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Case Report

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Esophageal Duplication Cyst Mimicking Hydatid Cyst in a Child: a Rare Diagnostic Challenge Case Report

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Abstract

Esophageal duplication cysts are rare congenital anomalies that account for approximately 0.5 to 2.5% of esophageal masses and are more commonly observed in males. These cysts are usually asymptomatic, but when symptoms do appear, they primarily manifest during childhood. In this study, we report a case of a 3-year-old child presenting with symptoms of chest pain, cough, and gastrointestinal complaints. Initially, the condition was suspected to be a hydatid cyst, but after serological testing and advanced imaging CT scan, the final diagnosis was identified as a foregut duplication cyst. Histopathological evaluations revealed the presence of squamous epithelium, granulation tissue, and giant cell reaction characteristics typical of duplication cysts. The patient underwent surgery and was treated with antibiotics, with follow-up showing no complications. This study emphasizes the necessity of accurate differential diagnosis among various pediatric pulmonary cysts and highlights the importance of further research to determine their prevalence, etiology, and treatment methods.

Keywords: Anti-bacterial agents, Chest pain, Cough, Cysts, Echinococcosis, Epithelium, Granulation tissue

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Introduction

Esophageal duplication cysts are rare congenital anomalies, accounting for approximately 0.5 to 2.5% of all esophageal masses, with an estimated prevalence of about 1 in 8,200 individuals, occurring more frequently in males. While the exact etiology remains unclear, they are believed to result from a failure in the intrauterine vacuolization process of the esophagus between the 4th and 8th weeks of embryonic development (1). These cysts are typically asymptomatic; however, when symptoms do occur, they most often present during childhood. Cysts located in the upper esophagus may cause respiratory symptoms such as dyspnea, stridor, and retrosternal pain, whereas those in the lower esophagus are more commonly associated with dysphagia, food obstruction, and vomiting. Proposed mechanisms for their development include the notochordal cleft theory, incomplete recanalization during foregut development, and persistence of enteric diverticula (2). From an embryological perspective, the foregut gives rise to the pharynx, respiratory tract, esophagus, stomach, and part of the duodenum. Duplications are congenital malformations arising from developmental abnormalities, and several theories have been proposed to explain their formation; these duplications typically present as cystic or spherical structures attached to segments of the gastrointestinal tract, characterized by a smooth muscle wall and a mucosal lining (3).

Duplication cysts are often discovered incidentally in asymptomatic patients. However, they may become symptomatic when complications such as infection, compression of adjacent structures, hemorrhage, rupture, or neoplastic transformation occur. Surgical excision of the cyst is generally recommended immediately upon diagnosis, regardless of the presence or absence of symptoms. On the other hand, surgical intervention for asymptomatic cysts may also result in long-term complications such as gastroesophageal reflux and heartburn, and carries a reported mortality risk of up to 1% (4).

Treatment of duplication cysts typically involves surgical resection, which can be performed *via* thoracotomy or thoracoscopic approaches. In cases of gastric duplications, surgical removal is advised to prevent complications such as ulceration or perforation. Bronchogenic cysts, which are usually located near the trachea, also require surgical intervention if they become symptomatic (5).

Case Presentation

A 3-year-old male presented with a history of chest pain and cough. The patient was feverish prior to admission. The onset of symptoms, including cough, hemoptysis, chest pain, and dyspnea, occurred 8 days before presentation. The patient was referred due to a cyst discovered on chest radiography and planned surgery for a suspected hydatid cyst. On admission, vital signs were: heart rate of 97/bpm and respiratory rate of 22/bpm. In addition, the laboratory parameters examined by the patient are also mentioned (Table 1). The patient also reported abdominal pain in the epigastric region for the past 3 months and constipation for a few days prior to admission. Given the history of contact with livestock, a hydatid cyst was initially suspected.

A subsequent pleural ultrasound was performed to

Table 1. Blood test results of the patient studied

Blood parameters	Result	Unit
WBC	11	10^3 *cells/µl*
RBC	4.66	10^3 *cells/µl*
Hgb	10.5	*g/dl*
Hct	33.6	%
PLT.c	451	×10^3/
MCV	72.1	*fL*
MCH	22.5	*pg*
MCHC	31.3	*g/dl*
RDW	14.4	%
ESR	97	*mm/h*
Creatinine	0.6	*mg/dl*
AST (SGOT)	23	*IU/Lit*
ALT (SGPT)	9	*IU/Lit*
Alk phosphatase	388	*IU/Lit*
Ca	9.6	*mg/dl*
Na	135	*mEq/lit*
K	4.3	*mEq/lit*
CRP	22	*mg/l*

