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Mediastinal and retroperitoneal transdiaphragmatic bronchogenic cyst with gastric mucosa

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ABSTRACT

Background: Congenital cystic lung lesions are rare lung malformations. They are mostly diagnosed by a routine antenatal ultrasound scanning. Clinical symptoms ranging from asymptomatic condition to life-threatening course.
Case and outcomes: We presented a case of congenital cystic lung malformation, the case demonstrated a 20- month- old boy who was reported to our hospital because he was diagnosed prenatally with a right-sided cystic lung mass. He was asymptomatic. A diagnosis of mediastinal retroperitoneal bronchogenic cyst was made radiologically. He underwent a successful right thoracotomy and the transdiaphragmatic cystic mass was completely resected. Histologic examination revealed bronchogenic cyst with gastric mucosa.
Conclusion: Early diagnosis improves outcomes in infants with congenital cystic lung anomalies. Surgical management is the treatment of choice for congenital cystic lung malformations.

1. Introduction

Congenital cystic lung lesions comprise cystic adenomatoid malformations, pulmonary sequestrations, congenital lobar emphysema, and bronchogenic cysts [1]. All these malformations are rare. Clinical symptoms are vary from asymptomatic to life-threatening [2]. Prenatal diagnosis of congenital lung malformations are most often found in prenatal imaging during pregnancy in the second trimester [3]. Surgical treatment plays a key role for the successful management of all these lesions, because of the risk of developing complications [4].

We describe a case of a 20- month- old boy with bronchogenic cyst. We report this case because of its rarity. While many studies have investigated bronchogenic cysts, we have searched medical English literature, but we haven't come across a case report of mediastinal retroperitoneal transdiaphragmatic bronchogenic cyst with gastric mucosa. We report what we believe to be the first documented case report that described this type of bronchogenic cysts.

Case Report: A 20- month- old infant was brought to our hospital in May 2021. He was diagnosed prenatally with a right-sided cystic lung mass. He was evaluated in a local hospital and was suspected to have a cystic lung mass on chest radiograph and hence referred to our hospital for further management and examination of the lesion. He was the third child to non-consanguineous parents. His neonatal period was uneventful and he was feeding well and active. At admission the patient was asymptomatic with no complaints of weight loss, fever, cough, or vomit. CXR revealed a well-defined rounded opacity in the right lower zone (Fig. 1). Chest and abdominal examinations showed no remarkable features. There was no obvious positive characteristics on physical exami-

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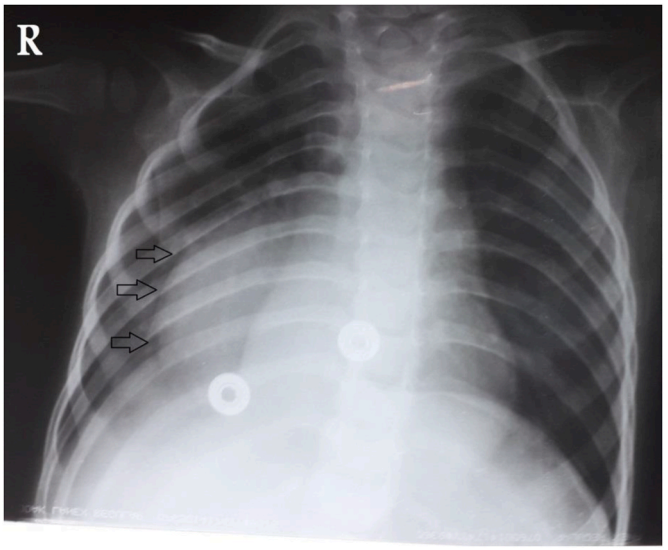

Fig. 1. CXR revealed a well-defined rounded opacity in the right lower zone.

