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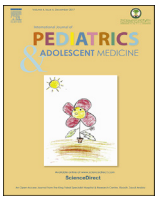

Sengstaken-Blakemore Tube: an extra mile

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ABSTRACT

Upper gastrointestinal bleeding (UGIB) in children has multiple etiologies but fortunately is not encountered commonly by pediatricians. Aorto-esophageal fistula (AEF) in children is a rare cause of UGIB and it is mainly secondary to accidental ingestion of foreign bodies, particularly disc batteries, or after cardiothoracic surgery. In this study, we report a case of a 3-year-old child who developed de novo AEF with no prior injury to the esophagus. The child presented with massive UGIB leading to hypovolemic shock, acute kidney injury, and cardiac arrest. The torrential bleed was controlled using a Sengstaken–Blakemore Tube (SBT), which allowed urgent chest CT angiography as well as subsequent thoracotomy and repair of the fistula Unfortunately, the child succumbed to repeated cardiac arrests secondary to the renal injury and severe acidosis. This case highlights the need for the early recognition of massive UGIB in children and the requirement to make appropriately sized SBTs available in all pediatric gastroenterology units.

1. Introduction

Upper gastrointestinal bleeding (UGIB) in children is a problem that is encountered infrequently by pediatricians. It is estimated to affect 11.2 per 10,000 of all pediatric discharges in the United States [1] and about 6% of children in pediatric intensive care units (PICUs) [2]. The mortality rate of 2% is relatively high [3].

The causes of UGIB in children are related to the age of the child. In children beyond toddler age, the most common causes include Mallory–Weiss syndrome, esophageal laceration, gastritis, and variceal bleeding [4]. Variceal bleeding usually involves a large bleed with potentially life threatening consequences and it has a reported frequency of 1:200,000 in Canadian children where liver transplant is available [5]. Variceal bleeding has a mortality rate of up to 19% within the following 35 days [6].

Aorto-esophageal fistula (AEF) is an extremely rare problem that is mostly fatal and it is not commonly mentioned as an etiology for upper gastrointestinal bleeding in children. AEF in children is attributed mainly to the ingestion of foreign bodies, particularly disc batteries [7]. In adults, thoracic aortic aneurysms are the most common causes of AEF [8]. Primary AEF was reported previously in a 2-year-old child who survived and the AEF was not caused by tuberculosis [9].

In this study, we report a rare case of large AEF in a 3-year-old child with no underlying cause, which was controlled using a Sengstaken–Blakemore tube (SBT) to allow definitive surgery.

2. Case report

A 3-year-old female with no prior medical history presented to her local hospital with a 2-week history of intermittent hematemesis in small amounts. The mother reported melena once previously and a bout of hematemesis with a large volume immediately prior to admission. On examination, she was in compensated shock with no evidence of icterus or signs of chronic liver disease. Her hemoglobin was 5 g/dl with normal platelets, coagulation profile, and liver functions test. The child was transfused urgently with packed red blood cells, stabilized, and referred to the Royal Hospital (RH), Oman, for tertiary care management.

In the RH, she went into florid hypovolemic shock with massive hematemesis and required intubation with large doses of inotropes in addition to 18 units of packed red blood cells, factor VII, and fresh frozen plasma. Her blood pressure did not stabilize despite these measures and she went into cardiac arrest. She was revived successfully but developed acute kidney injury. Diagnostic esophagogastroduodenoscopy (EGD) was performed urgently in the

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PICU, which indicated fresh bleeding from the middle third of the esophagus, with normal mucosa in the stomach and duodenum. No lesions could be visualized as responsible for the large bleed in the area. A size F18 Sengstaken–Blackmore tube (SBT) was inserted 4 hours after the initial bleed, which allowed immediate control of the bleeding and blood pressure stabilization. A peritoneal dialysis catheter was urgently inserted. CT angiography (CTA) of the chest detected an abnormal connection between the descending aorta and mid-esophagus, which was tamponaded using the SBT balloon (Figs. 1 and 2).

Urgent thoracotomy was performed 12 hours after the first massive hematemesis and 8 hours after control of the bleed using the SBT. EGD detected a fistula measuring 8–10 mm between the antero-medial aspect of the aorta connecting to the posterio-lateral wall of the esophagus. An additional posterior-medial esophageal wall tear was noted at the site of the SBT.

The AEF was ligated and the aortic defect was repaired using a bovine pericardial patch with complete hemostasis. Both the esophageal fistulous opening and the esophageal tear were repaired.

Histopathological analysis of tissue from the site of the fistula detected granulation and inflammatory changes with iron deposition, but a granuloma or foreign body was not identified.

Post-operatively, the child experienced severe acidosis, with cardiogenic shock and repeated cardiac arrests, and unfortunately was not revivable.

