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Iatrogenic diaphragmatic hernia in infants: Potentially catastrophic when overlooked☆

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ABSTRACT

Acquired diaphragmatic hernias are a rare occurrence. They can result from blunt, penetrating or inadvertent iatrogenic injury. When overlooked they can potentially be catastrophic. We report a case of iatrogenic diaphragmatic hernia in a six-month old infant presenting with acute respiratory distress as a result of strangulated bowel herniating into the left hemithorax caused from a traumatic chest tube insertion in the neonatal period.

The vast majority of diaphragmatic hernias encountered by pediatric surgeons are congenital, with a reported prevalence rate of 1 to 3000–5000 live births [1]. Acquired or iatrogenic diaphragmatic hernias on the other hand are probably under reported in the pediatric population, with only a single case report describing two uneventful cases after pericardial drainage following cardiac surgery [2]. Here we report a case of an incidental finding of an acquired diaphragmatic hernia resulting in strangulated small bowel following a traumatic chest tube that was inserted six months prior to presentation.

1. Case report

A six month old ex-premature baby boy, born at 29 weeks of gestation, who was scheduled for an elective left inguinal hernia repair, presented in the early hours of the day for his elective procedure with fever, lethargy and difficulty breathing, without any reported emesis. Physical exam showed patient was pyrexic with evidence of subcostal recession with decreased air entry on the left side. Labs revealed significant leukocytosis 43.9 and hyperkalemia, 6.8 mmol/L. An arterial blood gas was consistent with respiratory acidosis, with a lactate level 6.1. An urgent chest x-ray demonstrated a complete opacification of the left hemithorax (Fig. 1).

Patient's clinical condition deteriorated in the emergency department and was intubated. After providing the necessary resuscitation and stabilizing the patient, a non-contrast CT scan of the chest was obtained. The scans were initially read as massive pleural effusions with mediastinal shift (Fig. 2A). A percutaneous chest drain was then inserted by interventional radiology (Fig. 2B).

A chest x-ray was obtained post percutaneous chest tube insertion and did not show any improvement. Pediatric surgery was consulted for diagnostic thoracoscopy for evaluation of the left hemithorax for presumed multiple loculated pockets of empyema (Fig. 2B). Intra-operatively, multiple loops of ischemic looking bowel were seen during the diagnostic thoracoscopy. The operation was converted to a laparotomy confirming the diaphragmatic hernia (Fig. 3A) with the strangulated small bowel, 75.5 cm, herniating through a 1 cm defect (Fig. 3A–C). The defect had to be extended to help reduce small bowel into the abdominal cavity. The strangulated small bowel was resected, which started five cm from the ileocecal valve extending proximally, and an end-ileostomy was fashioned. Two hundred and fifty cm of healthy small bowel were

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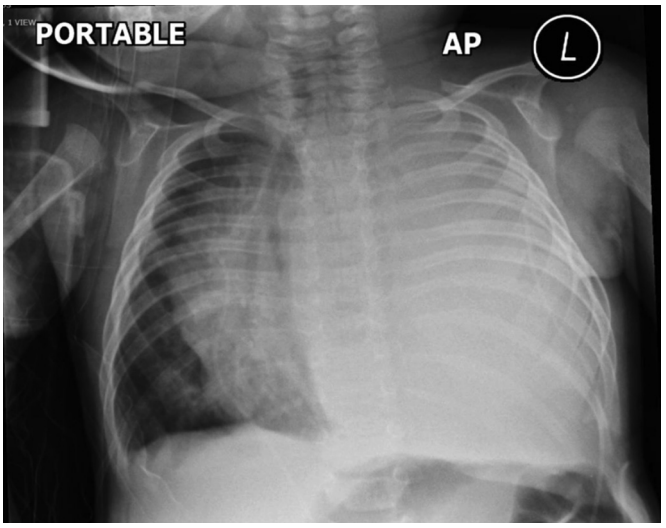

Fig. 1. Complete 'white-out' left lung with mediastinal shift.

left behind with an intact ileocecal valve. No diaphragmatic hernial sac was seen intra-operatively with the spleen and liver looking normal. The diaphragmatic defect was closed primarily with non-absorbable interrupted sutures. Patient had an uneventful post-operative recovery.

