The Difficult Coughing Infant

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Case Report

Keywords: Bronchoesophageal fistula, congenital, recurrent cough, fever, pneumonia

Posted Date: September 7th, 2023

DOI: https://doi.org/10.21203/rs.3.rs-3199655/v1

Additional Declarations: No competing interests reported.

Abstract

Bronchoesophageal fistula (BEF) is an anomalous connection between the bronchial tree and the esophagus, which can be either congenital or acquired. This case study presents the clinical scenario of a six-month-old male infant of African descent with a history of recurrent cough and breathing difficulties since birth. The patient had received treatment for pneumonia without improvement.

Following a thorough series of investigations, the diagnosis of Bronchoesophageal fistula (BEF) was confirmed prompting the decision to proceed with surgical intervention. The case highlights the importance of maintaining a high index of suspicion to accurately diagnose BEF.

Introduction

Bronchoesophageal fistula is a pathological connection between the bronchial tree and the esophagus. This condition can be either congenital or acquired. Acquired broncho-esophageal fistula may develop due to factors such as infections like tuberculosis, malignancy, trauma caused by prolonged endotracheal intubation or during endoscopic procedures, or even blunt chest injuries. Congenital forms of the condition are rare, occurring less frequently than tracheoesophageal fistulas by a ratio of 25 to 50%. The onset of symptoms can range from the neonatal period to adulthood (1, 2).

Typically, patients with broncho-esophageal fistula present with recurrent episodes of coughing, particularly during meals, and eventually develop recurrent pulmonary infections (3). There is no known racial or gender predisposition associated with this condition (1). This case report presents a case of a six-month-old infant who has experienced recurrent coughing and difficulty in breathing since birth. The patient was initially diagnosed with recurrent pneumonia but did not show any improvement with treatment.

Case presentation

The case involves a six-month-old African infant who was delivered at term following an uneventful triplet pregnancy and with a birth weight of 3.1kg. The infant presented with a history of recurrent cough, starting on the fourth day of life, which progressed from being dry to wet and occurred mostly during or after breastfeeding. Several weeks after the onset of coughing, the infant began experiencing difficulty in breathing and wheezing. At one month of age, recurrent episodes of fever spikes, ranging from low to high grade, also developed.

Seeking medical help, the infant was initially treated in multiple healthcare facilities as a severe pneumonia case but showed no improvement in symptoms. At four months of age, tracheoesophageal fistula was suspected, leading to the insertion of a nasogastric tube. However, the coughing and breathing difficulties persisted, ultimately resulting in a referral to the present hospital at six months of age.

Upon admission, the infant appeared lethargic, malnourished, and in severe respiratory distress. Physical examination revealed tachypnea (respiratory rate of 78 breaths per minute), oxygen saturation below 85% on room air, intercostal recessions, bronchial breath sounds with bilateral crepitations (more pronounced on the left lung), and normal heart sounds. Other systemic examinations were unremarkable.

Complete blood count results showed leukocytosis with neutrophil predominance and moderate microcytic hypochromic anemia. C-reactive protein levels were significantly elevated. Chest X-ray revealed diffuse cystic changes in the left lung and consolidation in the right lung, raising concerns of a congenital lung malformation. A chest CT scan was subsequently performed, confirming communication between the distal left main bronchus and the distal esophagus (**as shown in Image 1).** The CT scan's volume-rendered reformatting displayed extensive cystic changes, consolidation, and a fistulous tract **(as shown in Image 2).**

Based on the diagnostic findings, a diagnosis of bronchoesophageal fistula involving the distal left main bronchus and the distal esophagus, along with severe cystic changes in the left lung and a superimposed infectious process in the right lung, was established. The case was reviewed by a cardiothoracic surgeon who planned for surgical resection. During the operation, sequestered left lung segments containing pus were discovered, along with a bronchoesophageal fistula originating from the lower third of the esophagus, communicating with the left main bronchus. A left pneumonectomy was performed, and the fistula was ligated to address the condition.

