

Open Repair of Pediatric Aortoenteric Fistula from A Remote Gastric Transposition in Congenital Esophageal Atresia: A Multidisciplinary Approach

Keywords: Esophageal Atresia; Aortoenteric Fistula; Ulcer; Pediatric Surgery; Vascular Surgery

Abstract

A 12-year-old male with history of long gap esophageal atresia with a gastric transposition at one year of age presented with multiple episodes of hematemesis. He recently had been prescribed high dose NSAIDs for pericarditis. He underwent multiple endoscopic cauterizations of a large gastric ulcer and despite this required MTP. CTA was obtained showing hypoattenuation of the gastric conduit along the aorta near the area that was cauterized. The patient underwent a left-thoracotomy and gastrotomy. Once hematoma was evacuated, a large pulsatile bleed was encountered. Pressure was held and control of the aorta was obtained. The gastric conduit was dissected off the aorta, revealing a large defect. The gastric conduit was repaired, the aorta was repaired with bovine pericardium and pleural flap was placed. On POD 8 a swallow study demonstrated no leak and the patient was discharged on POD 15. Outpatient follow-up CTA demonstrated an intact repair.

Introduction

Aortoenteric fistulas are a fairly rare cause of gastrointestinal bleeding and if seen, it is typically seen in adults [1,2]. They are historically separated into primary aortoenteric fistula and secondary aortoenteric fistulas [1,2]. Primary AEF is defined as a spontaneous communication between native aorta and any portion of the gastrointestinal tract resulting from compression against an abdominal aortic aneurysm [1, 2]. Secondary AEF is defined as typically resulting from a complication following vascular surgery and rarely GI surgery [1-3]. When noted to be a sequela of GI surgery, AEF characteristically presents 2-3 weeks following the operation, typically an esophagectomy or esophago-gastrectomy at the anastomosis site [4-6]. Patients typically present with massive hematemesis and regardless of type of AEF, the mortality rate for AEF is high, with it reaching as high as almost 60% with an in hospital mortality rate almost 30% [7]. There is little to no literature demonstrating AEF in the pediatric population. We present a case of a 12-year-old male presenting with a thoracic gastro aortoenteric fistula following a gastric transposition for long gap esophageal atresia when he was 1-year-old.

Case report

The patient is a 12-year-old male with history of long gap esophageal atresia with a gastric transposition at one year of age presented with multiple episodes of hematemesis. Interval history involved primary care physician diagnosis of pericarditis associated



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Submission: 11 January, 2024

Accepted: 07 February, 2024

Published: 12 February, 2024

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with COVID vaccination, requiring high dose ibuprofen one month prior to his presentation for hematemesis. Four weeks following the diagnosis of pericarditis, he experienced hematemesis for which three attempts at endoscopic cauterization of a large gastric ulcer were attempted and unsuccessful. Computed tomography angiography (CTA) was obtained that showed hypoattenuation of the gastric conduit along the aorta near the area that was cauterized (Figure 1).

Due to failure of non-operative management and concern for AEF, operative intervention with pediatric surgery was indicated. Extensive operative planning was performed including resuscitative line placement, preparation for possible cardiopulmonary bypass, and vascular and cardiac surgery on standby. The patient underwent a left thoracotomy and gastrotomy. Once hematoma was evacuated from the conduit, a large pulsatile bleed was encountered. Pressure was held and proximal and distal control of the aorta was obtained. Vascular surgery was consulted emergently intraoperatively and the gastric conduit was dissected off the aorta, revealing a large defect



Figure 1: CTA demonstrating hypoattenuation of the gastric conduit along the aorta near the area that was cauterized with concern for aortoenteric fistula.

and an aortoenteric fistula (Figure 2). Proximal and distal control of the aorta was obtained using umbilical tape. A 20F chest tube was then used as a shunt and the aorta was clamped. Pediatric surgery worked to separate the gastric conduit from the aorta, while vascular surgery repaired the aorta with a rifampin soaked bovine patch sewed in using 4-0 prolene suture. Upon removal of the gastric conduit from the aorta, a large ulcer in the posterior aspect of the stomach was discovered and repaired with several figure of eight 3-0 PDS and the gastrotomy was closed with a blue load 60mm stapler. Cardiothoracic surgery was called intraoperatively to perform a thoracic pleural flap placed between the gastric and aortic repair as a buttress.

