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A 2-Year-Old Boy Who Developed an Aortoesophageal Fistula After Swallowing a Button Battery, Managed Using a Novel Procedure with Vascular Plug Device as a Bridge to Definitive Surgical Repair

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Male, 2-year-old
Final Diagnosis: Aortoesophageal fistula
Symptoms: Esophageal foreign body • gastrointestinal bleeding
Medication: —
Clinical Procedure: —
Specialty: Gastroenterology and Hepatology • Pediatrics and Neonatology

Objective: Rare disease

Background: Button batteries (BBs) can be inhaled or swallowed accidentally, particularly by infants and children, who can present as a surgical emergency with a fatal outcome. This report is of a case of a 2-year-old boy who developed an aortoesophageal fistula (AEF) after swallowing a button battery and was successfully treated using a novel vascular plug device as a bridge to definitive surgical repair.

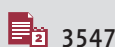
AEF is diagnosed using computed tomography angiography (CTA), after laparotomy, and using aortography. Aortic endovascular stenting and vascular occluder placement is a minimally invasive emergency option until definitive treatment can be provided. The surgical options include repair the aortic defect primarily, or the diseased segment of the aorta is replaced with a graft.

Case Report: A 2-year-old boy presented with hematemesis 9 days after ingestion of a button battery, which was removed endoscopically 16 hours after the incident. The patient was resuscitated aggressively and diagnosed to have AEF using computed tomography angiography (CTA). The aorta was successfully repaired using a vascular plug device, which kept the patient safe until the definitive surgical treatment was done 2 months later. The defect was repaired with direct anastomosis and completed with a patch of bovine pericardium, as well as closure of the fistula from the esophageal side with stitches.

Conclusions: This report presents a rare but previously reported complication of swallowing a button battery, resulting in an aortoesophageal fistula. The aorta was successfully repaired using a vascular plug as a minimally invasive emergency option, which is considered as a lifesaving procedure and a bridge for definitive repair.

Keywords: Fatal Outcome • Foreign Bodies • Pediatrics • Survival • Vascular Fistula

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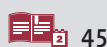
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Background

Foreign body (FB) ingestion is common among young children and is mostly an uneventful incident. Complications resulting from FB ingestion depend on the nature of the FB, size of the FB, site of dislodgement, duration of retention of FB, presence of any anatomical abnormalities, and the age of the child. Button batteries (BBs) are among the most dangerous objects that are commonly ingested, especially in the esophagus, and their removal requires emergency endoscopy [1]. Many serious complications have been reported with BB ingestion. These include perforation, mediastinitis, gastrointestinal (GI) bleeding, esophageal stricture, tracheoesophageal fistula, and aortoesophageal fistula (AEF) [2]. AEF was fatal in most reported cases. If AEF is suspected, CTA is the preferred initial diagnostic test. The management of AEF in children includes aggressive resuscitation, measures to control bleeding (eg, use of balloons), procedures like percutaneous endovascular aortic repair (EVAR), or our novel use of a vascular plug device to close the fistula as a bridge to definitive surgical repair.

Case Report

An otherwise healthy boy, age 2 year 6 months, presented to the Emergency Department on 17 May 2020 at 9 PM with a history of frequent vomiting and difficulty in swallowing semi-solid food. Accidental battery ingestion was suspected when his family noticed the absence of a battery in the glucometer device at 5 AM. The estimated time since the ingestion was

16 h. On physical examination, the patient generally looked well; he was not tachypneic or distressed. He was mildly dehydrated and showed stable vital signs, and the results of the systemic examination were unremarkable. Chest radiography revealed a BB located in the upper part of the esophagus. The patient was admitted to the hospital and was kept nil per os (NPO). An urgent upper gastrointestinal endoscopy (EGD) was performed under general anesthesia within 2 h of his arrival to the hospital, and the endoscope inserted to 15 cm. A disc battery was found, with a circumferential area of necrosis. The battery was fixed to the mucosa and it was removed using rat-tooth forceps. The area was found to have circumferential third-degree burns at 15 cm (Figure 1). The patient stayed in the hospital for 5 days. On the first 2 days, he was kept NPO and was administered intravenous omeprazole. Chest radiography was repeated, with normal results, and no contrast study (esophagogram) done. During the next 2 days, intake of only water was allowed, following which intake of other liquids was permitted. The patient tolerated feeding and was discharged in good condition on the fifth day. On 26 May 2020, almost 4 days after discharge, the patient presented to our Emergency Department at 9 PM with a history of melena for 1 day, and nasal bleeding and hematemesis on the day of admission. The mother gave a history of cough, runny nose, and fever since the last 2 days. On examination, the patient looked generally unwell, febrile, extremely pale, tachypneic, and dehydrated. His vital signs were as follows: temperature 38.3°C, heart rate (HR) 175 beats per min, respiratory rate (RR) 30 breaths per min, BP 72/44 mmHg, SpO₂ 100% on face mask 10 L/min, blood gas PH: 7.08, PCO₂ 42, HCO₃ 11.2, lactate 7.9, and random blood

