

The Diagnostic Challenge of an Accompanying Tracheal Bronchus and Meckel's Diverticulum: A Case Report

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ABSTRACT

Background: A tracheal bronchus is a rare congenital abnormality in which an abnormal bronchus arises from the trachea. The prevalence of a tracheal bronchus is approximately 0.9% to 3% in the population. Due to its low prevalence, association with other anomalies, and overlapping symptoms, this condition often goes undiagnosed or is diagnosed late.

Case Report: This article discusses a case of an infant born at 37 weeks' gestation. The baby required a right thoracotomy after experiencing cyanosis during feeding, leading to a diagnosis of tracheoesophageal fistula. During bronchoscopy, an abnormal orifice was noted above the carina. Subsequently, contrast material was introduced into the orifice and evaluated under fluoroscopy.

Conclusion: The fluoroscopic examination confirmed that the observed orifice did not represent a tracheoesophageal fistula (TEF). Fluoroscopy findings showed that the orifice was an accessory bronchus connected to the right lung. In this case, the use of Visipaque during fluoroscopy significantly aided the diagnosis. The routine use of contrast agents for diagnosing tracheobronchial diseases in children is uncommon. The significant aspect of this case is the utilization of contrast material during fluoroscopy, which facilitated an accurate diagnosis. The low incidence of tracheal bronchus has resulted in limited progress in diagnostic and treatment research. When tracheobronchial anomalies are suspected, the use of contrast agents during imaging can expedite diagnosis.

Keywords: Fluoroscopy, Tracheal bronchus, Tracheobronchial diseases

Introduction

A tracheal bronchus is a rare congenital airway abnormality (1). An atypical bronchus arises from the trachea near the cricoid cartilage (2). The prevalence of tracheal bronchus is approximately 0.9% to 3% in the population (3). The majority of cases occur on the right lateral wall (4). This anomaly is associated with several congenital conditions, including Down syndrome, VATER association, tracheoesophageal fistula, duodenal atresia, and congenital heart defects (5). Symptoms of this abnormality often manifest as

wheezing, stridor, persistent cough, pneumonia, and atelectasis (6). It can present with chronic cough symptoms that persist for several years (7). A classification system anatomically categorizes the tracheal bronchus in relation to the carina. In Type I, the tracheal bronchus is located more than 2 cm away from the carina, and the distal trachea is narrowed. In Type II, the tracheal bronchus is more than 2 cm away from the carina, but the distal trachea does not narrow. In Type III, the tracheal bronchus is situated less than 2 cm above

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the level of the carina. Both chest X-rays and chest CT scans can sometimes detect the presence of a tracheal bronchus (8). Multidetector CT (MDCT) is used for the non-invasive direct diagnosis of tracheal bronchus. However, most cases are discovered incidentally during bronchoscopy (9).

Meckel's diverticulum is a congenital anomaly of the digestive system that is associated with a variety of disorders, including imperforate anus (anorectal malformations), omphalocele, Crohn's disease, duodenal atresia, as well as various neurological and cardiovascular anomalies. It is the most prevalent congenital malformation of the gastrointestinal tract, affecting approximately 2 percent of the population. Notably, most individuals with Meckel's diverticulum remain asymptomatic and do not encounter complications. Furthermore, the frequent association of this condition with a broad spectrum of congenital disorders can result in diagnostic errors and delays in identifying other conditions (10). Due to the low prevalence of tracheal bronchus, its association with other anomalies, and overlapping symptoms, this anomaly often goes undiagnosed or is diagnosed late.

Case report

This article presents the case of an infant born at 37 weeks of gestation, with a birth weight of 2,200 grams and an Apgar score of 8 out of 10. The infant presented with cyanosis from birth, which was observed during the initial breastfeeding. This condition progressively exacerbated the respiratory symptoms, ultimately necessitating intubation. A tracheoesophageal fistula was diagnosed through clinical evidence and imaging, leading to a right thoracotomy and esophagostomy. During surgery, esophageal atresia and Meckel's diverticulum were also identified and treated at the same medical center. After the thoracotomy, a chest tube was placed due to the presence of right pleural effusion. Despite these interventions, the patient did not show signs of recovery and continued to exhibit cyanosis after feedings. A barium swallow test was conducted to investigate potential strictures in the upper digestive tract (Figure 1). Due to the esophageal stricture, esophageal dilatation was performed using a bougie. Despite dilating the esophagus, no improvement was noted in the infant's breastfeeding condition. Following feeding, the infant once again experienced episodes of cyanosis and aspiration.

Given the recurrence of these symptoms, a



Figure 1. Barium test shows signs of esophageal stricture

diagnosis of tracheoesophageal fistula (TEF) recurrence was considered. The patient underwent bronchoscopy. During the bronchoscopy, an abnormal orifice was observed above the carina. Contrast material was subsequently injected into the orifice and evaluated through fluoroscopy. The fluoroscopic examination confirmed that the identified orifice did not correspond to a tracheoesophageal fistula (TEF) (Figure 2). Fluoroscopy findings indicated that the observed orifice corresponded to an accessory bronchus connected to the right lung. Regrettably, during hospitalization, the patient developed sepsis and unfortunately succumbed despite treatment.

