CASE REPORT

# Infected Bronchogenic Cyst with *Mycobacterium tuberculosis* as a Cause of Respiratory Distress: A Case Report

*Ezekiel Olayiwola Ogunleye*[1], *Oyebola Olubodun Adekola*[2], *Olugbenga Oluseyi Olusoji*[1], *Augustine Jeremai Olugbemi*[1], *Saheed Babatunde Sanni*[1]

[1]Department of Surgery, Cardiothoracic Surgical Unit, College of Medicine, University of Lagos, Lagos, Nigeria

[2]Department of Anaesthesia, College of Medicine, University of Lagos, Lagos, Nigeria

***Correspondence to:*** Saheed Babatunde Sanni; email sannisaheed@yahoo.com

Received: 20 Nov 2020; Revised: 12 Dec 2021; Accepted: 27 Dec 2021; Available online: 22 Jan 2022

**Summary**

Bronchogenic cyst is a common primary cyst of the mediastinum, and it is usually located in the middle mediastinum and intrapulmonary regions. Bronchogenic cysts are lesions of congenital origin that occur due to abnormal budding from the primitive ventral foregut. They are common in the pediatric age group and have symptoms of respiratory distress, recurrent cough, wheezing, and stridor. Definitive management involves surgical excision. We herein report our experience of a bronchogenic cyst complicated by *Mycobacterium tuberculosis* infection in an 8-month-old infant. The patient underwent a right posterolateral thoracotomy, and an intrapulmonary bronchogenic cyst filled with purulent fluid was excised. The patient was given anti-tuberculous medication, and the post-operative outcome was good.

**Keywords:** Bronchogenic cysts, *Mycobacterium tuberculosis*, Respiratory distress

Ann Afr Surg. 2022; 19(3): 161-164
**DOI**: http://dx.doi.org/10.4314/aas.v19i3.6

**Funding:** None



## Introduction

Bronchogenic cyst is a congenital bronchopulmonary malformation resulting from abnormal ventral budding of the tracheobronchial tree during embryogenesis. It usually occurs between 26 and 40 days of gestation, with subsequent separation from the normally developing bronchi by 16 weeks of gestation. Early separation before 12 weeks leads to a mediastinal location of the cyst, whereas delayed separation results in a more peripheral location such as intrapulmonary parenchyma (1).

The epithelial lining of the cyst is usually pseudo-stratified columnar epithelium, and the wall consists of smooth muscle, cartilage, elastic tissue, and mucous glands. Bronchogenic cysts are usually unilocular and contain clear fluids devoid of air since they do not communicate with the tracheobronchial lumen. Infection of the cyst may cause the liquid content to become purulent. The cysts are mostly located in the mediastinum and lung parenchyma, but rarely in other locations (2).

Data on the management of an infected bronchogenic cyst with *Mycobacterium tuberculosis* are very sparse in our setting. We, therefore, report the case of an 8-month-old child with an intrapulmonary bronchogenic cyst that was infected with *M. tuberculosis*. The cyst was

successfully managed by surgical excision and post-operative anti-tuberculous medication.

**Case presentation**

An 8-month-old female infant was brought to our facility with a 6-week history of respiratory distress with increasing severity and associated cough, wheezing, and stridor. Fever and weight loss were absent, and there was no documented contact with anybody with infective cough or tuberculosis.

Previous trials of antibiotics and inhaled bronchodilator for presumed bronchopneumonia and asthma, respectively, were unsuccessful. The infant was born in the UK after a 9-month pregnancy, and delivery was uneventful. She was brought to Nigeria at the age of 4 months in good health, but she developed respiratory symptoms after 10 weeks of residing in Nigeria.

She was not vaccinated at birth against tuberculosis, but she was exclusively breastfed for 6 months, and her developmental milestones were normal for her age. She is the first child of well-educated parents who reside in the UK. Clinical examination revealed a well-nourished infant who was obese (>90th percentile compared with standard weight for age) and in severe respiratory distress, with expiratory stridor and wheezing. There were paroxysms of cough punctuating the clinical examination. Temperature was normal, with mild to moderate pallor. The respiratory rate was 88 cycles per minute, with moderately reduced air entry in the middle and lower zones of the right hemithorax. Only the first and second heart sounds were present, with mild tachycardia. All other systems were essentially normal. Her weight was 10 kg (>90$^{th}$ percentile), serum chemistry was normal, hematological profile showed mild leukocytosis (14,000/µL), with lymphocytic predominance, and increased erythrocyte sedimentation rate; the retroviral status was negative.

Chest x-ray (Figure 1) showed a well-defined oval-shaped homogenous opacity in the right mid-zone extending from the cardiac border to the chest wall laterally.

