Case Report

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Gastric duplication cyst as a differential for an intra-thoracic cystic mass

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ABSTRACT

We report a case of a neonate who presented with respiratory distress initially managed for a suspected pneumothorax before being transferred to a tertiary centre where he had a thoracotomy. A large cystic structure was excised later histologically confirmed to be a gastric duplication cyst. We discuss its management.

Key words: Bronchopulmonary foregut malformation, gastric duplication cyst, intra-thoracic mass

INTRODUCTION

Alimentary duct duplications have a reported incidence of 1 in 4500, and only 20% are located in the thorax. Gastric duplication cysts (GDCs) count for <4% of all duplication cysts.[1-4] Most GDCs are found along the greater curvature of the stomach and most are cystic and noncommunicating.[5] To our knowledge, this is the third reported case report in the English literature of an intra-thoracic occurring GDC. The first case report was by Daher *et al*. which was an antenatal diagnosed (33 weeks) intra-thoracic cystic mass with associated vertebral anomalies. The second case report was reported by Turkyilmaz *et al*. of a 10 days old presenting with respiratory distress who was found to have two intra-thoracic masses but no associated anomalies. Both patients had a thoracotomy and GDC confirmed on histology.

CASE REPORT

The male patient was born at full-term via uneventful vaginal delivery with a birth weight of 4 kg. All antenatal scans were reported as normal. There were no immediate postnatal concerns and the child was discharged routinely. At 17 days of age, he initially attended his local hospital following a blue episode after choking on feeds with subsequent vomiting. Our patient experienced episodes of vomiting, lethargy and increased difficulty in feeding in the 24 h prior to presentation. The recorded observations on attendance revealed tachypnoea though the patient maintained adequate oxygen saturation in air. The attending team diagnosed bilateral pneumothoraces on chest X-ray [Figure 1], greater on the right. A right chest drain was promptly inserted. The patient was commenced on intravenous antibiotics empirically for suspected chest sepsis.

After a short period, the patient developed increasing oxygen requirement, and a second chest drain was inserted on the right side. The patient was then discussed with our surgical on-call team and subsequently transferred to our high dependency unit.

On arrival, we found the patient to have minimal respiratory distress and did not require additional oxygen supplementation. On reviewing the original

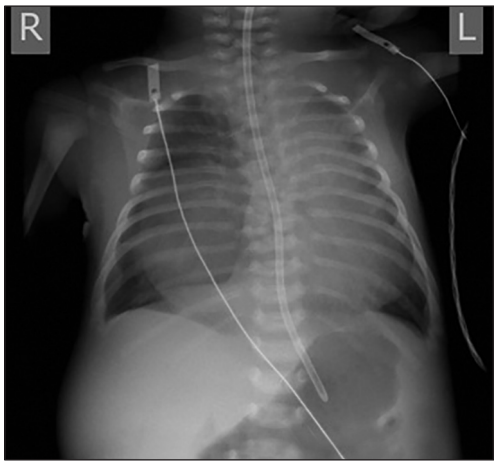

Figure 1: Chest X-ray taken at initial presentation-showing mass in right hemi-thorax causing mediastinal displacement. Thoracic vertebral anomalies can also be identified

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chest X-rays taken by the referring hospital, our impression was the patient had a thoracic mass resulting in mediastinal displacement. There was no evidence of a pneumothorax however we could identify anomalies of T5 to T7 vertebrae. Subsequent computed tomography (CT) [Figure 2] of the chest confirmed a large cystic mass (measuring 4.5 cm) in the posterior mediastinum, displacing the oesophagus anteriorly and remaining mediastinal contents to the left. The tip of one chest drain was seen within the lung parenchyma, and consolidation of the left lower zone was reported. In view of the vertebral anomalies, magnetic resonance imaging (MRI) [Figure 3] was conducted. T5 hemivertebra and split butterfly T6 and T7 were reported. Abdominal ultrasound was reported to be normal.

