PST | PEDIATRIC SURGERY IN TROPICS

AN UNUSUAL CASE OF A MISSED ESOPHAGEAL FOREIGN BODY PRESENTING WITH A BRAIN ABSCESS

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Keywords	**Abstract**
Foreign body Tracheoesophageal fistula Esophago-vascular fistula Gastrointestinal bleeding	**Introduction:** Foreign body ingestion is a common occurrence in children. Majority of the ingested foreign bodies usually pass spontaneously without complications. One percent of these ingested foreign bodies result in life threatening complications such as esophageal perforation, tracheoesophageal fistula, esophago-vascular fistula (EVF), extraluminal migration, penetration into adjacent organs and death. EVF typically presents with massive upper gastrointestinal bleeding. **Case presentation:** We present a case of a 2-year 6-month-old boy with a missed sharp foreign body, an open safety pin, in the esophagus with an unusual presentation. The pin perforated the esophageal wall into the right common carotid artery and caused a pseudoaneurysm. This patient presented with a right parietal brain abscess and neurological fallout. The brain abscess was drained, the safety pin removed and the pseudoaneurysm resected with primary repair of the common carotid artery. **Conclusion:** EVF is rare and has a high mortality. There are no standardized protocols to manage these cases. We advocate for a multidisciplinary team approach and individualizing the management of each case.
Abbreviations	FB: Foreign body TEF: Tracheoesophageal fistula EVF: Esophago-vascular fistula

	AEF: Arterio-esophageal fistula UGIB: Upper gastrointestinal bleeding CT: Computed tomography CTA: Computed tomography angiogram MDT: Multidisciplinary team MRA: Magnetic Resonance angiography

INTRODUCTION

Foreign body ingestion is a common occurrence in children mostly between the ages 6 months and 3 years. [1,2] Ingested foreign bodies typically include coins, button batteries, toys and sharp objects such as pins, nails, toothpicks and bones. [3,4] Up to 90% of ingested foreign bodies usually pass spontaneously without complications. [2] Those that get impacted in the esophagus often only require endoscopic removal under short anesthesia. Impaction typically occurs in one of the narrow areas in the esophagus, namely the cricopharyngeus; the aortic notch; the left mainstem bronchus, and the lower esophageal sphincter. [8] One percent of ingested foreign bodies result in life threatening complications requiring surgical intervention. [2] Some of the reported complications include esophageal perforation, tracheoesophageal fistula, esophago-vascular fistula (EVF), extraluminal migration, penetration into adjacent organs and death. [1,2,4]

EVF, an abnormal communication between a vessel and the esophagus, has a high mortality due to exsanguination. [5] Sharp objects such as pins and bones are some of the frequently implicated foreign bodies to cause EVF. Aorto-esophageal fistula is the more commonly described type of EVF. [3]

Arterio-esophageal fistula (AEF), a communication between an artery and the esophagus, occurs less commonly but holds a high mortality rate of about 40-60%. [3] Patients with AEF typically present with upper gastrointestinal bleeding (UGIB) with or without a previous history of foreign body ingestion. Often, the patient initially has a herald bleed which is then followed by an exsanguinating UGIB.

Due to its rarity and high fatality, the management of AEF remains individualized or institutionally based with no universal standardized protocols or guidelines. [5]

We present a case of a toddler with an unusual presentation of AEF and his subsequent management.

CASE REPORT

A 2-year 6-month-old boy presented to a peripheral hospital with fever, lethargy, weakness of the left upper and lower limbs, and decreased level of consciousness for one day. This was preceded by a two-week history of refusal of feeds. A computed tomography (CT) scan of the brain revealed a brain abscess in the right parietal lobe region, measuring 28mm x

42mm x 29mm with midline shift to the left; mild subfalcine and uncal herniation; and extensive perilesional oedema. (Fig 1) A foreign body was also noted in the esophagus: an open safety pin with its sharp end at the level of T1/T2, pointing to the right. (Fig 2 and 3) The sharp end of the pin was piercing through the esophageal wall into the right common carotid artery. Its tip was lodged into the proximal part of the artery and had caused a pseudoaneurysm a few millimeters above the bifurcation of the right brachiocephalic and common carotid arteries. (Fig 4)

Fig 1. Right parietal brain abscess (asterisk) and perilesional edema (arrow)