Discussion and conclusion

Congenital bronchoesophageal fistulas are a group of rare congenital malformations that were first reported in 1929. Diagnosing bronchoesophageal fistulas can be clinically challenging due to their nonspecific symptoms, as observed in many cases, including the one presented in this study involving a child with a history of recurrent cough and fever. Initially, the child was treated for pneumonia, but symptoms did not improve. Subsequently, the possibility of tracheoesophageal fistula was considered, leading to the insertion of a nasogastric tube, yet the symptoms persisted. Diagnosing this deformity requires a high index of suspicion, as it presents with nonspecific symptoms and may sometimes remain silent until adulthood, especially when not associated with esophageal atresia (1, 5).

Braimbridge and Keith classified bronchoesophageal fistulas into four types. Type I involves congenital bronchoesophageal fistulas associated with congenital esophageal diverticulum, type II refers to simple bronchoesophageal fistulas, type III involves bronchoesophageal fistulas with intralobar cysts, and type IV describes bronchoesophageal fistulas communicating with pulmonary sequestration. In the case of the child discussed in this study, the classification is bronchoesophageal fistula with pulmonary sequestration, specifically type IV, as evident in the CT scan (**as shown in Image 1)** (6).

Bronchoesophageal fistula is an exceptionally rare condition, and a high index of suspicion is required for its diagnosis, especially when a child presents with bouts of coughing, worsened during feeding, resulting

in recurrent respiratory infections. Radiological investigations play a crucial role in accurately diagnosing such cases.

Declarations

Acknowledgements

Authors acknowledge the support from the Muhimbili National hospital. Furthermore, we thank parents and the child to be involved in this case report.

Ethics approval and consent to participate: Not applicable

Consent for publication: Written informed consent for publication and associated images were obtained from child's parents.

Availability of data and materials: Not applicable

Conflicts of interests: The authors have no competing interests to declare.

Funding: Not applicable

Acknowledgements:

Authors acknowledge the support from the Muhimbili National hospital. Furthermore, we thank parents and the child to be involved in this case report.

Authors' contribution: Aika Shoo and Evance Godfrey where primary pediatricians attending this child and responsible for manuscript preparation. Fredrick Lyimo is consultant radiologist who provided radiologist expertise and contributed in manuscript writing. Alphonce Simbila, Rachel Kajuna and Livin Mumburi were involved management and overall follow up when the child was admitted in our hospital. Mary DeAlmeida and Namala Mkopi where pediatric critical care specialist who provided expertise management of this case and in manuscript preparation.