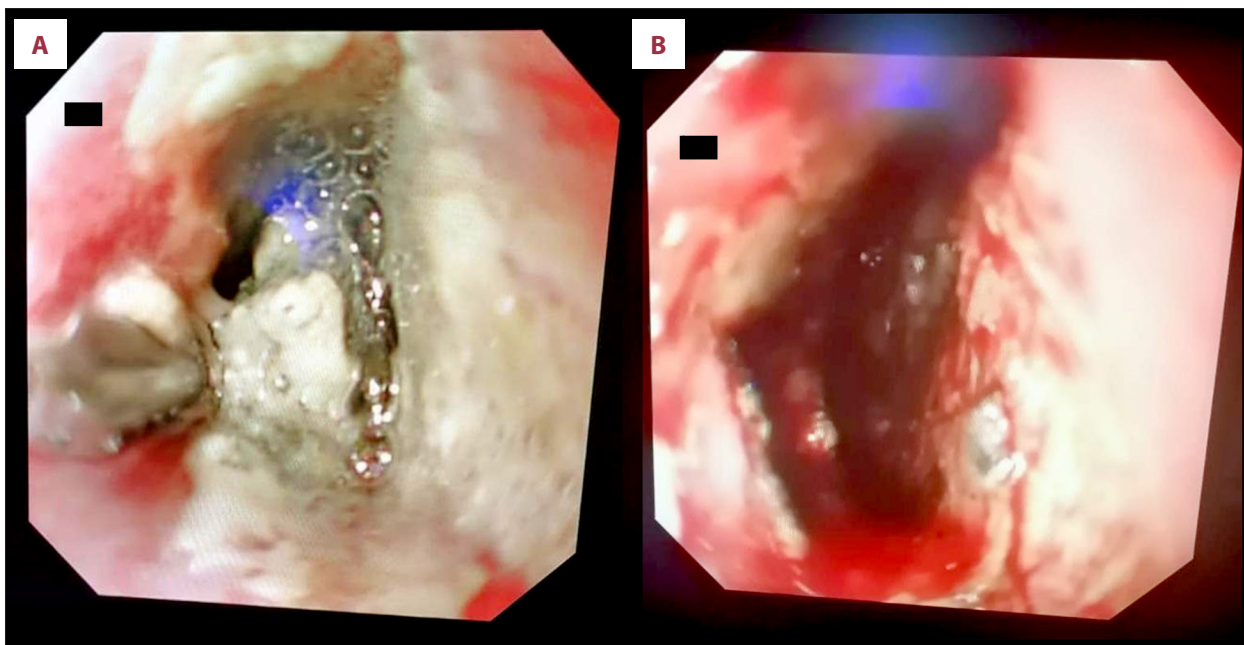


Figure 1. Endoscopic view of esophageal injury before (A) (shows circumferential exudate and battery in place) and after (B) removal of button battery (shows circumferential deep ulceration, necrosis, and hemorrhage).