nation. He had stable vital signs. Laboratory tests including complete blood counts, liver function test and blood chemistry were normal. Anti-hydatic cyst antibody test was negative. The mass was further characterized by a thoracic and abdominal computed tomography scan which demonstrated a hypodense, thin-walled cystic lesion on the right in the posterior mediastinum with extension into the retroperitoneum, no calcifications or solid components were noted (Fig. 2). Echocardiography was normal. The child underwent a right thoracotomy and the transdiaphragmatic cystic mass was completely resected and the diaphragmatic defect was directly repaired. The mass was not associated with diaphragmatic hernia or other congenital anomaly. There was no communication with any abdominal structure (Fig. 3). The cyst was filled with thick, brownish mucoid secretions (Fig. 4). . Cultures of the fluid did not yield any bacteria or fungi. The patient's intra-operative and postoperative evolution was uneventful with a rapid recovery. After surgical X-ray of chest showed no cystic lesions with right lower zone consolidation (Fig. 5). The patient was discharged home on post-operative day three. The cyst was submitted for microscopic examination. On gross examination, the specimen consisted of a cystic lesion (Fig. 6). Histologic examination showed presence of muscular fibers, fibrosis, cavity formation lined by ulcerative respiratory epithelium, with hemorrhage, severe mixed inflammatory infiltrate with hemorrhage and fibrosis. Focal presence of thin gastric mucosa surrounded by muscular fibers. No evidence of dysplasia or malignancy (Fig. 7). The diagnosis of bronchogenic cyst with gastric mucosa was finally confirmed.

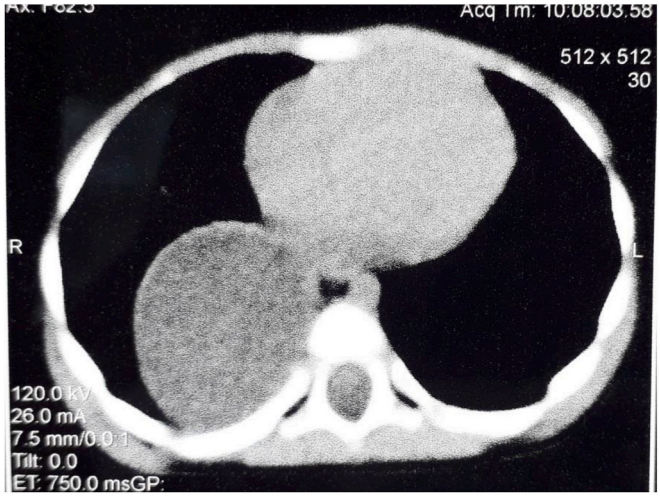

Fig. 2. Thoracic computed tomography scan demonstrated a hypodense, thin-walled cystic lesion on the right in the posterior mediastinum with extension into the retroperitoneum, no calcifications or solid components were noted.

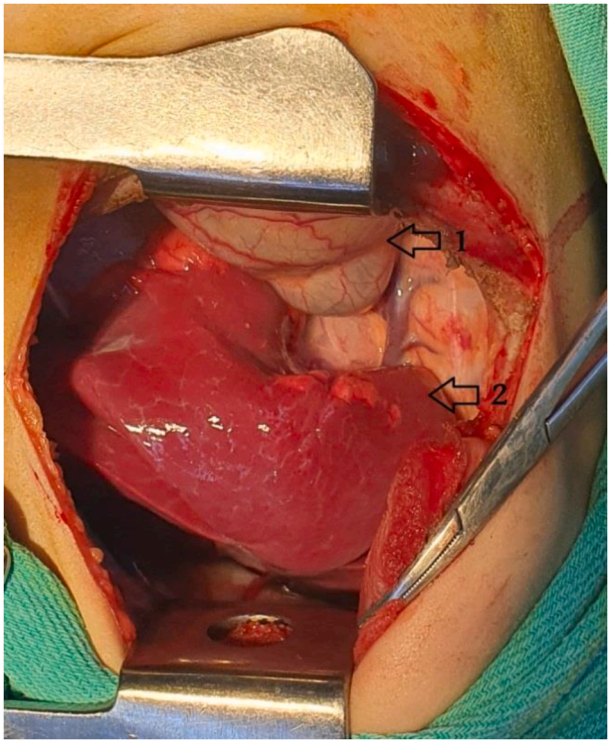

Fig. 3. Intra-operative view showing: 1- the bronchogenic cyst, 2- right lower lobe of the lung.

