Online Case Report

Multiple antenatally diagnosed foregut duplication cysts excised and the value of thoracoscopy in diagnosing small concurrent cysts

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A case report of a female neonate referred with antenatally diagnosed cystic lesions within the right hemithorax and under the left hemidiaphragm is presented to highlight the ease with which these lesions can be resected in an asymptomatic post-natal infant with minimally invasive surgery. The diagnostic and cosmetic benefits of minimally invasive surgery are also demonstrated. While excising the documented thoracic cyst, another small cyst was identified adherent to the right main bronchus. An elective laparoscopic excision of the abdominal cyst was also successful. A brief review of the complications and treatment of foregut duplication cysts is detailed.

Keywords: Foregut duplication cysts – Antenatal ultrasonography – Video-assisted thoracoscopy – laparoscopy

Foregut duplication cysts are rare but well documented. They are known to be the cause of a variety of symptoms, some of which may not present until later childhood, or even beyond. Morbidity is associated with complicated and infected foregut duplications and their resection can be a surgical challenge. Fetal sonography in recent years has been achieving antenatal diagnosis of cystic lung lesions with increasingly accuracy.¹ Postnatal radiological investigations help to localise the lesion and give clues to its pathology in most instances. Our unique case with antenatal diagnosis of intrathoracic and intra-abdominal cystic lesions is reported to highlight post-natal assessment of these lesions and its preferred surgical management. The report discusses the optimal management of the foregut duplication cysts and the advantages of minimally invasive resection techniques.

Case report

A full-term, female neonate was referred to us with an antenatal ultrasound diagnosis of persistent cystic lesions within the right hemithorax and under the left hemidiaphragm. Postnatally, she was entirely asymptomatic but found to have a palpable cystic mass in the left upper quadrant. She had a normal chest X-ray and an ultrasound confirming a large abdominal cyst in close proximity to stomach and spleen. Computed tomogram (CT) scanning with intravenous contrast showed a tubular cyst in the right thoracic cavity running parallel to the oesophagus to the diaphragm and a large cyst posterior to the stomach extending from the diaphragm superiorly, spleen laterally, and extending across the midline (Fig. 1d). Post-natal contrast swallow showed

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Figure 1 Histopathological slides showing a variety of epithelia within the resected cysts: (a) simple columnar epithelium with cilia at places and a smooth muscle wall in the cyst in close proximity to the right main bronchus; (b) immature oesophageal squamous type epithelium with residual cilia within the cyst adjacent to the oesophagus; and (c) gastric-type mucosa and smooth muscle within the cyst adjacent to stomach. (d) Computed tomogram showing a lower oesophageal duplication cyst in the right hemithorax and a second subdiaphragmatic cyst on the left behind the stomach and medial to the spleen.

characteristic indentations of the cyst on the oesophageal and stomach walls suggestive of foregut duplications.

Elective excision of the thoracic cystic lesion was successfully carried out at 20 months of age by video-assisted thoracoscopy using three 5-mm ports. While excising the tubular cyst parallel to the oesophagus, another small cyst (0.5 cm in diameter) was identified adherent to right main bronchus and separate to the oesophageal cyst. This small cyst was not diagnosed on pre-operative investigation and would have been easily missed at an open surgical resection. The histopathology confirmed these cysts

as foregut duplication lined by characteristic epithelium and outer muscle walls as a bronchogenic cyst (Fig. 1a) and an oesophageal duplication cyst (Fig 1b).

The abdominal cyst was excised subsequently by laparoscopy. This minimally invasive procedure was entirely uneventful and was carried out using three ports and achieving dissection behind the stomach through the lesser sac. Histopathology of this cyst revealed it to be a gastric duplication cyst (Fig. 1c). The child was discharged home the next day after both the procedures and had very minimal analgesic requirements. She has remained

asymptomatic after 2 years of follow-up with an excellent cosmetic result. Her growth and development are entirely normal.

Discussion

Foregut duplication cysts are well-recognised congenital malformations that may remain asymptomatic. The most common symptoms of dysphagia, acute respiratory distress or chest pain will depend upon the location and size of the intrathoracic cyst.² Late presentations in complicated intrathoracic cysts with infection or bleeding within the cysts could be related to either compression or their spontaneous rupture with fistulation into neighbouring structures such as the bronchus, oesophagus, pericardium or pleura.^{1,2} Sudden infant deaths have been reported in association with bronchogenic cysts.² Chronic symptoms such as asthma-like illness, failure to thrive, anaemia and dysphagia have been reported.^{1,2} Our case demonstrates that an elective excision is safe and can be achieved with minimal morbidity with minimally invasive techniques. Therefore, an elective resection of foregut duplication is advocated and is generally accepted surgical management.^{3,4}

With the advances in antenatal imaging, cystic lesions are now being diagnosed antenatally.⁵ Antenatally diagnosed intrathoracic or intra-abdominal cystic lesions are best investigated and delineated by a postnatal CT scan as invariably a plain chest X-ray fails to demonstrate such lesions. A contrast swallow delineates a characteristic impression and shift of the oesophagus caused by a cystic duplication.¹ The contrast swallow is indicated in cystic lesions situated in the posterior mediastinum.

The published literature suggests that these cysts are best managed by an early resection, as these cysts have a tendency to cause significant and often life-threatening complications.² Minimally invasive surgery offers low morbidity, speedy recovery and superior cosmetic results,³ and also reduces postoperative hospital stay and postoperative pain. The success rate of minimally invasive technique is higher in uncomplicated foregut duplications. We advocate this technique as the resection option of choice as it is well tolerated and gives better cosmetic results in young infants. Our unique case illustrates the benefits of antenatal detection, early investigations and successful elective excision by thoracoscopy and laparoscopy. The magnified vision and unhindered view offered by this technique in our case were helpful, in addition, to diagnose a small bronchogenic cyst, which would have been otherwise missed.

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