

Foregut Duplication Cyst Mimicking Pulmonary Tuberculosis: A Rare Presentation

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Abstract

Foregut duplication cyst is a rare congenital anomaly in children. Most of the children are asymptomatic and are diagnosed incidentally when evaluated for other problems. Complications include infection, rupture, and malignant transformation. Here, we present a case of complicated foregut duplication cyst in a 10-year-old female which mimicked tubercular spondylitis on imaging.

Keywords: Foregut duplication cyst, heterotopia, tubercular spondylitis

INTRODUCTION

Foregut duplication cyst results from the abnormal budding of the embryonic foregut. Duplication can occur anywhere along the gut. However, the most common location is along the lower thoracic esophagus and on the right side.^[1] Although most of the children are asymptomatic, symptoms occur depending on the location of the cyst as well as complications. Our case presents a rare complication of foregut duplication cyst with rupture into the tracheobronchial tree and infiltration of the lung parenchyma.

CASE REPORT

A 10-year-old female presented to the pediatric outpatient department with complaints of chest pain, hemoptysis, and nonbilious vomiting for 1 month. There was no history of fever. There was a history of admission at the age of 1 and 8 years for pneumonia. Chest X-ray done during the current admission revealed a soft-tissue opacity in the right paravertebral region. There was evidence of fusion of D3 and D4 vertebra [Figure 1a]. Contrast-enhanced computerized tomography (CECT) revealed a heterogeneously enhancing soft tissue showing areas of necrosis in the center in the pre- and paravertebral region extending from D2–D7 level with fusion of D3–D4 vertebra and bony sclerosis. There was also a thin-walled cavitory lesion in the right upper lobe adjacent to the soft tissue with the presence of surrounding ground-glass opacity [Figure 1b-d]. On the basis of the current

imaging, diagnosis of tubercular spondylitis was considered. Meanwhile, the CECT which was performed when the child was admitted at the age of 8 years was available and it was reviewed. It revealed a well-marginated cystic lesion in the pre- and right paravertebral location which was closely related to the esophagus causing extrinsic compression on the trachea and right main stem bronchus [Figure 2]. In view of the previous imaging, a final diagnosis of foregut duplication cyst with rupture into tracheobronchial tree and adjacent lung infiltration was made. Tc-99m pertechnetate radionuclide scan was done which showed a tracer uptake in the lesions suggesting the presence of ectopic gastric mucosa in the cyst which further confirmed the diagnosis. The child underwent right posterolateral thoracotomy, and a duplication cyst of size 7 cm × 5 cm × 4 cm was found in the right paravertebral location densely adherent to the right upper and lower lobe near the fissure. The cyst was excised in toto with some part of the right upper lobe. Postoperative period was uneventful. The child was discharged on postoperative day 7 and was doing well till the last follow-up. Histopathological

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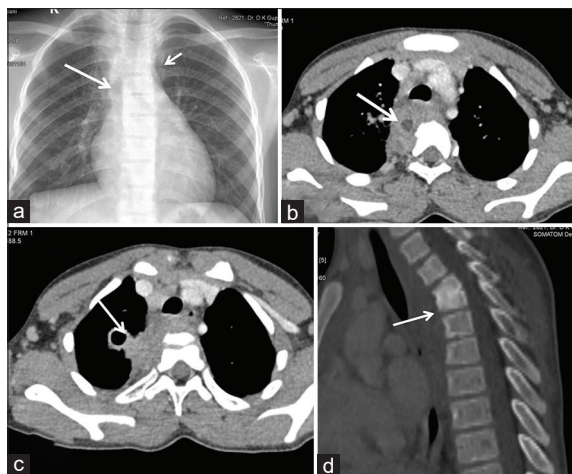


Figure 1: (a) Chest X-ray posteroanterior view shows well-marginated soft-tissue opacity with convex margin seen in the right paravertebral region (large arrow) with fusion of D3–D4 vertebra (small arrow). (b and c) Contrast-enhanced computerized tomography axial view depicts the heterogeneously enhancing soft tissue in the pre- and paravertebral region showing areas of necrosis (arrow in b) with adjacent lung infiltration and cavity formation in the right upper lobe (arrow in c). (d) Sagittal reformatted sections of the bone window show fusion of D3–D4 vertebra with the presence of bony sclerosis

examination revealed the presence of fibromuscular wall lined by gastric-type mucosa. Pancreatic heterotopia was also noted in the wall. There was no evidence of dysplasia present.