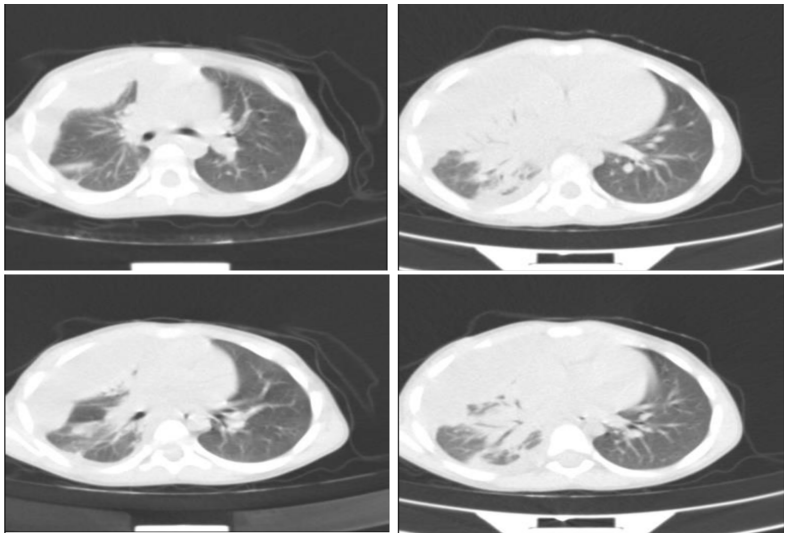


Figure 1. Spiral CT scan of the thorax without IV contrast: Pleural thickening with loculated effusion is seen in the right hemithorax, raising suspicion for empyema. A cystic lesion with adjacent passive subsegmental atelectasis is noted in the Right Lower Lobe (RLL).

evaluate the pleural effusion, which was reported as mild. The pleural ultrasound showed a mild right-sided pleural effusion and a cystic lesion measuring 60×47 *mm*. Due to the mild nature of the pleural fluid, fluid analysis and thoracentesis were not deemed necessary at this stage.

The radiologist had requested a contrast-enhanced CT scan for further evaluation (Figure 1). However, given the strong clinical suspicion of a hydatid cyst as the definitive pre-operative diagnosis, the priority was to proceed with surgery as soon as possible. The diagnosis of a duplication cyst was confirmed. This diagnosis was supported by the clinical presentation (chest pain and cough), imaging findings (cyst on CT scan), and histopathological results (Figure 2).

Intraoperatively, the surgery commenced with the assumption of a hydatid cyst. It was discovered that the lesion was, in fact, a foregut (duplication) cyst, appearing to have a connection with the esophagus. Intraoperative findings suggested empyema, but no compressive effect on adjacent structures was observed. Following surgery, the diagnosis of a foregut cyst was made, and endoscopy was not included in the diagnostic and treatment protocols.

Postoperatively, the patient received Cefixime and Metronidazole for follow-up care and was scheduled for a chest X-ray in 10 days (Figure 3). Follow-up radiography showed improvement (Figure 4).

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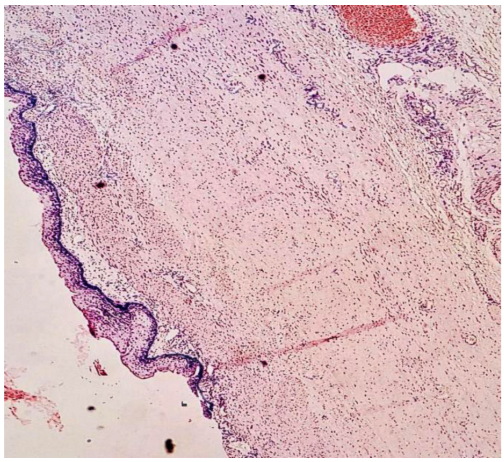


Figure 2. The resected lesion was a 7.5×7×3.1 *cm* foregut duplication cyst, characterized by a benign squamous lining with granulation tissue and giant cell reaction on microscopy.

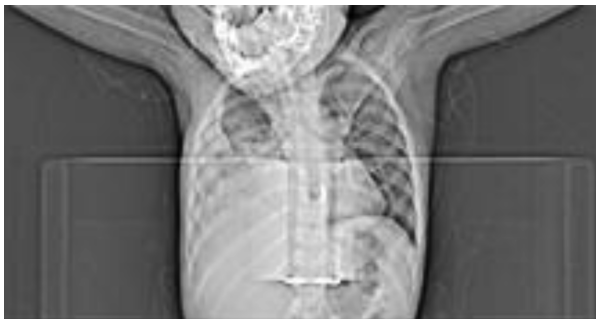


Figure 3. Pre-treatment chest X-ray.