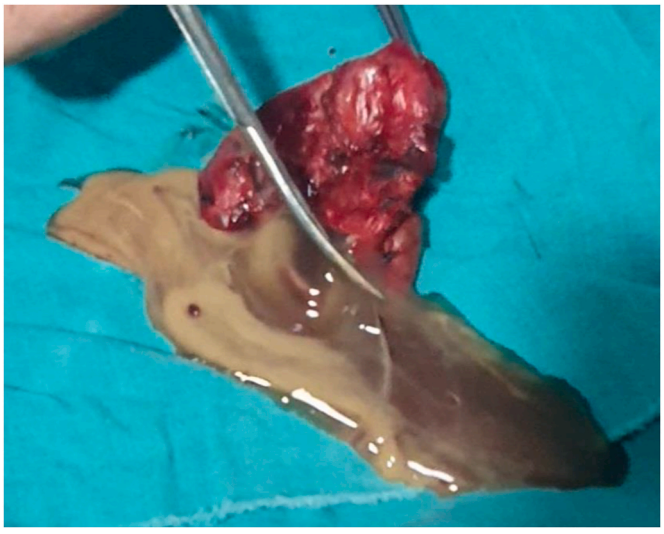

Fig. 4. The cyst was filled with thick, brownish mucoid secretions.

2. Discussion

Foregut duplications are rare cystic masses. It represents 20% of mediastinal masses. These duplications include a wide spectrum of malformations, usually enteric cysts or bronchogenic anomalies. Bronchogenic cysts (BC) are the most common representing 50–60% of cases and can be found in many locations (paraesophageal, paratracheal, perihilar, intraparenchymal; rarely in the tongue, neck, back), but they are usually found in the upper airways [6]. In our case, the cyst located on the right in the posterior mediastinum with extension into the retroperitoneum. Most foregut cysts arise from aberrant foregut which develop between the 4th and 8th week of gestation. BCs develop from abnormal budding of the ventral foregut. The BC is lined by ciliated, pseudostratified, columnar epithelium and it frequently includes elements of the tracheobronchial tree such as bronchial glands and smooth muscle bundles. Enteric cysts result from abnormal budding of the dorsal foregut and they are usually lined with gastrointestinal epithelium

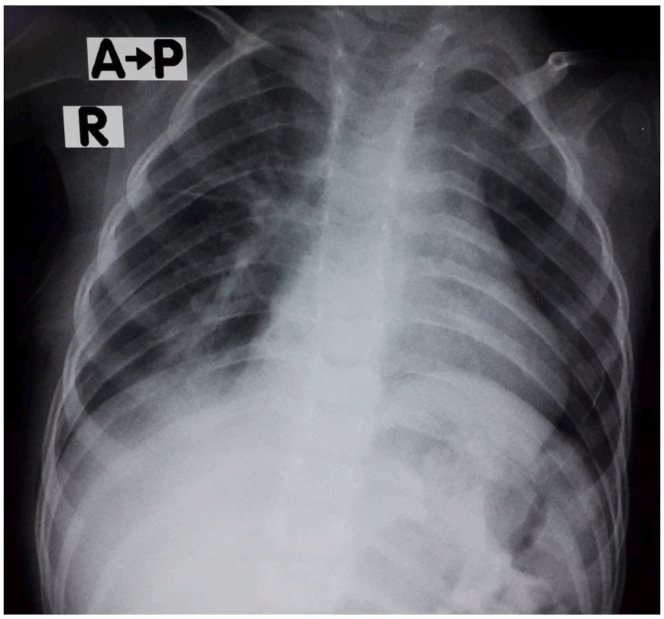

Fig. 5. After surgical X-ray of chest showed no cystic lesions with right lower zone consolidation.

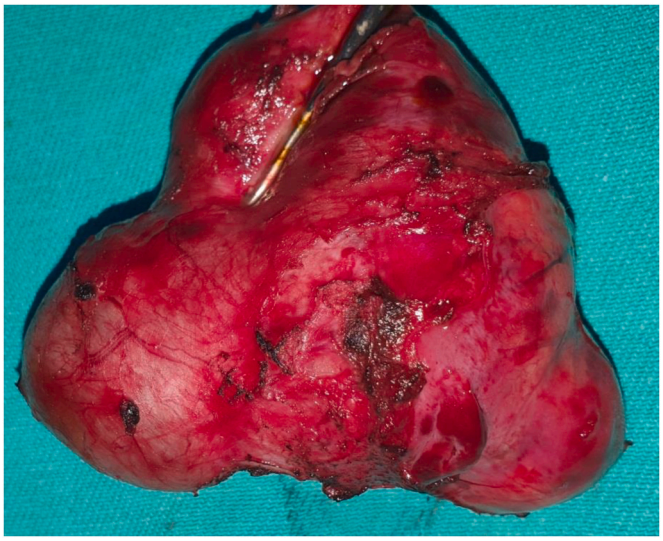

Fig. 6. Gross appearance of the resected cyst.