Post operatively, he was resuscitated in the pediatric intensive care unit for several days. Post-operative medications included IV antibiotics for a 6-week course, twice daily proton-pump inhibitor, and Plavix which was started on post-operative day (POD) three. On POD 8 we performed a swallow study demonstrating no leak and diet was advanced (Figure 3). He was discharged on POD 15 with an outpatient follow-up with pediatric surgery and vascular surgery and progressed well.

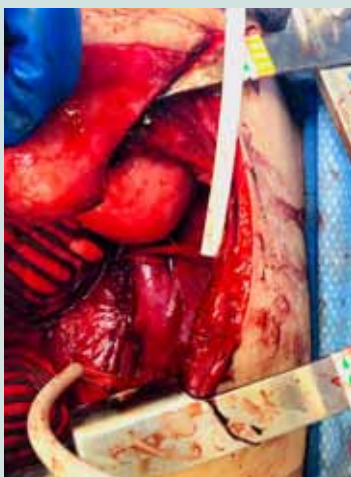


Figure 2: Intra-operative findings demonstrating the aortoenteric fistula tract.

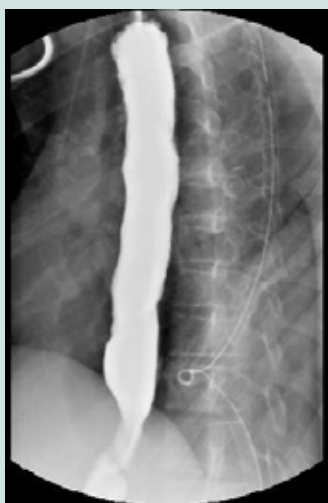


Figure 3: Post-operative swallow study demonstrating intact repair.

Discussion

Aortoenteric fistulas are rare, however if found they are typically in the adult population predominantly associated with aortic aneurysms or following vascular operations [1, 3, 7, 8]. Non-aneurismal aortoenteric fistulas are extremely rare, but have been reported in adults to be associated with carcinomas, tuberculosis, abscess, radiation, or duodenal ulcers [8]. The presentation of these is usually with gastrointestinal bleeding, and if from an operative cause, typically within one month postoperatively [4]. Management in the adult population often consists of endovascular stent placement and resection and reconstruction with intestinal reconstruction [6,7]. Our case is unique in many ways, our patient is a 12-year-old male without history of vascular disease or surgery who presented with gastrointestinal bleeding in the form of hematemesis 11 years post operatively. He was prescribed high dose ibuprofen one month prior to his episodes of hematemesis which almost certainly led to the development of the gastric ulcer found intraoperatively eroding in the aorta. His unique anatomy following the gastric pull through likely contributed to the ease of development of the fistula from the gastric ulcer. Our patient was trial managed non-operatively multiple times with failure leading to hemorrhagic shock. We would recommend early involvement of a pediatric surgery team when dealing with unique anatomy and continued hematemesis following endoscopic intervention.

To our knowledge, this is the first case of this nature in pediatrics and it presented several challenging components. Our institution is fortunate to have all needed available specialties for adequate operative repair. Obtaining proximal and distal control in this instance was crucial, we were able to use a chest tube as a shunt to avoid prolonged ischemic time. Ideally, the aorta would have been repaired primarily, however the ulcer site tissue was extremely friable and thus sutures did not hold. Rifampin soaked bovine patches have shown good results in adult literature for vascular repair, and we would recommend that for treatment should primary repair not be attainable. Removal of the ulcerated gastric tissue and repair of the



Figure 4: Outpatient CTA demonstrating intact repair of the aortoenteric fistula.

ISSN: 2332-4139

area is crucial in this operation as well, and we were able to primarily repair this without evidence of leak. In a hostile field, the worry would be that this would recur for this patient, which prompted us to perform the thoracic pleural flap to act as a barrier between the stomach and the newly repaired aorta, which is crucial in this case. He recovered well postoperatively and has close follow up surveillance with outpatient CTA demonstrating an intact repair (Figure 4). We would recommend follow up imaging with CTA's at 3 months, and one year due to the serious nature of this aortoenteric fistula and his unique anatomy.

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