

Bronchogenic Cyst in an Infant

DKK NG, AKW LAW, WF LAU, PKH TAM

Abstract

We report a case of bronchogenic cyst incidentally identified from a chest radiograph in a child with roseola infantum. This is not a common congenital malformation. Age of presentation can be very variable. Mediastinal cyst accounts for the majority of cases. Clinically it can be totally asymptomatic or it may cause severe respiratory distress especially in young infants. Chest radiograph and CT scan thorax are the mainstay of investigation.

Key words

Bronchogenic cyst; Infant

Introduction

Bronchogenic cyst is a rare congenital pulmonary anomaly.^{1,2} It is uncommon but the incidence in Hong Kong is unknown. Asymptomatic cyst may not present until complications arise or it may be diagnosed incidentally from routine health check. We report here a case of bronchogenic cyst identified from a chest radiograph taken for an infant. Diagnosis and management of this congenital problem are also discussed.

Case Report

WKF was admitted to our hospital at the age of eight months because of high fever, cough, runny nose, poor

feeding and vomiting for a day. Temperature was up to 40°C. He was born at full term and the neonatal period was uneventful. Immunization status was up-to-date and his past health was good. He was the only child of a non-consanguineous marriage. Family history of lung diseases or congenital lung malformation was absent. Physical examination was unremarkable except for mild dehydration and a mildly congested right eardrum. Chest examination showed symmetrical expansion and normal vesicular breath sound. Provisional diagnosis was viral upper respiratory tract infection. Complete blood picture showed a normal total white cell count. Chest radiograph (Figure 1) demonstrated a well-delineated circular opacity with smooth outline over the right upper zone. Its medial border could not be defined from that of mediastinum. Symptomatic treatment including paracetamol and chlorpheniramine was given. Intravenous fluid supplement was provided for his poor feeding and dehydration. Fever subsided two days later with the appearance of generalized maculopapular rash. The clinical diagnosis was roseola infantum and he recovered uneventfully.

Subsequent CT scan thorax (Figure 2) showed a well-defined middle mediastinal homogenous lesion with soft tissue attenuation of 21 H.U., located above and adjacent to right hilar region. The lesion attenuation was less than the surrounding soft tissue. It was internally homogenous and it did not have a well-defined thin wall. It was compatible with bronchogenic cyst. Flexible bronchoscopy revealed no obstruction in the tracheobronchial tree. Follow

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Figure 1 Chest radiograph showing a well defined roundish opacity (arrow) projected over right upper zone and a well preserved lateral mediastinal margin.



Figure 2 Computed tomography of thorax showing a well defined, low density and homogenous lesion (arrow) seen above and adjacent to right hilar region.

up chest radiograph six months later showed increase in size of the cyst. The child remained asymptomatic and his parents were unwilling to have surgical removal at that time. He was thriving well and there was no respiratory complication. At the age of 30-month, his father agreed to surgery. Right thoracotomy was performed. The cyst (Figure 3) was identified. Part of the cyst wall was adhering to right upper lobe probably due to previous subclinical infection. Segmental resection of right upper lobe was required in addition to excision of cyst. Both pieces of lung tissue showed marked congestion and focal inflammation. The child recovered well from operation without complication.

Discussion

Our patient had an asymptomatic mediastinal bronchogenic cyst. Incidental finding in chest radiograph of bronchogenic cyst is not an uncommon presentation.^{3,4} Neurenteric cysts, enterogenous cyst, neural tumour, teratoma, thymoma, cystic hygroma and lymphoma are differential diagnoses of middle mediastinal mass.³ Other rarities include pericardial cyst, intrathoracic meningocele and hiatus hernia. Mediastinal cyst constitutes about 65% of all cases of bronchogenic cyst and is centrally located in regions like carina, hilum, paratracheal and paraoesophageal space. Very rarely, bronchogenic cysts occur in cutaneous tissue, subcutaneous tissue,⁵ neck, pericardium, diaphragm and abdomen.

During the fetal period, the respiratory tract is an outgrowth of the pharyngeal portion of foregut.⁶ Bronchogenic cysts are actually aberrant budding from ventral diverticulum in the third trimester of gestation. Early abnormal budding at the level of carina results in mediastinal cysts whilst late budding at distal tracheobronchial tree ends up with intraparenchymal cysts. Histologically, bronchogenic cysts are lined with ciliated, mucus secreting respiratory columnar epithelium.⁶ Cartilage and bronchial glands can be found in the fibrous wall. Alveolar sacs are absent. Usually, there is no communication with the tracheobronchial tree. If a pathway does exist, the cyst may contain air and pus as well.