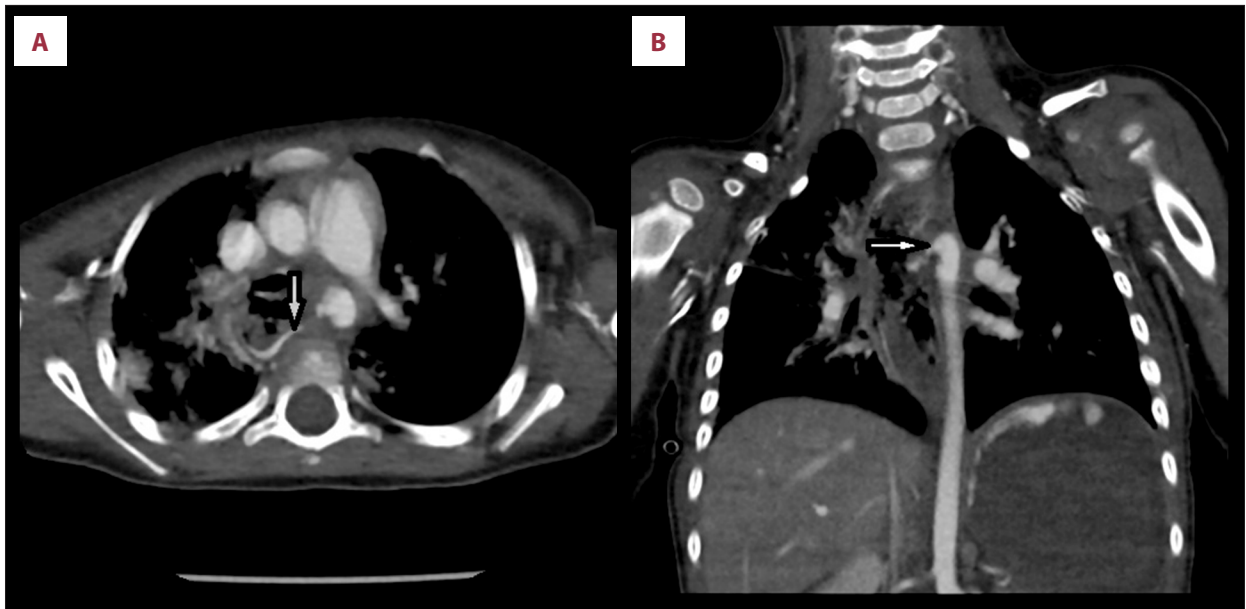


Figure 2. (A) Axial CT scan of the chest showing focal irregularity and bulging of the medial side of the aorta with the fistula communication between the aorta and dilated esophagus (shown by the arrow). Also there is fistula between the esophagus and the right main bronchus. (B) Coronal view with contrast showing focal aorto-esophageal fistula with contrast extravasation from the aorta into dilated esophagus. Also, there is contrast outlining the stomach wall reaching from the aorto-esophageal fistula, indicating significant bleeding at time of presentation.

sugar 278 mg/dl (possibly due to stress, as it later normalized). Cardiovascular examination revealed normal first and second heart sounds and hemic murmur; no gallop was heard. Examination of the respiratory system revealed equal bilateral air entry, with no added sounds. Gastrointestinal examination showed a mildly distended but soft and lax abdomen with no organomegaly. In the emergency room, normal saline boluses of a total 40 ml/kg were administered, as well as packed red blood cells (PRBCs) 10 ml/kg, tranexamic acid stat, omeprazole, and paracetamol, as he was febrile. An urgent computed tomography (CT) with contrast was performed after he was sedated with 2 doses of midazolam and lorazepam. CT angiography (CTA) of the chest and abdomen showed a fistula communication between the aorta and esophagus at the level of the T4 vertebra distal to the left subclavian artery. The fistula measured approximately 3.3 mm in transverse diameter, with contrast media filling the dilated esophagus and extending to the stomach, suggestive of active bleeding (Figure 2). A suspicious fistula was noted between the trachea and esophagus at the level of the right main bronchus. The patient was shifted to the pediatric intensive care unit (PICU) after resuscitation with 2 boluses of normal saline at 20 ml/kg. His BP was 86/45 mmHg, HR 140 bpm, and he was tachypneic. The patient received PRBCs a second time immediately in the PICU. Elective rapid sequence intubation was performed. The interventional cardiologist was ready to intervene and close the fistula from the aortic side using the device. Therefore, an urgent referral was made to the nearby cardiac center (10 to 15

minutes away from our hospital). In the cardiac center, the patient received 1 unit of PRBCs attached, a central line was inserted in the internal jugular vein under ultrasonography (US), and complete aseptic technique was used. Another unit of PRBCs and 1 unit of platelets (PLT) and fresh frozen plasma (FFP) were administered owing to massive bleeding during the procedure. The patient was administered sodium bicarbonate twice for acidosis. The procedure was performed under general anesthesia and intubation. The right femoral artery was punctured, and a 5F sheath was introduced using the Seldinger technique. A 4F JR diagnostic catheter (JR 3.5, 100 cm×0.038", Cordis Company, Florida, USA) was introduced into the descending aorta, and an aortogram was obtained using hand injection contrast. A selective fistulogram was performed to delineate the course of the fistula. A guidewire (radiofocus guidewire, 0.035×150, Terumo Corporation, Japan) was used to cross the fistula and was placed inside the esophagus. Thereafter, a 5F-long delivery system was introduced in exchange with a short sheath. The Amplatzer Duct Occluder II 6×6 mm (ADO II; St. Jude Medical, St. Paul, MN, USA) was used to occlude the fistula (Figure 3). The device position was checked using an aortogram from the side arm of the long delivery sheath. The device was released in a good position, and an aortogram was obtained at the end, which showed no more leakage. At the end of the procedure, all catheters and wires were removed, and manual compression was used to stop bleeding from the punctured side. The patient remained well sedated in the PICU on fentanyl 4 µg/kg/h and midazolam