Computerized tomography scan of the chest (Figure 2) showed an intrapulmonary location of the lesion attached to the second carina on the right side. The lesion was 10×12 cm, and the mass appeared isodense to adjacent soft tissues, non-fatty, and non-calcified with attenuation of 40 HU. There was no significant enhancement of the mass on administration of contrast.

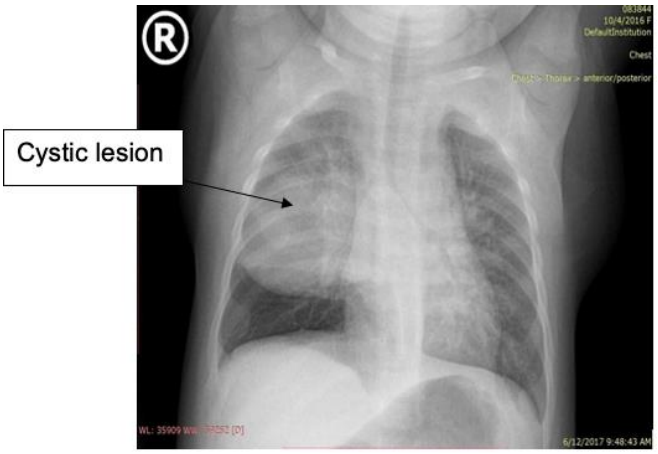


Figure 1. Pre-operative chest X-ray: anteroposterior view.

The mass was oblong-shaped and homogenously hypodense with a smooth, well-defined wall.

A diagnosis of bronchogenic cyst located within the right lung was made. Emergency right posterolateral thoracotomy was performed.

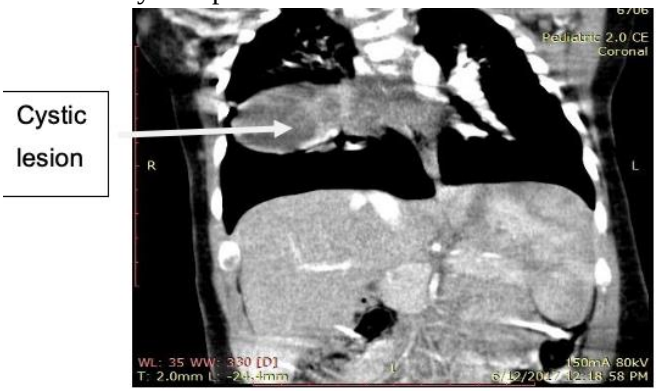


Figure 2. Pre-operative chest computerized tomography scan.

Findings include a large cystic mass, containing purulent fluid, attached to the tracheobronchial tree at the division of the right main bronchus to the right upper lobe bronchus and bronchus intermedius and compression atelectasis of the middle and lower lobes and a solitary lobar node. The mass was excised and sent for histopathology. The fluid aspirated from the cystic mass was sent for culture and sensitivity. The atelectatic middle and lower lobes were inflated through the endotracheal tubes with good lung expansion.

Thoracotomy closure was done, and patient was transferred to the intensive care unit for elective ventilation for 12 hours with chest tube in situ. Medications include intravenous antibiotics and analgesics.

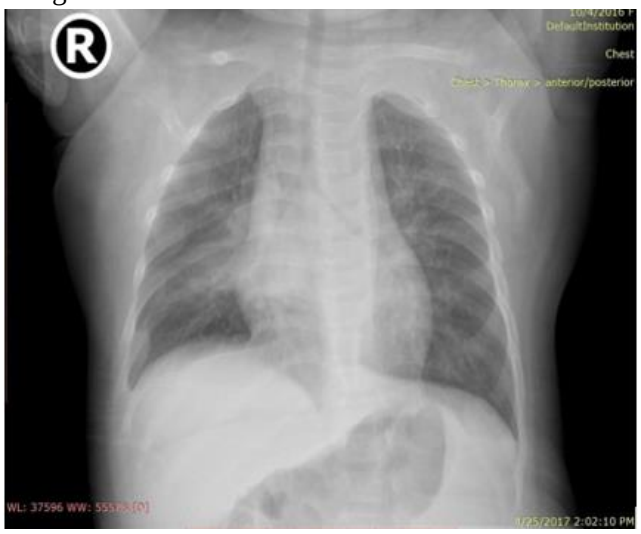


Figure 3. Post-operative chest X-ray.

The post-operative chest x-ray showed adequate lung expansion, and she was subsequently weaned off ventilation at 12 hours post-operatively. However, tachypnea persisted. Wound inspection on the 5th post-operative day showed complete wound dehiscence from the skin to the costal wall. The cystic fluid culture revealed no growth. Histopathology of the cyst wall showed massive infiltration by caseating granulomatous lesion indicative of *Mycobacterium* infection. Screening for tuberculosis was not done, as it was not expected.