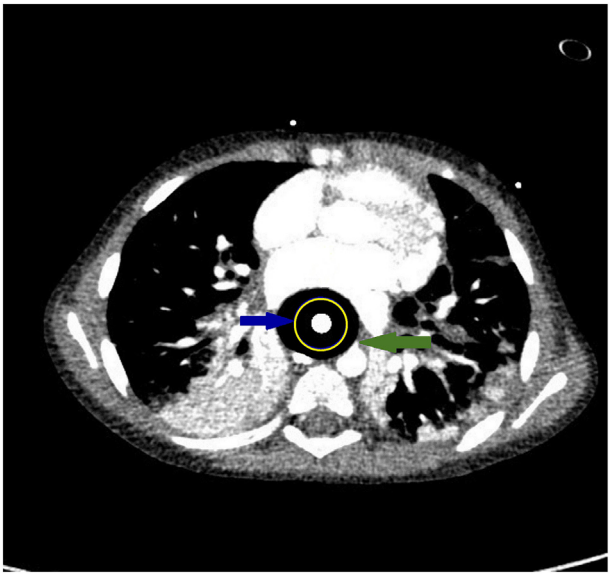

Fig. 1. Axial contrast-enhanced CT of the chest showing a linear enhanced structure arising from the anterior mid-thoracic aorta, where the "arrowhead" represents the aorto-esophageal fistula. Note the inflated Sengstaken–Blakemore tube within the mid-esophagus, as indicated by the "arrowed circle".

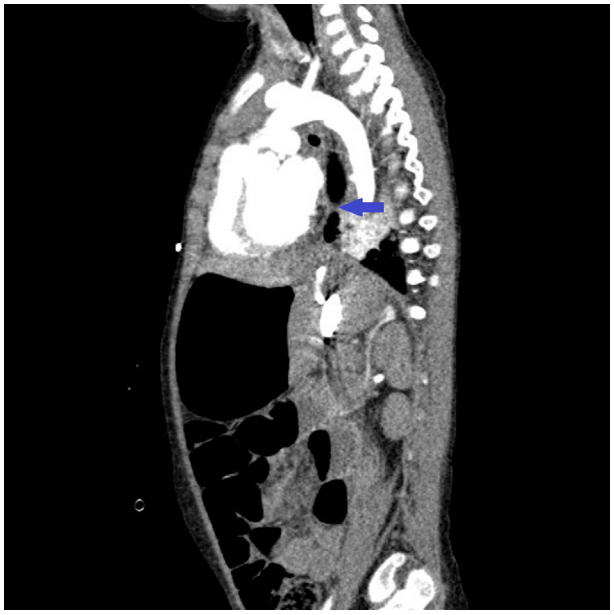

Fig. 2. Sagittal reformat based on contrast-enhanced CT of the chest indicating a linear enhanced structure arising from the anterior mid-thoracic aorta where the "arrowhead" represents the aorto-esophageal fistula.

3. Discussion

AEF is an abnormal communication between the aorta and esophagus. Massive UGIB causing hemodynamic instability is rare in children and should always suggest the possibility of variceal bleed as well as AEF. AEF should be considered even in the absence of any history of foreign body, surgery, or trauma to the child. A thorough examination should always be performed for any child with a massive UGIB to consider liver disease, trauma, and earlier cardio-esophageal surgery.

In this study, we report a case of a previously well child with no history of liver disease or ingestion of foreign body who presented with sentinel bleeding for 2 weeks prior to an episode of massive hematemesis. AEF was obviously not considered initially because the amount of bleeding was not substantial. However, after massive bleeding was noted and resuscitation commenced, the possibility of vascular bleed was more obvious. EGD is the recommended mode for investigation in patients with UGIB [10].

In this patient, despite the active bleeding and hemodynamic instability, rapid diagnostic EGD was performed while the child was being ventilated in the PICU.

The EGD was uneventful and led to the insertion of a size F-18 SBT to tamponade the bleeding fistulous opening. This immediately stabilized the blood pressure and subsequently allowed the patient to be moved to the radiology department for CTA and finally for the definitive surgery.

CTA was needed to accurately localize the possible point of bleeding. CTA was the least invasive, most sensitive [11], and most convenient test for delineating the anatomy of the fistula.

Thoracotomy was suggested because the bleeding came from the middle third of the esophagus and CTA indicated an abnormal connection in the thoracic aorta. Laparotomy would have been performed if the bleeding was due to esophageal varices or gastric ulcers.

Thoracotomy and fistula ligation was the definite treatment for the fistula. An esophageal tear was noted at the site of the SBT due to the larger size of the F18 tube rather than a more suitable size of F14–F16, as used in other cases [12].

Esophageal tear is a well-recognized complication of SBT [13]. However, the tear was clean with no hematoma, fluid, or air leakage, as observed during thoracotomy. Hence, we consider that this complication caused no significant co-morbidity in this child.

The child deteriorated by the end of surgery, which occurred 12 h after the first large hematemesis and 8 h after stabilization with the SBT. It is likely that the cardiogenic shock and multiple arrests were due to severe ischemia during the first 4 h after initial presentation leading to acute kidney injury and severe metabolic acidosis until the bleeding was controlled by the SBT. The ischemia could have been further aggravated by the high doses of

vasoconstrictors required to raise the falling blood pressure.

It is possible that early EGD, insertion of a SBT, and earlier peritoneal dialysis might have limited the impact of severe ischemia on different organs, thereby leading to a better outcome.

4. Conclusion

This case highlights the importance of the early recognition of massive UGIB in children and the need for early transfer to the PICU. EGD should be performed as early as possible and a SBT with an appropriate size should be utilized in esophageal vascular bleeding even if non-variceal.

Conflict of interest

There is no conflict of interest to declare.

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