CASE REPORT

Oesophageal inflammatory paediatric chylothorax

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SUMMARY

Paediatric chylothoraces are rare, particularly outside the operative setting. Cases of spontaneous chylothorax are often demanding diagnostically and frequently associated with patient morbidity. We present a challenging case of paediatric chylothorax associated with inflammatory oesophageal perforation likely related to foreign body ingestion.

BACKGROUND

Spontaneous paediatric chylothoraces are an unusual entity. Non-surgical cases are typically associated with congenital abnormalities or thoracic malignancy. In a small cohort of patients the underlying pathology is not identified resulting in a diagnosis of idiopathic chylothorax. Oesophageal pathology has infrequently been described in the development of paediatric chylothorax and oesophageal inflammatory perforation-associated chylothorax has not previously been described.

CASE PRESENTATION

A 3-year-old boy, with no significant medical history, was referred from a peripheral centre with a 3-day history of lethargy, poor appetite and vague abdominal pain. Further questioning revealed worsening shortness of breath and urine output particularly over the previous 24 h period. Thoracic examination revealed asymmetrical chest wall movement with absent breath sounds affecting the left hemi-thorax. He was haemodynamically stable with a respiratory rate of 36 breaths per min and peripheral oxygen saturations of 97%. Chest X-ray revealed massive left-sided hydrothorax with marked midline shift. Of note, during subsequent thoracic CT scan an area of extraluminal gas was noted at the mid-oesophageal level with an adjacent area of low attenuation (figure 1) consistent with inflammatory perforation. No foreign body was identified.

TREATMENT

Urgent tube thoracostomy was performed with an initial volume of 250 mL of chylous pleural fluid (figure 2) drawn off prior to a drain clamp being applied. Fluid analysis revealed a triglyceride level of 26.3 mmol/L with a >80% mature lymphocyte fraction. The remaining fluid was drained at two hourly intervals and thereafter medium-chain triglyceride diet (via nasogastric tube for oesophageal protection) and octreotide therapy were initiated. Over subsequent days barium studies and flexible oesophagoscopy revealed a nodular lesion with central necrosis (figure 3) in the mid-oesophagus

Figure 1 Area of high attenuation, free thoracic contrast and adjacent free air.

with no further evidence of perforation. It was hypothesised that the central necrosed area may have been due to trauma from a button battery with consequent inflammation resulting in oesophageal perforation and compression of the thoracic duct. Biopsies confirmed evidence of a benign inflammatory process.

Despite maximal medical management massive thoracostomy losses of 600–1000 mL/24 h persisted over the following 10 days. With neutropenia, coagulopathy and weight loss placing the patient at high risk of morbidity and pleurodesis felt to offer an inferior outcome, a right-sided minithoracotomy and thoracic duct ligation was performed. Under direct vision a small area of scarring was noted at the mid-oesophageal level with no palpable masses identified.

OUTCOME AND FOLLOW-UP

A significant reduction in pleural losses was noted postoperatively and with continued improvement the chest drain was removed at day 8. This period

Figure 2 Chylous fluid.

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Figure 3 Nodular oesophageal lesion with central necrosis.

permitted for stepwise reduction of octreotide therapy, establishment of a low-fat oral diet and close observation for rebound chylous accumulation. The patient was discharged from hospital 3 days later and remained clinically and radiologically stable when reviewed 3 months postoperatively.

DISCUSSION

Paediatric chylothorax, the collection of chyle in the pleural cavity, is a rare process. It typically manifests as a result of underlying congenital anomalies, thoracic malignancy or most commonly post cardiothoracic surgery. Vomiting¹ and child abuse² have been reported as unusual causative factors. Furthermore, idiopathic chylothorax has infrequently been documented.^{3,4}

To our knowledge, paediatric chylothorax due to oesophageal inflammatory thoracic duct compression associated with foreign body ingestion has not previously been described.

Long intrathoracic courses expose the thoracic duct to a myriad of thoracic pathology. This makes the initial diagnosis difficult in these unusual cases. As noted in this case an inflammatory process may underlie. Mediastinal and hilar adenopathy due to underlying tuberculosis⁵ and sarcoidosis⁶ have been described to cause chylothorax. In addition, Kairamkonda⁷ links traumatic oesophageal perforation during the intubation of a neonate with the development of chylothorax. Raised intrathoracic pressures associated with vomiting has also been seen to compress the duct resulting in bilateral chylothorax.¹ Critically, child abuse must be considered prior to definitive diagnosis of idiopathic chylothorax.²

Many of these cases require urgent pleural decompression to relieve respiratory compromise. However, it is often the subacute diagnosis and medical management that prove most challenging to the treating physician. Continued high-output chylous drainage exposes these paediatric patients to a significant risk of immune compromise and malnutrition therefore prompt action is required.

Once the initial effusion is identified on chest X-ray chylothorax can be differentiated from alternative pathology by the aspiration of milky fluid from the pleural space. Chylothorax is confirmed by the presence of >1.1 mmol/L triglycerides, >1000 cells/ μ L and a lymphocyte fraction $>80\%$ ⁴ on laboratory

fluid testing. Differential exclusion is achieved with cytological, biochemical and microbiological analysis of blood and pleural fluid. This process can in turn be augmented by thoracic CT and in this case flexible oesophagoscopy.

Management involves conservative and invasive measures. Epaul *et al*³ describe a series of six idiopathic paediatric chylothoraces. They concluded that initial management should involve a medium-chain triglyceride diet with total parenteral nutrition initiated in those failing simple measures. Two cases required pleuroperitoneal shunting while somatostatin was trialled unsuccessfully in one case. Chylothorax stabilised in all patients. Paul *et al*⁸ report a 96% treatment success associated with thoracic duct ligation. Additionally, pleurodesis enjoyed an 83% success rate. Novel percutaneous ductal management has also been proposed as a potentially minimally invasive management option.

Oesophageal inflammation induced by foreign body ingestion may result in the development of paediatric chylothorax. In similar challenging cases we recommend close CT inspection of paraductal structures for causative inflammatory processes. In this case initial conservative management was unsuccessful, thus, persistent massive chylous output dictated surgical exploration was required for therapeutic purposes.

Learning points

- ▶ Oesophageal inflammation secondary to foreign body ingestion may underlie paediatric chylothorax.
- ▶ On thoracic imaging paraductal structures should be scrutinised for causative pathology.
- ▶ Acute management of massive chylothorax requires urgent thoracic decompression with subsequent institution of conservative measures.
- ▶ Surgical duct ligation is an effective option in resistant paediatric chylothorax.

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Competing interests None.

Patient consent Obtained.

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