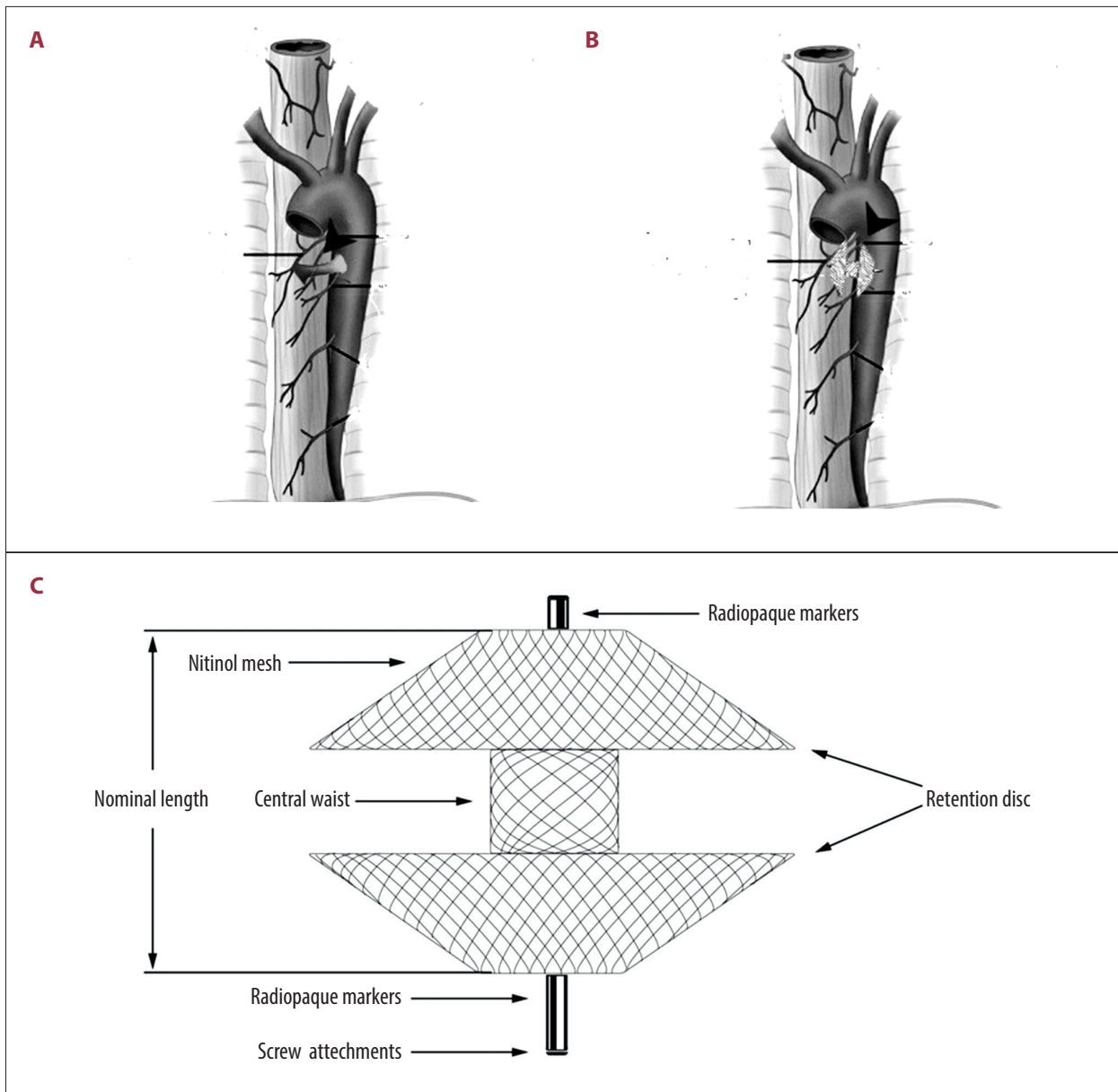


Figure 3. Procedure illustrations of vascular plug device. AEF, Aortoesophageal fistula (A, arrowhead), position of the device after placement (B, arrowhead), dimension and shape of the Amplatzer Duct Occluder II (C).