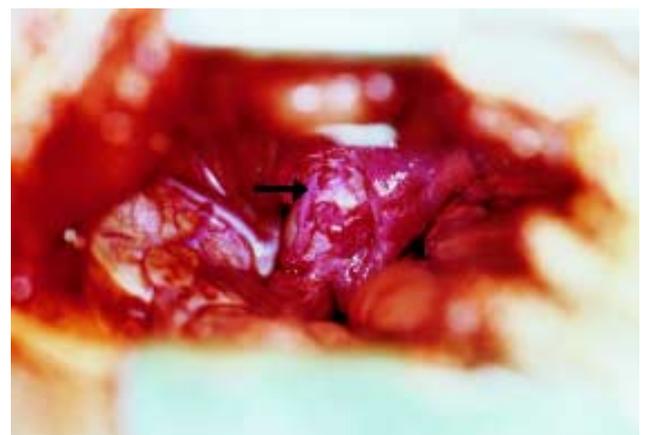


Figure 3 Infected bronchogenic cyst (arrow) at time of surgery.

Bronchogenic cyst is usually single, unilocular and spherical. Its size is usually less than ten centimeter in diameter⁴ and it commonly occurs on the right side. Associated anomalies include laryngeal cyst, congenital bronchomalacia and accessory left upper lobe.⁶ It is often clinically silent³ and is found in between the trachea and oesophagus. It was reported that a small bronchogenic cyst caused acute respiratory distress in a term neonate just one hour after birth.⁷ Reported complications included superior vena cava syndrome, bronchial atresia, pericardial compression, arrhythmia, left atrial compression, pulmonary artery compression or stenosis, pneumothorax, pleural effusion⁸ and giant tension bronchogenic cyst. There also exists a possibility of malignant degeneration. In view of all these potential problems, early operation of the cyst was usually suggested.⁹

Bronchogenic cyst is usually suspected from the appearance in chest radiograph. Characteristically, bronchogenic cyst is round or oval in shape. A well-marginated shadow of fluid attenuation in middle mediastinum is quite suggestive.⁴ Occasionally, an air fluid level or peripheral calcification can be found. Intraparenchymal cyst occurs in any part of the lung field. Contrast CT scan with selected thin slices through the region of interest affords detailed delineation of the relationship of the cyst to the surrounding structures. These anatomical details are important for the pre-operative surgical planning. Magnetic resonance imaging is also potentially useful for differentiating problematic soft-tissue-attenuation cysts from mediastinal neoplasia. Prenatal diagnosis with ultrasound scan was also reported.⁴ However, definitive tissue diagnosis can be established only by surgical excision.

Surgery should be the treatment of choice even when the cyst is asymptomatic.^{9,10} There will be dense pericyclic adhesions to adjacent structures at times of acute infection. Frequency of operative difficulty in symptomatic cases is thus higher when compared with asymptomatic cases.¹¹ Simple excision is all that is required for mediastinal cyst but an intraparenchymal cyst may require segmentectomy

or lobectomy as well. All lesions are approached via thoracotomy. In our patient, the cyst was infected at the time of surgery although the child was clinically asymptomatic. The operation would be much more difficult should the cyst be severely infected. Recurrence may occur if excision is incomplete. Post-operative complications are rare. Reported problems include pneumonia and transient paresis of right phrenic nerve.

In summary, congenital malformations of lung are relatively rare. Circular and homogenous mediastinal shadow in a chest radiograph should prompt one to include bronchogenic cyst in the differential diagnosis. Early surgery is technically easier and can help to prevent potential complications that may occur later in life.

References

1. Kravitz RM. Congenital malformations of the lung. *Pediatr Clin North Am* 1994;41:453-72.
2. Defossez SM, DeLuca SA. Bronchogenic cysts. *Am Fam Physician* 1989;39:129-32.
3. Jeffries JM 3rd. Asymptomatic bronchogenic cyst of the mediastinum. *Postgrad Med* 1987;81:235-40.
4. McAdams HP, Kirejczyk WM, Rosado-de-Christenson ML, Matsumoto S. Bronchogenic cyst: imaging features with clinical and histopathologic correlation. *Radiology* 2000;217:441-6.
5. Bagwell CE, Schiffman RJ. Subcutaneous bronchogenic cysts. *J Pediatr Surg* 1988;23:993-5.
6. Fowler CL, Pokorny WJ, Wagner ML, Kessler MS. Review of bronchopulmonary foregut malformations. *J Pediatr Surg* 1988; 23:793-7.
7. Dale PJ, Shaw NJ. Bronchogenic cyst presenting in the neonatal period. *Acta Paediatr* 1994;83:1102-3.
8. 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.
9. Cioffi U, Bonavina L, De Simone M, et al. Presentation and surgical management of bronchogenic and esophageal duplication cysts in adults. *Chest* 1998;113:1492-6.
10. Di Lorenzo M, Collin PP, Vaillancourt R, Duranceau A. Bronchogenic cyst. *J Pediatr Surg* 1989;24:988-91.
11. Aktogu S, Yuncu G, Halilcolar H, Ermete S, Buduneli T. Bronchogenic cysts: clinicopathological presentation and treatment. *Eur Respir J* 1996;9:2017-21.