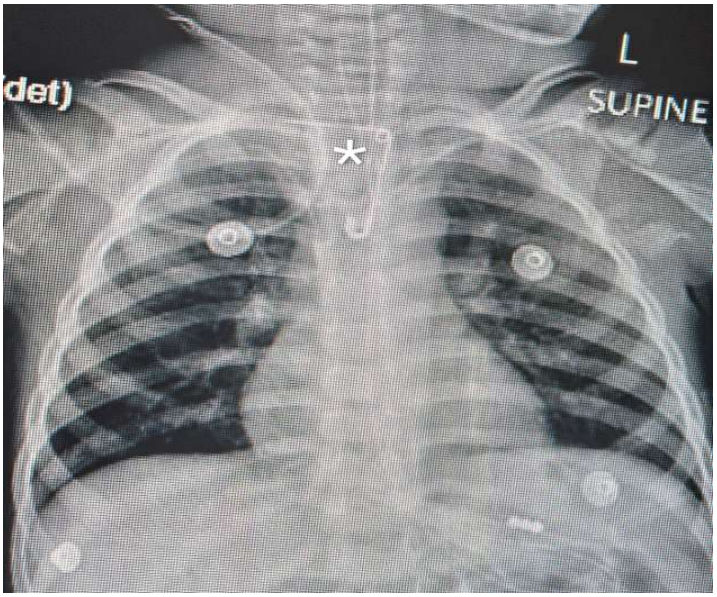

Fig 2. Open safety pin in esophagus (asterisk)

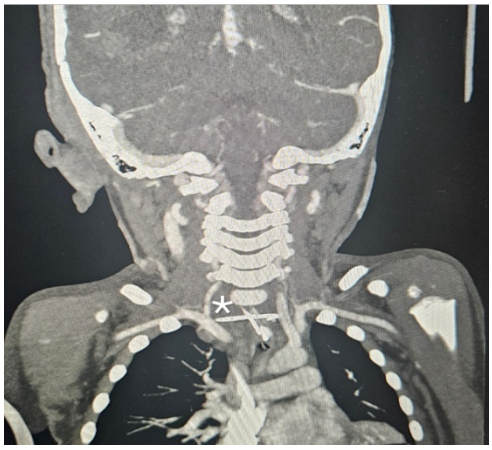

Fig 3. Safety pin with the sharp end to the right (asterisk)

Fig 4. Right common carotid pseudoaneurysm (arrows). Tip of the safety pin in the carotid artery

The patient was transferred to our hospital for further management by various specialties. A multidisciplinary team consisting of pediatric surgeons, neurosurgeons, cardiothoracic surgeons, vascular surgeons and pediatric intensivists were brought together. The brain abscess was drained twice, first at the peripheral hospital and again upon admission at our

hospital. A decision was made to approach the pseudoaneurysm via neck dissection and proceed according to the findings. Provision was made for sternotomy in case the need arose. The patient was then taken to theatre for removal of the foreign body. Right neck exploration was performed. Despite a lot of fibrosis, the right common carotid artery was identified and adequately exposed. (Fig 5) The pseudoaneurysm was identified in the proximal part of the artery. (Fig 6) Proximal and distal common carotid artery control was also obtained. The foreign body was not visible or palpable from the wound. A rigid esophagoscope was passed and the safety pin was seen embedded in the wall of the esophagus. The safety pin was grasped with metallic crocodile teeth grasper and pulled under vision. During the pulling, the safety pin started bending on itself and got dislodged from the esophageal wall which resulted in a less traumatic removal of the safety pin. (Fig 7) No esophageal perforation was noted. A nasogastric tube was passed for feeding. The pseudoaneurysm was excised and the common carotid artery primarily repaired without tension.

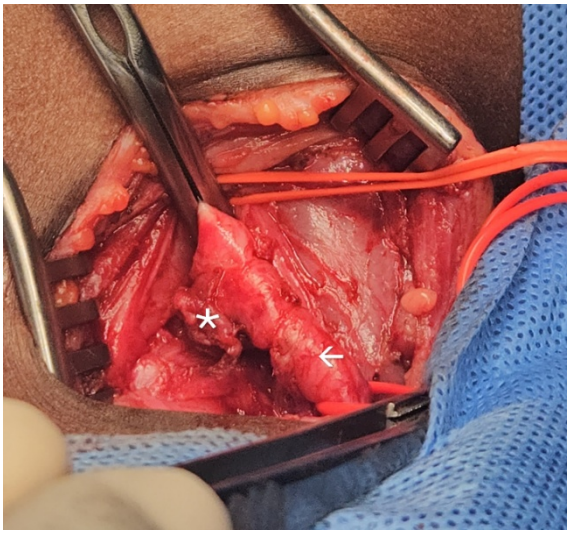

Fig 5. Right common carotid artery (arrow) exposed. The pseudoaneurysm in the proximal part of the artery (asterisk)