4 $\mu\text{g}/\text{kg}/\text{min}$, ventilated on SIMV pressure control. The NPO was maintained, and NGT was inserted by a pediatric surgeon to decompress the distended stomach, and the patient was started on total parenteral nutrition. As the patient still had fever, ceftriaxone was discontinued, and meropenem plus vancomycin was started. A chest CTA, obtained on the fifth day, showed interval placement of the vascular plug through the previously seen AEF. Currently, it is seen extending between the aorta and esophageal lumen with no active extravasation (Figure 4). The esophagus remained dilated with a persistent fistula noted between the trachea and the esophagus at the level of the right main bronchus (esophageal-bronchial

fistula. A plain CT scan of the brain showed a well-defined hypodensity involving the right occipital lobe with surround mass effect and effacement of the cortical sulci, denoting brain infarction involving the right PCA territory. Echocardiography was normal. The patient was extubated after 1 week and was stable with only melena in the first few days, although without a significant drop in hemoglobin levels, as the patient pulled out the NGT and he was therefore continued on total parenteral nutrition (TPN) the next week. An upper GI contrast study revealed fistula communication between the esophagus and the right main bronchus. The esophageal stricture could not be fully assessed as a small amount of contrast was given to avoid

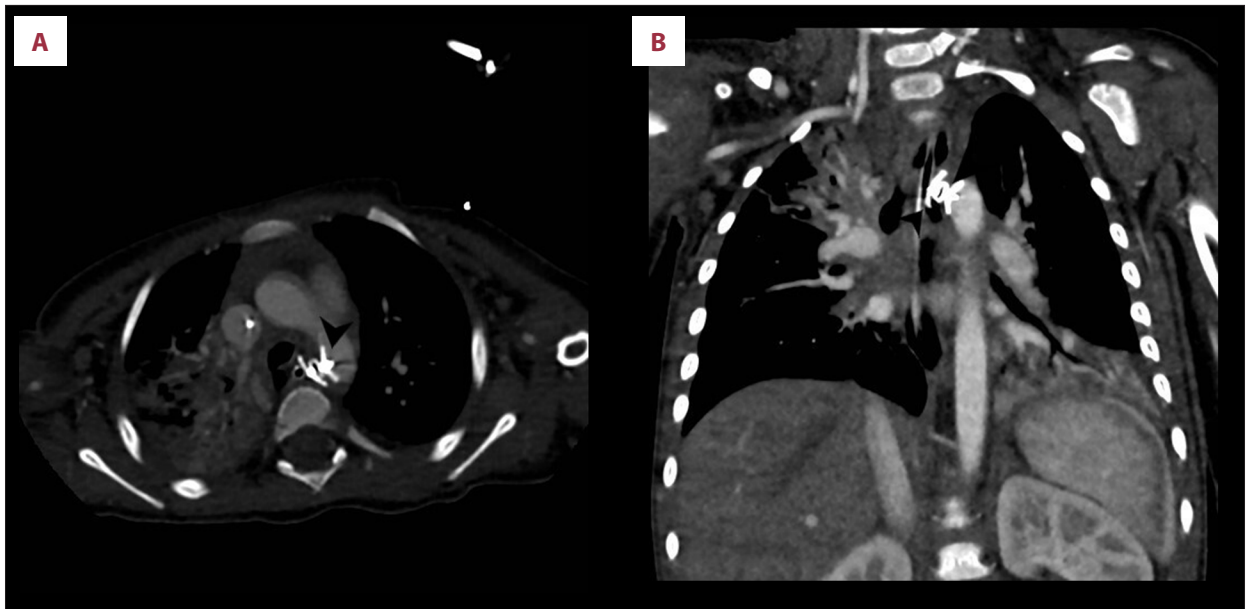


Figure 4. Axial (A) and coronal (B) CT scan of the chest post insertion of the vascular plug device (black arrowhead) through the aorto-esophageal fistula securing both sides of the fistula.

aspiration, so laparoscopic gastrostomy tube insertion was performed. Enteral feeding was advanced, and TPN was weaned. The patient was discharged in the fourth week. CTA was repeated in the sixth week after device insertion, and shows closure of the tracheoesophageal fistula and the device in place. The patient was referred to a higher-level care center for an opinion regarding the long-term plan for this patient. As the long-term plan for this device is unknown for them, cardiothoracic surgeons prefer to remove this device electively, and they repair the defect posteriorly with direct anastomosis between the 2 ends and anteriorly completed with a patch of bovine pericardium and closure of the fistula from the esophageal side with stitches according to the medical report from the center. The patient was doing fine during follow-up, and 1 year later was able to tolerate soft and solid food without dysphagia, as the gastrostomy tube came out by itself a few months after the surgery. Also, he resumed normal physical activities without a neurological sequel. We are unable to do any further endoscopy or contrast studies as the patient has completed follow-up.

