Cardiology in the Young

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Iatrogenic diaphragmatic hernia in an infant following cardiac surgery: the culprit in a case of unresolved respiratory distress: Case report and review of the literature

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Brief Report

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Abstract

Acquired diaphragmatic hernia is a rare complication of pediatric intervention or surgery. In this study, we report an infant with iatrogenic diaphragmatic hernia following neonatal complex congenital cardiac surgery, and then we review the associated literature.

Acquired diaphragmatic hernia is a rare finding in children and is most often the result of a blunt or penetrating abdominal trauma that compromises the integrity of the diaphragm and allows the bowel to enter the chest cavity.[1,2] Case reports of iatrogenic diaphragmatic hernia are scarce in the literature, but diaphragmatic hernia is a recognised complication of hepatic and upper gastrointestinal surgery, sub-xiphoid pericardial and pleural drain placement, and classic median sternotomy.[3–7] We present a case of iatrogenic diaphragmatic hernia following complex neonatal congenital cardiac surgery with a median sternotomy, pleural chest tube, and pericardial drain, whose primary clinical manifestation was respiratory distress; we then review the associated literature.

Case report

A 3270 gram female neonate with a prenatal diagnosis of non-mirror image dextrocardia, D-transposition of the great arteries, tricuspid atresia, and hypoplastic aortic arch with coarctation of the aorta underwent a modified Norwood stage 1 operation at 7 days of life. The patient left the operating room with a closed chest and a sub-xiphoid chest tube that was subsequently removed on post-operative day 5. The post-operative course was complicated by a large posterior, loculated pericardial effusion treated with placement of a 5-French pigtail catheter via the sub-xiphoid approach, as well as left vocal cord paralysis leading to placement of a gastrostomy tube. The patient remained in the cardiovascular ICU for a total of 8 weeks and was discharged home with a pulse oximeter monitor as per the Johns Hopkins All Children's Hospital Heart Institute protocol for inter-stage monitoring of patients with functionally univentricular cardiac anatomy. Three days following discharge, the patient presented to the emergency department with intermittent bradycardia, tachypnoea, and oxygen saturations in the low 70s (expected saturations being >80%). Following a 15-day admission with a negative infectious workup and multiple reassuring chest radiographs and echocardiograms, the patient was discharged home. The patient was followed frequently as an outpatient because the family had poor compliance with the inter-stage monitoring protocols, including pulse oximetry and daily weights. Two weeks later, at the outpatient clinic, the parents reported a 2-day history of cough, intermittent wheezing, nasal congestion, and fever of 101.4°F. On examination, the patient was afebrile with oxygen saturations of 88% on room air, but examination was significant for sub-costal retractions and rhonchi on auscultation. The patient was re-admitted to the cardiovascular ICU, tested positive for rhinovirus, and discharged after 48 hours of observation. Nine days following discharge, the patient re-presented to the emergency department with a cough, increased work of breathing, fever of 101.1°F, and a new complaint of daily emesis. However, the family reported reassuring oxygen saturations since her previous hospital discharge. Chest radiography showed bilateral pulmonary infiltrates that were most prominent in the right lower lobe. The patient was again admitted for observation and treatment with intravenous antibiotics, and was discharged 3 days later on oral amoxicillin. Over the subsequent weeks, the patient continued to have unresolved tachypnoea with retractions, diaphoresis, and increased frequency of emesis with feeding. The patient was then enrolled in a medical daycare, which reported to the cardiology

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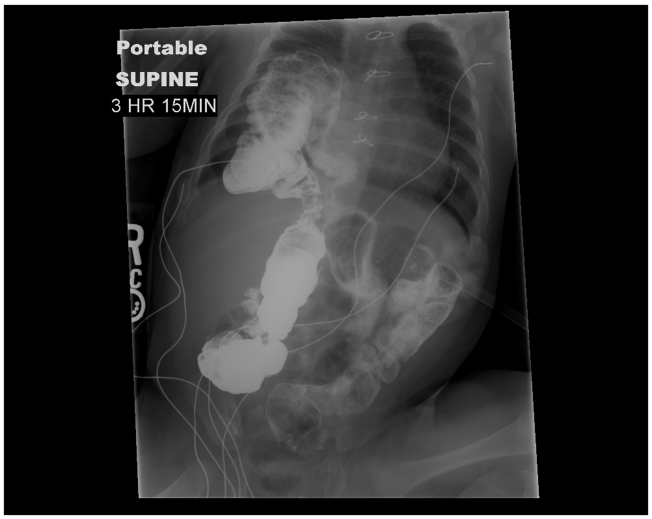