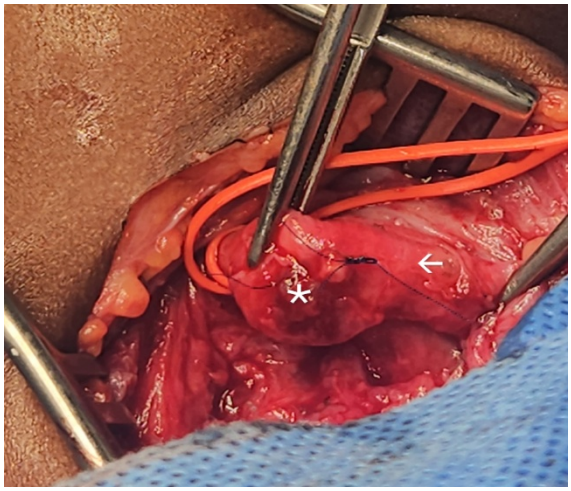

Fig 6. The right common carotid artery pseudoaneurysm (asterisk) exposed. Right common carotid artery (arrow)

Fig 7. The safety pin

A contrast swallow was performed a few days later and the esophagus was intact. Doppler ultrasound was also performed three weeks after the common carotid artery repair and showed normal flow in the common carotid artery.

The patient recovered remarkably after the last operation (removal of the foreign body). He could feed orally and started gaining strength on the left side of his body. He was referred to a different center for further rehabilitation.

DISCUSSION

Ingested sharp foreign bodies is associated with high morbidity and mortality. In general, ingested foreign bodies that get lodged in the intestine cause perforation in <1% of cases whereas with sharp foreign bodies, perforation can be as high as 15-35%. [(2)] Delay in diagnosis and management of ingested foreign bodies increases the risk of complication. [(2,4)]

Early diagnosis and prompt endoscopic intervention reduce the incidence of these complications. [(4)] Unfortunately, a number of these foreign body ingestions are not witnessed and therefore only present later and often with complications. EVF is one of the rare but well described complications that are seen with impacted esophageal foreign bodies. Patients with EVF typically present with massive UGIB. This is usually preceded by a small self-limiting bleed known as a herald bleed. [(5)] Our patient in this case report presented in an unusual way with a brain abscess and neurological fallout. The sharp end of the safety

pin was embedded in his esophagus and traversed the esophagus through to the right common carotid artery and formed a fistula between the two structures. He subsequently developed a pseudoaneurysm in the proximal common carotid artery. This led to septic emboli to his right brain resulting in abscess formation and neurological fallout. A computed tomography angiogram (CTa) showed the tip of the pin in the proximal common carotid artery with the pseudoaneurysm a few millimeters from the brachiocephalic artery.

Management of EVF is variable and depends on the site of the fistula, the presentation of the patient and resource availability per institution. There are no standard treatment guidelines owing to the rarity of these cases. Many authors emphasize the need for a multidisciplinary team (MDT) approach involving pediatric surgeons, pediatric cardiothoracic surgeons, interventional radiologists, vascular surgeons, pediatric anesthesiologists and pediatric intensive care. [1,3,5,6] Reddy et al. proposed a guideline which entails early recognition and diagnosis of EVF with massive bleeding, resuscitation with permissive hypotension, application of direct pressure or placement of a Sengstaken-Blakemore tube, ventilatory support; if stable, CTAthen stent the bleeding vessel, or CTA and operative repair of the vessel. [5] In addition, they advocate for mandatory involvement of the MDT.

Surgical intervention options are variable and include endovascular procedures such as stenting, angiographic embolization, use of vascular plugs, occlusive aortic balloon and open repair of the vessel via thoracotomy or sternotomy with or without cardiopulmonary bypass. [1,3,5-7]

No one procedure is considered more effective than the other as these cases are rarely managed. The use of endovascular stents in children remains controversial due to lack of long-term data on the outcome of stenting vessels that are still growing. As such, most surgeons prefer the open approach over endovascular intervention. [3] Open repair is not without its challenges. It can be very difficult to localize the fistula during open surgery. Unfortunately, patients may be too unstable to allow imaging before the surgical intervention. In the event CTA or Magnetic Resonance angiography (MRA) is not feasible due to patient's instability, ongoing resuscitation angiography in the interventional radiological suite with ongoing resuscitation is another possible option. [3] Some surgeons utilize endovascular stents as a temporizing measure whenever possible. Different institutions have variable resources available for optimal management of this condition under extreme time pressures without much room for error. As such, management of each case should be individualized and the best approach determined by the MDT. [5]

CONCLUSION

EVF is a rare but well described complication. AEF is even rarer. These patients almost always present with massive UGIB usually preceded by herald bleeding. Brain abscess with

neurological deficit as a presenting sign of a missed esophageal foreign body is highly unusual. Management of EVF remains a challenge with no validated guidelines. Cases are individualized and their management is greatly influenced by the resources available at the institution involved. MDT is of great importance in determining the optimal management approach for each case.

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