Ethical Approval

written informed consent was obtained from the patient's parents for the publication of this case report and any accompanying clinical images. all procedures performed in this study were in accordance with the ethical standards of the Shahid beheshti university of medical sciences.

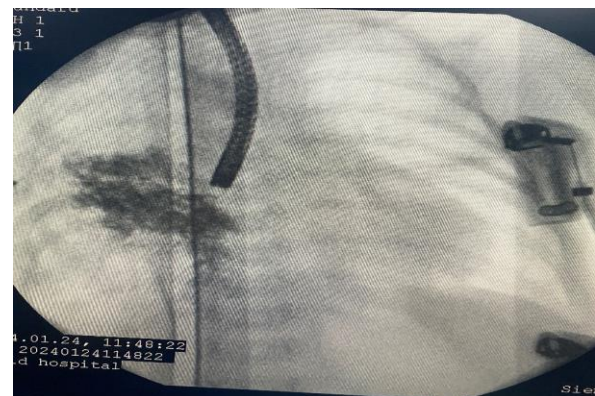


Figure 2. Fluoroscopic findings showing an accessory bronchus

Discussion

Congenital tracheobronchial anomalies in pediatric patients necessitate a comprehensive investigation, especially in those with vague respiratory and digestive symptoms. These anomalies can often lead to troubling signs, including persistent shortness of breath, noticeable cyanosis, and frequent respiratory infections. A detailed assessment is crucial for accurately diagnosing and managing these conditions, which can significantly impact the child's overall health and quality of life (11). The association between tracheobronchial disorders and a range of congenital anomalies often results in overlapping symptoms, which significantly complicates the process of accurate diagnosis. This complexity underscores the critical need for timely identification of patients with a tracheal bronchus, as several essential implications are tied to this diagnosis.

Firstly, patients with a tracheal bronchus require heightened caution during intubation procedures. The unique anatomical variations associated with this condition mean that improper placement of the endotracheal tube can lead to atelectasis, a partial or complete lung collapse, which may result in hypoxia.

Moreover, patients with a tracheal bronchus often experience recurrent respiratory infections without prompt and accurate diagnosis. This is due to the compromised airway structure, which may hinder normal respiratory function and promote the accumulation of secretions, leading to infection. Therefore, swift recognition and management of this condition are essential to mitigate complications and enhance patient outcomes.

The association between Meckel's diverticulum and a diverse set of congenital disorders, such as duodenal and esophageal atresia, often leads to diagnostic errors and delays in identifying related diseases. Considering the association of esophageal and duodenal atresia with tracheoesophageal fistula, paying attention to Meckel's diverticulum in the presence of TEF during surgery is helpful (12, 13). Interestingly, current investigations reveal that the co-occurrence of Meckel's diverticulum and tracheal bronchus has yet to be reported in the medical literature. In this specific case, we observed the presence of two distinct disorders, each contributing to the complexity of the clinical picture.

The case detailed in this article underscores the paramount importance of timely diagnosis, as

delays can result in grave consequences, including the potential death of an infant. In this particular instance, the diagnosis was delayed due to the small size of the orifice. The application of contrast agents in medical imaging has demonstrated significant advantages in diagnosing tracheobronchial diseases. This case highlights the valuable role of Visipaque, a non-ionic iodinated contrast agent, during fluoroscopic examinations. The use of Visipaque not only enhanced the visualization of the tracheobronchial structures but also provided crucial information that aided in reaching an accurate diagnosis.

Notably, the routine application of contrast agents in diagnosing tracheobronchial diseases among pediatric patients has yet to be widely adopted. This may be due to concerns regarding the safety and potential adverse effects of contrast materials in children. However, the positive outcomes observed in this case suggest a need for further exploration into the use of contrast agents in pediatric settings.

Overall, this case underscores a critical insight into the effectiveness of using contrast material during fluoroscopy. This approach can facilitate a more precise diagnosis, ultimately leading to improved management and treatment of tracheobronchial conditions.

Conclusion

The rarity of tracheal bronchus disease hampers research on its diagnosis and treatment. This case emphasizes the need for timely and accurate diagnosis in suspected cases. Using contrast agents in imaging procedures like CT scans or bronchograms can improve the visibility of tracheobronchial structures, resulting in faster diagnoses.

Additionally, sharing detailed case reports is essential for enhancing our understanding of this condition. These reports offer valuable insights into its clinical presentation and progression, helping to identify effective diagnostic and treatment strategies. Increasing awareness of tracheobronchial diseases can improve patient outcomes and encourage further research.

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Conflicts of interest

The authors declare that no competing interest exists.

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