Figure 1. Upper gastrointestinal contrast study with small bowel follow through showing contrast-filled bowel within the right hemithorax.

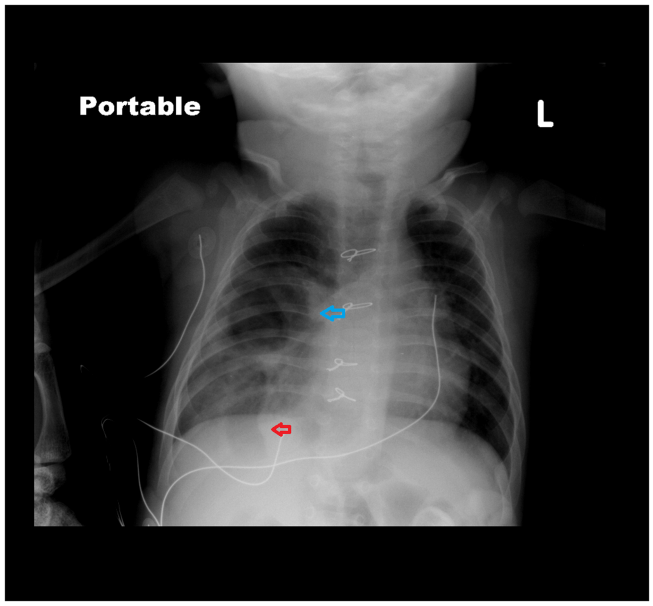

Figure 2. Chest radiograph performed three months after Norwood Stage 1 Operation showing bowel-like tubular lucency traversing the right hemi-diaphragm (red arrow) and right upper lobe lucency (blue arrow).

service that the patient had multiple episodes of decreased oxygen saturations each day. Given the history of vocal cord paralysis, frequent emesis, and worsening respiratory status, the patient was re-admitted to the cardiovascular ICU for evaluation of possible aspiration pneumonia. The patient's chest radiography continued to show opacification of the right lower lobe, and the care team continued to suspect aspiration pneumonia despite a negative pH probe. An upper gastrointestinal contrast study with small bowel follow through revealed contrast-filled bowel within the right hemithorax consistent with a right-sided diaphragmatic hernia (Fig 1). At 4 months of age, the child underwent surgical repair of the diaphragmatic hernia via a midline incision of the chest that extended inferiorly to the former chest tube site. Bowel was reduced from a small hole in the diaphragm just to the right of midline. The bowel had herniated directly through this small diaphragmatic defect that was suspected to be related to the chest tube from the Norwood stage 1 operation. The patient recovered quickly and was discharged home on room air with feedings via the gastrostomy tube. The patient underwent a successful stage 2 bidirectional cavopulmonary anastomosis at 5 months of age and a complete stage 3 extra-cardiac Fontan operation at 3 years of age.

A retrospective review of the patient's chest radiographs prior to neonatal cardiovascular surgery are consistent with an intact diaphragm. In contrast, the post-operative chest radiographs performed when the patient had symptoms of shortness of breath and vomiting reveal air-fluid levels in the right lower lobe (Fig 2). This patient had multiple echocardiograms during each hospital admission. A retrospective review of these studies does not reveal any discontinuity in the diaphragm or any clear evidence of bowel in the chest cavity. The cardiac silhouette never appeared displaced into the left chest nor was there any evidence of bowel herniation into the pericardium or cardiac tamponade.