The chest drains were removed serially with no sequelae of parenchymal injury. On day 7 of admission, the patient underwent microlaryngoscopy and bronchoscopy, which demonstrated possible compression of right bronchus intermedius. He then had a right thoracotomy. The cyst was identified in the middle mediastinum, it was large and tense. Thick yellow fluid was aspirated to facilitate access to its posterior attachment and afferent vessel from the thoracic aorta. The cyst was then carefully dissected out. A chest drain was left *in-situ*. The patient was extubated 18 h after surgery.

The patient made prompt recovery and was discharged by day 7 after surgery.

He appears in excellent condition at his 2 months review. A final repeat chest X-ray shows normal lung expansion and mediastinal contours. Histology confirmed the cyst to be of gastric origin; walls containing gastric mucosa, a double layer of muscularis propria and visible myenteric plexus with ganglion cells.

DISCUSSION

Bronchopulmonary foregut malformations (BPFM) should be considered in the presence of an intra-thoracic mass. BPFM results from anomalous budding of the foregut and tracheobronchial tree during embryological development. This encompasses congenital pulmonary airway malformations, pulmonary sequestrations, foregut duplication cysts (neuroenteric cyst, bronchogenic cyst, enteric cyst).[6] Intra-thoracic cystic lesions pose a diagnostic as well as therapeutic challenge. The first modality of investigation is still the chest X-ray, AP and lateral views help to delineate the position within the chest.[6] Evidence of spinal abnormalities should raise the suspicion of neuro-enteric cysts arising from the thecal sac.[7] To further delineate the local anatomy of the cyst, a CT or MRI is useful.

Early resection of intra-thoracic cysts has been advocated due to potential complications. Respiratory distress occurs with increasing size of the cyst resulting in compression of the bronchi and lung parenchyma;[2,8] infection and dysplasia have been described;[2,5,9] haematemesis and haemoptysis secondary to acid production by ectopic gastric mucosa.[2,10-13] Thoracosopic excision has been described but often difficult if mass is very large.

In conclusion, we suggest that BPFM should be included as a differential diagnosis during assessment of a mediastinal mass.

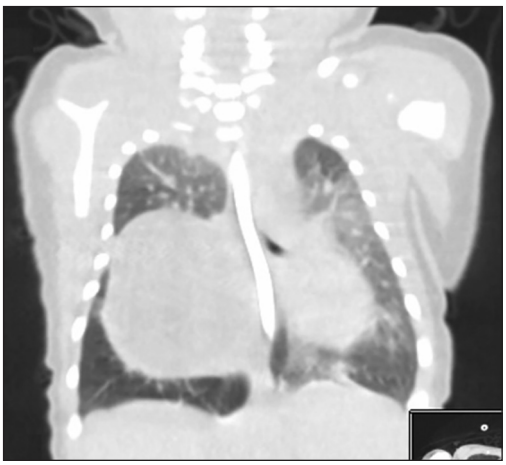

Figure 2: Coronal computed tomography with contrast demonstrating right intra-thoracic mass causing mediastinal shift to the left and obstructive emphysematous changes of right lower and middle lung

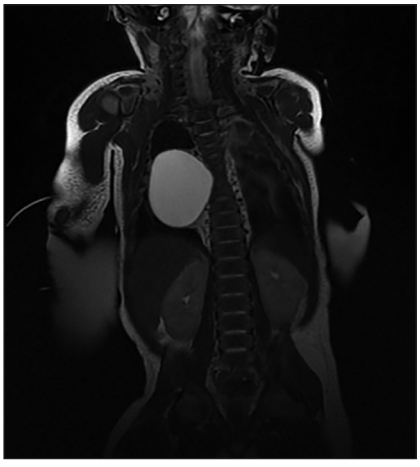

Figure 3: Magnetic resonance imaging, coronal section-demonstrating cystic mass in right hemi-thorax

Computed tomography or magnetic resonance imaging is necessary for pre-operative planning. Patient and cyst anatomy will determine the mode of excision; open or thoracoscopic.

Histopathological examination is mandatory for final confirmation of this diagnosis.

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