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Figures

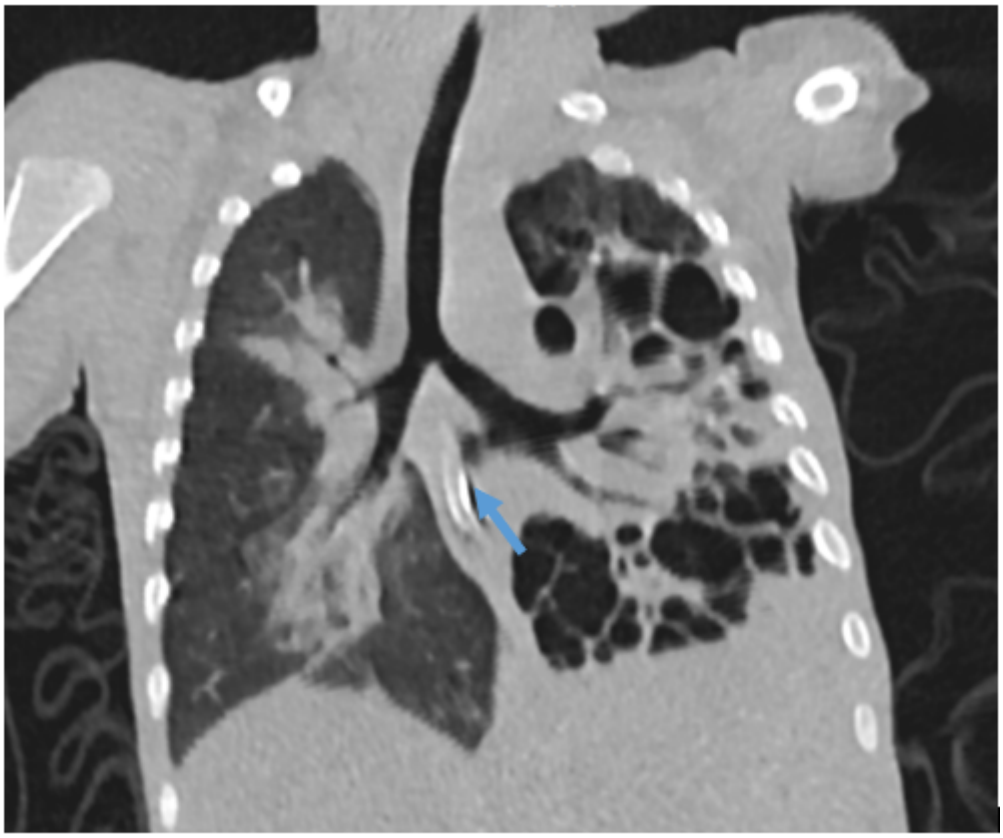

Figure 1

Chest CT coronal reformatted view showing a fistulous connection between a distal left main bronchus and the esophagus (Arrow) and cystic changes.

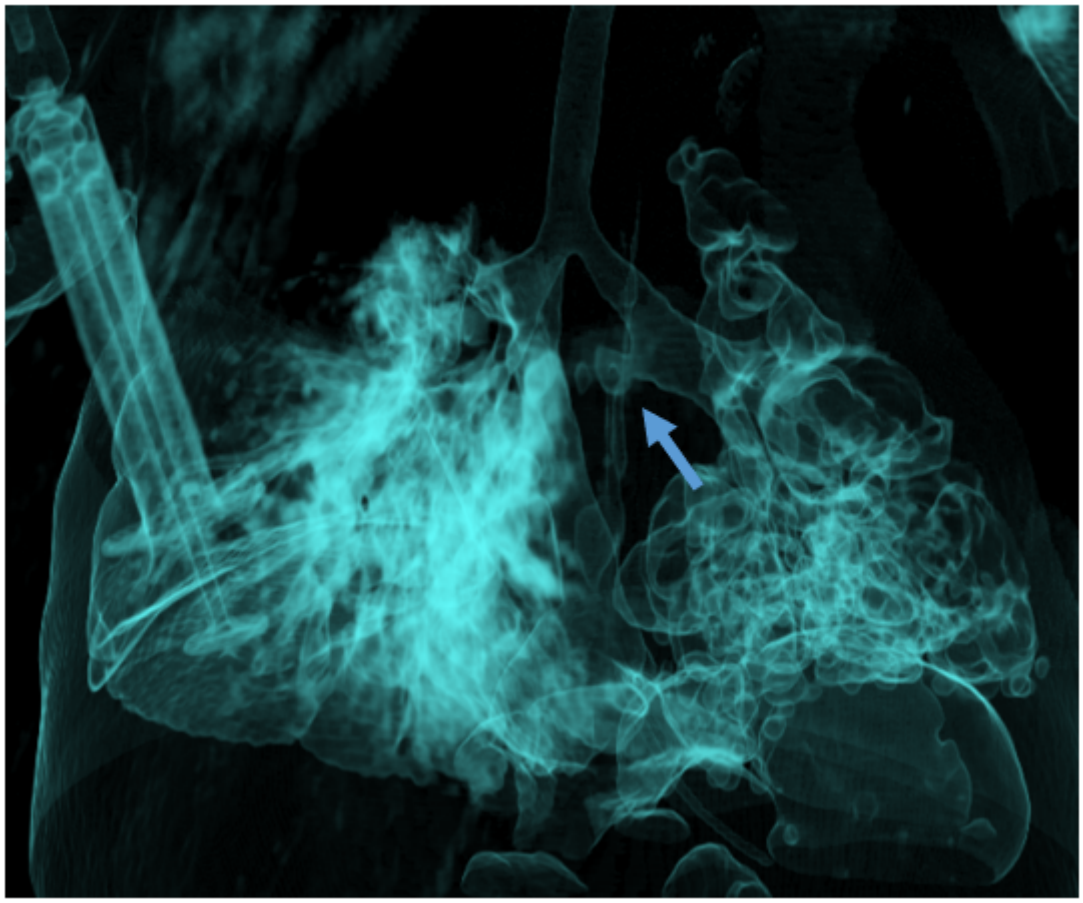

Figure 2

A volume-rendered CT image showing fistulous connections between the distal left main bronchus and the esophagus