst to the left upper bronchus and blood clots were seen coming from the cyst
Suen <i>et al</i> ¹⁷	Case Series	1993	42	26	37 based on Histopathology 5 based on Clinico-radiologically	Abnormalities were found in 9 patients 7 had extrinsic compression of the tracheobronchial tree 1 had blood-tinged material, seen coming from the middle and lower lobes 1 had direct communication to the bronchus intermedius from a cyst
Loren <i>et al</i> ¹⁸	Case Series	1989	26	4	Histopathology	Abnormality was found in 1 patient but details of the findings were not mentioned
Kim <i>et al</i> ¹⁹	Case Report	2005	1	1	Histopathology	Peduncular polyp about 2 cm in length, originating from the anterior segment of right upper lung
Ohba <i>et al</i> ²⁰	Case Report	2014	1	1	Histopathology	Smooth protrusion of antero-lateral wall of the trachea, and the tracheal lumen was severely occluded
Sarkar <i>et al</i> ²¹	Case Report	2013	1	1	Clinico-radiologically	Mucosal thickening and mild narrowing of lumen of right intermediate bronchus due to extrinsic compression
Okabayashi <i>et al</i> ²²	Case Report	2002	1	1	Histopathology	Erosive mucosal lesion overlying the area of extrinsic compression at the right mainstem bronchus
Mampilly <i>et al</i> ²³	Case Report	2005	1	1	Histopathology	Bulging of the posterior wall of left bronchus into the lumen, which was filled with granulation tissue
Yohena <i>et al</i> ²⁵	Case Report	2005	1	1	Histopathology	Stenosis of the entrance of the left B6 segmental bronchus without mucinous discharge

Bronchogenic cysts can occur in the mediastinum or be intra-pulmonary. The mediastinal cysts are classified into five groups: para-tracheal, carinal, hilar, para-esophageal and miscellaneous.⁸ Most common location is the middle mediastinum (65%-90%).¹ Parenchymal (intra-pulmonary) cysts are typically perihilar and have predilection for lower lobes.⁹ Other uncommon locations include neck, cutaneous, pericardium extending across the diaphragm and appearing dumb-bell shaped and retroperitoneal.^{1,10,11} In our case the cyst was located in the antero-superior mediastinum which is a rare occurrence.

The majority of children with intra-thoracic foregut cysts are symptomatic and present with dyspnoea, stridor or persistent cough.¹² However, in adults symptoms are present in only about one-third of the patients.⁶ In our case also, patient had symptoms of cough and recurrent lower respiratory tract infection since childhood. Studies have reported differences in symptomology varying from 30%-90%.^{5,13-15}

Bronchogenic cysts are rarely associated with pleural effusion. To the best of our knowledge, till date only three cases have been reported.^{3,16} Cause of associated pleural effusion is thought to be probably due to inflammation of the pleura because of the huge cyst size. Khalil *et al*³ reported two cases of bronchogenic cyst with effusion and had mentioned about difficulties in surgical excision of the cyst. Similarly in our case also cyst excision was difficult.

Various studies have described bronchoscopic findings in bronchogenic cysts.^{2,4,17,18} Individual case reports have also described various bronchoscopy findings like endobronchial mass/protrusion, mucosal thickening/erosion, extrinsic compression and stenosis.¹⁹⁻²⁵ In one report⁴ fiberoptic bronchoscopy in seven patients with bronchogenic cyst showed bulging of the tracheal wall with no specific sign. In another study¹⁷ fiberoptic bronchoscopy in 26 cases with bronchogenic cyst revealed abnormalities in nine cases predominantly in the form of extrinsic compression. Bronchoscopic findings were very unusual in our case. Tracheal compression was present only during expiration and disappeared during inspiration. This probably contributed for the absence of compressive symptoms even though the size of cyst was 8.69cm × 7.61cm. To the best of our knowledge, such bronchoscopic findings has never been described in the previously reported cases.

To conclude, our case highlights several rare features including the presence of associated right-sided pleural effusion, bronchoscopic finding of tracheal compression present only during expiration and the location of the bronchogenic cyst in the antero-superior mediastinum.