[7]. In our case, the cyst was lined by respiratory epithelium with focal presence of gastric mucosa. The majority of children with foregut duplications are asymptomatic at birth. Occasionally some patients may have symptoms like acute respiratory distress, stridor, dysphagia, hematemesis, and sudden appearance of a cervical mass [8]. In our case the child was asymptomatic. One-third of foregut cysts at least will be discovered incidentally on chest X-ray, computed tomography, or prenatal ultrasound., the other two-thirds of patients will present with a wide range of clinical symptoms. Any evidence of a cystic mass on chest X-ray should be followed by a CT scan or MRI, which may aid in preoperative planning, not only help in diagnosis [9]. The clinical presentation of BC is variable, and mainly appears during childhood, although some cases have been described in adults. During the antenatal period, the diagnosis can be made using echography [4]. In our case the diagnosis was established prenatally by ultrasound examination. Treatment of BC consists of complete surgical resection, due to the potential complications and the risk of malignant change [5]. Lucile Fievet et al. reviewed 36 patients with BC managed from 2000 to 2011. They concluded that early surgical resection of BCs provides better conservation of pulmonary parenchyma, a lower incidence of inflammatory lesions, and a reduced rate of complications, and should be proposed after prenatal diagnosis, between the 6th and 12th month of life [10]. In our case, the patient referred to our hospital at the age of twenty month old, the decision of surgical resection was made after a case discussion with pediatric pulmonologist and thoracic surgeon.

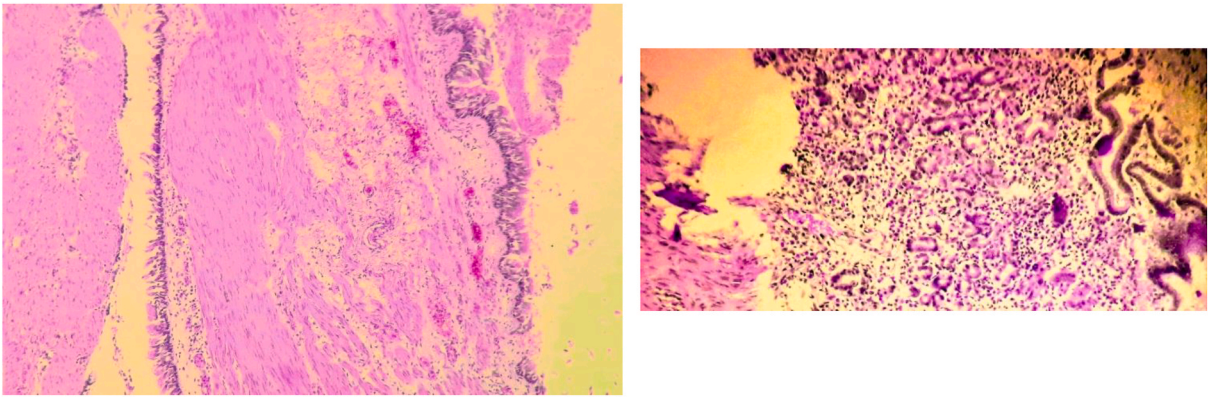

Fig. 7. Histologic findings of bronchogenic cyst lined by respiratory epithelium (on the left), with focal presence of thin gastric mucosa surrounded by muscular fibers (on the right).

3. Conclusion

Congenital cystic lesions of the lung include a spectrum of malformations. Understanding of the antenatal and postnatal presentation of these malformations is important for investigations, diagnosis, and treatment.

Author contributions

All authors contributed to the development of the manuscript and the care of the patient presented. All authors approved the final manuscript.

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Ethical approval

Institutional review board approval is not required for deidentified single case reports or histories based on institutional policies.

Consent

The patient's mothers consented to the publication of this deidentified case reports.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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