Daily wound dressing was commenced, along with initiation of anti-tuberculosis therapy following the histopathology result. The chest tube was removed when the daily output was insignificant and there was radiological and clinical evidence of lung expansion on the 6th post-operative day.

The mother requested for a transfer to the UK, where she resides for continued care, as well as for a pathology cell block of the specimen for confirmation of the histopathology report.

The repeat histopathology in the UK also confirmed tuberculosis. The patient has since been improving steadily on anti-tuberculous medication. The dehisced post-operative wound has closed completely with weight gain. The patient's cough and other respiratory symptoms have abated.

**Discussion**

Bronchogenic cysts, which are commonly in the middle mediastinum or within the lung parenchyma, result from abnormal budding from the tracheal diverticulum of the foregut during early weeks of gestation. They are usually in contact with the tracheobronchial tree. They have been described in more remote locations, including the interatrial septum, neck, and abdomen, including the retroperitoneal space (3).

In our index case, the bronchogenic cyst was located within the lung parenchyma in the oblique fissure and attached to the division of the right main bronchus into the upper lobe bronchus and bronchus intermedius (Figure 2).

They are usually unilocular and discrete, but they vary in size. The content is usually fluid and does not communicate with the tracheobronchial lumen. The fluid content may be serous or mucoid, but it can become purulent if infected.

Infection of a bronchogenic cyst is not uncommon, and several organisms have been identified including *Hemophilus influenza*, *Streptococcus pneumonia*, *Streptococcus pyogenes*, *Escherichia coli* and *Salmonella enteritidis*. However, infection with *M. tuberculosis* is uncommon, especially in infants (4).

This is the first case of bronchogenic cyst infected with *M. tuberculosis* in children in our center. Infection of a bronchogenic cyst can lead to an increase in its dimensions due to increased fluid production and acute presentation due to compression on adjacent structures such as the tracheal and lung parenchyma. This could account for her presentation after living in Nigeria for approximately 10 weeks.

The presentation of our patient is particularly noteworthy because she was born in the UK and was not immunized against tuberculosis where she stayed for 4 months before coming to Africa, where tuberculosis is prevalent. It shows the importance of immunization against childhood diseases, including tuberculosis.

The other reported complications of bronchogenic cyst include dysphagia, arrhythmia, malignant

transformation, and fatal air embolism. Notable tumors include mucoepidermoid carcinoma and melanoma (5). Total or near-total surgical excision of the cyst, as was done in the index case, is the best option to avoid recurrence; however, simple aspiration, marsupialization, or unroofing of the cyst for preventing injury to the tracheobronchial wall due to proximity has been described. This, however, is usually associated with recurrence. Pre-operative cyst drainage either done percutaneously or endoscopically has also been described especially for severe respiratory distress (6, 7). A recent follow-up of the patient 4 months after surgery showed remarkable improvement, with lung expansion on chest x-ray (Figure 3).

This case has again highlighted the high level of tuberculosis prevalence in our environment in Africa and the attendant risk to the populace, including health workers exposed daily to these patients. An equally important fact that cannot be over-emphasized is the role of childhood immunization against tuberculosis.

In conclusion, a high index of suspicion for bronchogenic cyst as a cause of repeated chest infection and respiratory distress in children is necessary. Pre-operative screening for tuberculosis may be important in confirmed cases (of bronchogenic cyst) in children, particularly those who are unimmunized against tuberculosis or are immunosuppressed.

**Author contributions**

All authors contributed equally to writing and editing the original draft.

**References**


1. Limaiem F, Ayadi-Kaddour A, Djilani H. Pulmonary and mediastinal bronchogenic cysts: a clinicopathologic study of 33 cases. Lung. 2008;186:55-61.
2. Chhabria BA, Agarwal R, Garg M, et al. A rare cause of airway obstruction: mediastinal cyst secondarily infected with Mycobacterium tuberculosis. Lung India. 2018;35(5):421-424.
3. Limaiem F, Mlika M. Bronchogenic cyst. 2020 Oct 16. In: StatPearls [Internet]. Treasure Island, FL: StatPearls Publishing; 2021.
4. Lin SH, Lee LN, Chang YL, et al. Infected bronchogenic cyst due to mycobacterium avium in an immuno-competent patient. J Infect. 2005;51:131-133.
5. Cortes-Santiago N, Deutsch GH. Pediatric cystic lung lesions: where are we now? Surg Pathol Clin. 2020;13(4):643-655.
6. Zobel M, Gologorsky R, Lee H, et al. Congenital lung lesions. Semin Pediatr Surg. 2019;28(4):150821.
7. Brisky TA, Rosthenberg SS. Foregut duplication cysts. Pediatr Thorac Surg. 2009;30:385-387.