Discussion

Paediatric cardiac surgery predisposes the patient to multiple, well-described complications, including bleeding, infection, effusion, arrhythmia, and issues with feeding.[8] An iatrogenic injury to the diaphragm following a cardiovascular procedure is infrequently reported in the paediatric literature and requires a high index of suspicion to diagnose. In this case report, the patient presented with respiratory symptoms, fevers, and radiographic findings that all pointed to an infectious ideology. It was not until the patient developed vomiting that an upper gastrointestinal contrast study confirmed the diagnosis of iatrogenic diaphragmatic hernia.

Previously, five paediatric cases of iatrogenic diaphragmatic hernia that are not associated with upper gastrointestinal surgery are found in the literature.[3,4,7,9] Bettolli et al described two infants that underwent cardiovascular surgery with pericardial drain placement, who were diagnosed at 6 and 12 months of age with herniation of the peritoneal contents into the chest via an iatrogenic diaphragmatic hernia.[3] Among the two infants, one patient presented with cough and vomiting, and the other was diagnosed incidentally during laparoscopic-assisted gastrostomy tube placement. Ashour et al report a 6-month-old infant who previously underwent complex neonatal cardiovascular surgery via median sternotomy with placement of a sub-xiphoid chest tube and a mediastinal drain. This infant was diagnosed with an iatrogenic diaphragmatic hernia by chest radiography after presenting with cough and poor feeding.[7] Panda et al also describe a 6-month-old who previously underwent complex neonatal cardiovascular surgery and who presented with shortness of breath.[9] Echocardiography revealed a mass in the pericardium and findings of cardiac tamponade. Subsequently, an intra-operative diagnosis of diaphragmatic hernia into the pericardium was made. Finally, El-Gohary et al describe a 6-month-old ex-29-week pre-term infant who presented with respiratory failure, sepsis, and complete opacification of the left hemithorax on chest radiography.[4] This patient was diagnosed intra-operatively with ischaemic herniated bowel in the chest via an iatrogenic diaphragmatic hernia. Of interest, this patient had not undergone cardiovascular surgery, but did have a history of

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a tension pneumothorax on day 1 of life. At that time, two chest tubes were placed, the second of which resulted in an intra-abdominal haemorrhage from a suspected injury of the abdominal organs.

Speculation of the cause of the iatrogenic injury to the diaphragm varied in these reports. Some conclude that herniation was secondary to extension of the sternotomy incision below the xiphoid process in order to facilitate exposure during neonatal cardiovascular surgery, with a resultant breach of the peritoneum.[3,7,9] However, the patient described by El-Gohary et al did not have a sternotomy. All of the previously described cases, including our case, did have sub-xiphoid drains placed as part of their postoperative care. On direct inspection, the surgical team in our case confirmed the location of the diaphragmatic defect as likely related to the prior chest tube placement. Bettolli et al speculated that with time, the intra-thoracic pressure gradient during times of Valsalva may increase the size of the defect and allow for herniation of the peritoneal contents into the pleural space or pericardial cavity. Their hypothesis supports our patient's clinical presentation of symptoms that were remote from the immediate post-operative period and the discontinuation of chest tubes and drains. Over some period of time, the area of prior diaphragm breech allowed the bowel to migrate into the chest cavity. In our case, the time period from drain removal to clinical presentation with new onset daily emesis was approximately 3 months. In consideration of the previously reviewed cases in the literature and this case, a high index of suspicion and careful radiographic assessment is needed to make the diagnosis of iatrogenic diaphragmatic hernia. The majority of patients presented with non-specific respiratory and gastrointestinal symptoms and the diagnosis was made by chest radiograph several months following the interventional procedures. In review of the chest radiographs in this case, perhaps the findings of a right lower lobe infiltrate should have been suspect for a bowel herniation.

In summary, a breach in the integrity of the diaphragm is a rare complication of paediatric cardiothoracic interventions, including median sternotomy, chest tube placement, and pericardial drain placement. A high index of suspicion is necessary to make this diagnosis, and repair can be live-saving.

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Conflicts of Interest. None.

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