DISCUSSION

Esophageal duplication cysts (EDCs) and bronchogenic cysts are alterations of primitive foregut development and are rare entities. They are together grouped as foregut duplication cysts. Most of these are asymptomatic and are discovered incidentally. Few of these cysts can become symptomatic, and they depend on the size as well as location. Symptoms are present when they are large and cause compression of adjacent structures or when they get secondarily infected.^[2]

The most common location of the EDC is on the right side of the mediastinum. Majority are located along the lower esophagus, followed by mid and upper esophagus. Upper esophageal cysts cause compression on the tracheobronchial tree and present with cough and recurrent respiratory tract infections.^[3] Those located near mid and lower esophagus present with dysphagia, vomiting, and chest pain. In our case, the child presented with nonbilious vomiting. Ectopic gastric mucosa in the duplication cyst can be seen in 17%–36% of cases.^[4] Intracystic hemorrhage and fistula with tracheobronchial tree can occur resulting in hemoptysis and pneumonia. In our case, the cyst was seen in the mid esophagus region which got ruptured into the tracheobronchial tree. Complications of foregut duplication cysts include infection, rupture, and malignant transformation.^[5] Initial investigation of choice is chest X-ray which can show a soft-tissue opacity in the paravertebral region with or without vertebral anomaly. Barium swallow can demonstrate the mass effect/indentation on the esophagus. CECT/magnetic

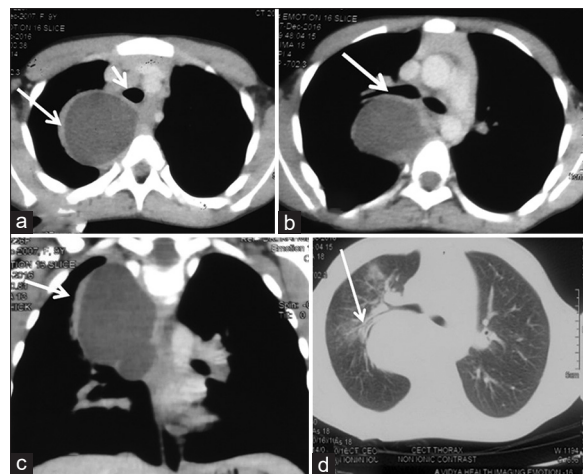


Figure 2: (a-d) Contrast-enhanced computerized tomography axial view of the previous study shows cystic lesion in the middle and posterior mediastinum (large arrow in a) in relation to the esophagus causing indentation on the trachea (small arrow in a) and right main stem bronchus (arrow in b). Coronal reformatted image (c) delineates the extent of cyst (arrow in c). Lung window (d) shows ground-glass opacity in the right upper lobe adjacent to the cyst

resonance imaging is done subsequently to localize the cyst, nature of cyst – simple or infected, mass effect on the adjacent structures, and associated lung changes. In our patient, the cyst had possibly ruptured into the tracheobronchial tree, and there was adjacent lung infiltration. Meckel scan can help in demonstrating ectopic gastric mucosa.

One of the differential diagnoses of a mediastinal lesion in children is esophageal cyst. However, in our case, the possibility of tubercular spondylitis was considered due to the presence of a necrotic soft tissue in pre- and paravertebral abscess with reduced disc space and presence of thin-walled cavitory lesion in the right upper lobe. However, when the previous imaging was evaluated, the diagnosis of ruptured foregut duplication cyst with lung involvement was made.

Surgical resection is done for symptomatic as well as asymptomatic lesions in order to avoid complications.^[6] The cyst should be removed completely as incomplete removal can lead to recurrence. Resection can be either open or thoracoscopic depending on the location of the cyst and the surgeon's expertise. In the index case, right posterolateral thoracotomy was done with excision of cyst.

CONCLUSION

EDC, although is rare, should be included in the differential diagnosis in a child presenting with vomiting and chest pain. CECT is the modality of choice to diagnose and also to look for complications. Meckel scan should be done to look for ectopic gastric mucosa, especially when it has ruptured.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the parent has given his

consent for the images and other clinical information to be reported in the journal. The patient understand that their child's names and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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