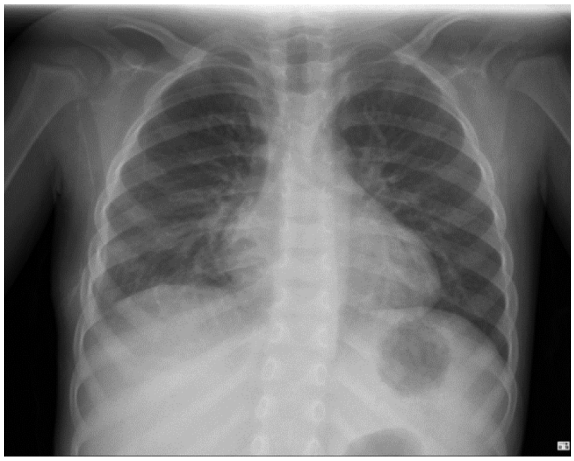


Figure 4. Follow-up chest X-ray after treatment and bronchoscopy showing improvement.

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Discussion

These cysts are classified into three main types: foregut duplication cysts, small intestine duplication cysts, and large intestine duplication cysts. Foregut duplication cysts are further subdivided into esophageal, bronchogenic, and neuroenteric cysts, based on their embryologic origin. Bronchogenic and esophageal cysts are considered to result from abnormal development of the foregut during the 5th to 8th weeks of gestation. However, the exact embryological origin of these cysts remains incompletely understood. Approximately 50-70% of foregut duplication cysts are enterogenous, while 7-15% are bronchogenic. These cysts account for about 6-15% of primary mediastinal masses.

Duplication cysts are most commonly found in the ileum, esophagus, and colon, and can present as either cystic (80%) or tubular (20%) lesions. They may be located within the wall of the gastrointestinal tract or external to it (6). In 1968, Gerle *et al* (7) introduced the term "foregut bronchopulmonary malformations" to describe a spectrum of congenital abnormalities related to foregut development. Today, the term "foregut cyst" is often used to encompass a variety of bronchopulmonary anomalies, including bronchogenic cysts, pulmonary sequestrations, and combined bronchogenic-esophageal duplications. However, this classical classification does not encompass all known variants. This hybrid nature is evident in gastric duplications lined by respiratory epithelium, or in esophagogastric lesions that contain smooth muscle, cartilage, and disorganized bronchiolar-like structures lined with mixed ciliated and squamous epithelium (8). The clinical manifestations of foregut duplication cysts may include abdominal symptoms such as a palpable abdominal mass, pain, vomiting, gastrointestinal bleeding, and weight loss (9).

Other reported symptoms include epigastric discomfort or pain, chronic cough, and hemoptysis. However, most cases are asymptomatic and are diagnosed incidentally (10,11).

In the present case, the foregut duplication cyst was discovered incidentally, as the patient was investigated due to a history of close contact with livestock carrying hydatid cysts. Depending on the epithelial lining of the cyst, several cases of adenocarcinoma, squamous cell carcinoma, and neuroendocrine carcinoma have been reported (12).

Conclusion

Esophageal duplication cysts, though rare, should be considered in the differential diagnosis of thoracic cystic lesions in children, especially when presenting with non-specific respiratory and gastrointestinal symptoms. In this case, a foregut duplication cyst mimicked a hydatid cyst, underlining the importance of advanced imaging and histopathological confirmation for accurate diagnosis. Surgical excision remains the definitive treatment, and postoperative management is crucial for preventing complications. This case highlights the necessity of clinical vigilance and multidisciplinary evaluation in diagnosing congenital thoracic anomalies and advocates for further research into their etiology, classification, and optimal management strategies.

Limitations of study

The clinical presentation, diagnostic process, and treatment outcomes described may not reflect the full spectrum of foregut duplication cysts in other pediatric patients. Additionally, long-term follow-up data were not available to assess potential delayed complications or recurrence. The absence of advanced molecular or genetic analysis also limits a deeper understanding of the underlying etiology.

Ethical statement

The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian has given consent for images and other clinical information to be reported in the journal. The guardian understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

The study was approved by the Ethical Committee of the National Research Institute of Tuberculosis and Lung Diseases (NRITLD) of Shahid Beheshti University of Medical Sciences (IR.SBMU.NRITLD.REC.1404.032). All the steps performed in this study were based on the terms of the Helsinki Convention.

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Conflict of Interest

There are no conflict of interest.

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