References

1. Josep M, Castellote A. Pulmonary malformation beyond neonatal period. In: Lucaya J, Baert AL, Strife JL, editors *Pediatric Chest Imaging, Chest Imaging in Infants and Children*. New York: Springer 2007;p.93-112.
2. Sarper A, Ayten A, Golbasi I, Demircan A, Isin E. Bronchogenic cyst. *Texas Heart Institute* 2003;30:105-8.
3. Khalil A, Carette MF, Milleron B, Grivaux M, Bigot JM. Bronchogenic cyst presenting as mediastinal mass with pleural effusion. *Eur Respir J* 1995;8:2185-7.
4. Ribet ME, Copin MC, Gosselin B. Bronchogenic cysts of the mediastinum. *J Thorac Cardiovasc Surg* 1995;109:1003-10.
5. Cuypers P, De Leyn P, Cappelle L, Verougstraete L, Demedts M, Deneffe G. Bronchogenic cysts: a review of 20 cases. *Eur J Cardiothorac Surg* 1996;10:393-6.
6. Neel R, Malcolm M. Congenital cysts of the mediastinum: bronchopulmonary foregut abnormalities. In: Fishman P, editor *Fishman's Pulmonary Diseases and Disorders*; 5th edition. USA: McGraw-Hill Education; 2015;p.2823.
7. Staatz G, Honnef D, Piroth W et-al. *Pediatric Imaging*. George ThiemeVerlag. 2007.
8. Maier HC. Bronchiogenic cysts of the mediastinum. *Ann Surg* 1948;127:476-502.
9. Yoon YC, Lee KS, Kim TS, Kim J, Shim YM, Han J. Intrapulmonary bronchogenic cyst: CT and pathologic findings in five adult patients. *AJR Am J Roentgenol* 2002;179:167-70.
10. Onol FF, Baytekin F, Dikbas O, Ergonenc T, Tanidir Y. A retroperitoneal bronchogenic cyst mimicking adrenal tumour in an adult: is differential diagnosis truly possible? *J Clin Pathol* 2009;62:187-9.
11. Pradeep KE. Cutaneous bronchogenic cyst: an under-recognised clinicopathological entity. *J Clin Pathol* 2009;62:384.
12. Eraklis AJ, Griscom NT, McGovern JB. Bronchogenic cysts of the mediastinum in infancy. *N Engl J Med* 1969;281:1150-54.
13. Aktogu S, Yuncu G, Halilcolar H, Ermete S, Buduneli T. Bronchogenic cysts: clinicopathological presentation and treatment. *Eur Respir J* 1996;9:2017-21.
14. Sirivella S, Ford WB, Zikria EA, Miller WH, Samadani SR, Sullivan ME. Foregut cysts of the mediastinum: results in 20 consecutive surgically treated cases. *J Thorac Cardiovasc Surg* 1985;90:776-82.
15. St-Georges R, Deslauriers J, Duranceau A, Vaillancourt R, Deschamps C, Beauchamp G, et al. Clinical spectrum of bronchogenic cysts of the mediastinum and lung in the adult. *Ann Thorac Surg* 1991;52:6-13.
16. Zaman MU. Intact bronchogenic cyst presenting as a lung mass provoking a pleural effusion: a rare presentation. *W V Med J* 2012;108:12-15.
17. Suen HC, Mathisen DJ. Surgical management and radiological characteristics of bronchogenic cysts. *Ann Thorac Surg* 1993;55:476-81.
18. Di Lorenzo M, Collin PP, Vaillancourt R, Duranceau A. Bronchogenic cysts. *J Pediatr Surg* 1989;24:988-91.

19. Kim JH, Jang AS, Park JS, Lee JH, Park SW, Koh ES, *et al.* Polypoid endobronchial lung cyst with bronchoscopic removal: a case report. *J Korean Med Sci* 2005;20:892-4.
20. Ohba G, Toma M, Komori K, Hirobe S, Fukuzawa R. Intramural tracheal bronchogenic cyst: a case report. *Springer Plus* 2014;3:262.
21. Sarkar A, Pandit N. Bronchogenic cyst: a case report. *Int J Case Rep Images* 2013;4:731-4.
22. Okabayashi K, Motohiro A, Ueda H, Ondo K, Kawahara K, Shirakusa T. Subcarinal bronchogenic cyst with high carbohydrate antigen 19-9 production. *Japanese J Thorac Cardiovasc Surg* 2002;50:46-8.
23. Mampilly T, Kurian R, Shenai A. Bronchogenic cyst: cause of refractory wheezing in infancy. *Indian J Pediatr* 2005;72:363-4.
24. Mazzei JA, Barro A, Mazzei ME, Portas T, Esteva H. Biphasic flow volume curve due to obstruction of main bronchus by bronchogenic cyst. *Respir Med* 2011;4:116-8.
25. Yohena T, Kuniyoshi M, Kono T, Uehara T, Miyahira T, Kawasaki H, *et al.* Novel approach for a pulmonary bronchogenic cyst: a report of a case. *Ann Thorac Cardiovasc Surg* 2005;11:249-51.