When investigating the possible cause of the hernia, it transpired that the patient had a stormy post-natal course developing spontaneous tension pneumothorax on day of life (DOL) 1 (Fig. 4A). A 10 French chest tube was inserted which did not resolve the pneumothorax, thus a second 10 French chest tube was inserted which was traumatic resulting in life threatening hemorrhage necessitating transfusion with packed red blood cells, fresh frozen plasma and platelets (Fig. 4B,C). The bleeding spontaneously stopped and the chest tube was subsequently removed on DOL 9 with no evidence of pneumothorax. All subsequent chest x-rays taken prior to hospital discharge were normal, with the last chest x-ray taken four months prior when the patient presented to the emergency room with bronchiolitis (Fig. 4D).

2. Discussion

The vast majority of diaphragmatic hernias are congenital, and is rarely associated with a number of different syndromes including, Beckwith-Weidman, Goltz Syndrome, and Denys-Drash Syndrome [3]. Bockdaleck diaphragmatic defects remain the commonest type of congenital diaphragmatic hernia, with the size of the diaphragmatic defect being closely correlated with the number of other embryologic defects, most notably cardiovascular malformations [4].

The majority of the handful of iatrogenic diaphgramatic hernias reported in the literature are in adults, with only one previously reported case report in the pediatric population [2]. Bettolli et al. [2] reported two cases of incarcerated bowel following peri-cardial drain placements. Here we report a case of iatrogenic diaphragmatic hernia resulting in strangulated bowel. This was found during a diagnostic thoracoscopy for presumed empyema with loculations on pre-operative imaging. Although this is the first reported case of iatrogenic diaphragmatic injury secondary to a traumatic chest tube insertion in the pediatric population, we believe that this might be under reported. The current case highlights two important points. Firstly, the importance of correctly placing chest tubes in premature neonates. Secondly, the need to have a high index of suspicion for an iatrogenic injury to a viscus or diaphragm after difficult chest tube insertion. Accidental insertion of chest tubes into the liver organ has been previously reported with right-sided CDH's associated with pleural effusions [5–8]. Thoracoscopy is a valuable tool in evaluating chest pathologies.

There are several different indications for inserting tube thoracostomies in the pediatric population. In the current case, the chest tube was inserted due to spontaneous pneumothorax in a preterm neonate, where the reported incidence of symptomatic pneumothorax in all live births is 0.08% [9] and 5%–7% in infants with a weight $\leq$1500 g [10,11]. In this current case report the patient had developed tension pneumothorax with respiratory distress (Fig. 4A). Chest tube thoracostomies can be associated with significant discomfort and, although rare, potentially life threatening complications [12]. Complications associated with chest tube placement include a malpositioned chest tube, infection, trauma resulting in lung laceration [13], hemorrhaging of a major vessel or puncture of a viscus with the path of the tube [12], phrenic nerve paralysis [14] and chylothorax [15]. It has recently been suggested that these stiff bore chest tubes can be replaced with 8.5 French soft pleural catheter, due to it being a less intrusive technique using the Seldinger technique, associated with less pain and are equally effective compared to large-bore stiff tubes [12,16]. Martin et al. [12] revealed in their series of 23 pediatric patients requiring soft pleural catheter insertion of which seven were neonates, that seven were indicated for spontaneous pneumothorax and were all successfully resolved. However, the majority of complications in their series were related to tube dislodgment or blockage. The three notable complications recorded which required the soft pleural catheter to be adjusted or replaced due to unacceptable placement included, firstly, subcutaneous placement of the catheter, secondly, too distal placement of the catheter and lastly, hemorrhage secondary to injury to the

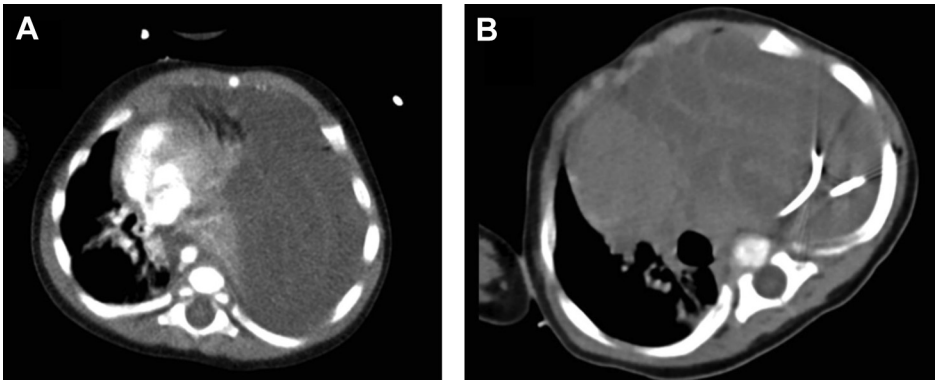

Fig. 2. (A) Massive left sided pleural effusion. (B) Percutaneous drain placed in left chest by interventional radiology.