Discussion

Ingestion of FBs is common in children younger than 6 years. The most common objects ingested are coins and bones of chicken and fish. Ingestion of BBs accounts for less than 2% of FBs ingested by children [3]. The most harmful and serious complications occur with a battery size of 20 mm [2,4,5]. The injury is attributed to various causes, including electric currents leading to the formation of corrosive hydroxyl ions, and

leakage of the alkaline contents of the batteries, causing corrosion and pressure necrosis [4]. While most ingested BBs pass spontaneously without any complications, especially beyond the stomach [3], complications usually occur from BBs that are stuck in the esophagus. The most frequent site of esophageal dislodgment is in the upper esophagus [3,6].

Complications from BB include pneumonia, mediastinitis, hemorrhage, esophageal ulceration, sepsis, tracheoesophageal fistula, perforation of esophagus, tension hydropneumothorax, pneumoperitoneum, and esophageal-vascular fistulas, such as erosion into the subclavian artery or thyroid vessels, AEF, and esophageal stenosis [5,7]. AEF is rare and poses a life-threatening emergency, and it is fatal in most affected patients. Causes of AEF in children include FB ingestion, which mainly include BBs and sharp objects such as pins, and fish and chicken bones. Other FBs, such as vascular rings, coins, and NGT, have been reported, especially in the presence of anatomical abnormalities such as a vascular ring.

Most BB ingestion cases were unwitnessed [2], and the presentation in these patients varied, including non-specific vomiting and vomiting after ingestion of solid and liquid food or only after ingestion of solid food, while liquid intake was tolerated. Additionally, there have also been reports of refusal to eat, dysphagia, odynophagia, sialorrhea, abdominal pain, abdominal distention, hematemesis, melena, fever, irritability, fussy behavior, crying, lethargy, anorexia, respiratory symptoms such as tachypnoea and stridor, dehydration, acrocyanosis, and seizures. A lack of a history of ingestion led to recurrent ER visits and misdiagnosis of upper respiratory tract

Table 1. Literature review of surviving cases of aorto-esophageal fistula (AEF).

Ref. (Year)	Age \ Sex	Cause of AEF	Presenting symptoms	Diagnosis	Managements
McComas BC (1991) [8]	8m F	Straight pin	Melena, massive GI hemorrhage	During operation	Aorta(A) defect repaired primarily
Yahagi N (1992) [9]	9d F	DAA+NGT	Pulsatile hemorrhage from the mouth & nose	During operation	Aorta(A) defect repaired primarily
Othersen HB (1996) [10]	2m M	DAA+NGT	Life-threatening upper GI hemorrhage	– CT fail to demonstrate AEF – During operation	– SB Tube – Aorta(A) defect repaired primarily
Othersen HB (1996) [11]	6w M	DAA+NGT	Hematemesis, melena	During operation	– SB Tube – Aorta(A) defect repaired primarily
Eckehard A (2001) [11]	7y M	Coin in the stomach. ? vascular ring	Hematemesis, mid-epigastric pain	During operation	– 24-F Foley balloon catheter – The diseased segment of aorta was replaced with graft
Jiri Snajdauf (2005) [12]	12.5y F	Esophagitis DUE TO a leaf of house plant Dieffenbachia	Hematemesis, melena	During operation	– SB Tube – Aorta(A) defect repaired primarily
D'Angelisa ARDV (2006) [13]	4m ?	DAA+NGT	Hypovolemic shock, hematemesis, melena	– Endoscopy: Esophageal ulcer – Aortography: non-conclusive – During operation	– SB Tube – Aorta(A) defect repaired primarily
Burns BJ (2008) [14]	11y M	Mycotic pseudoaneurysm in undiagnosed coarctation	Vomiting, mild chest & upper abdominal, hypertension, hematemesis	CTA: fail during operation	Diseased segment of aorta was replaced with graft
Hill SJ (2010) [15]	9y F	Retained internal bolster from the PEG tube	Chest pain and odynophagia massive hematemesis with cardiovascular collapse	Aortogram confirmed the AEF	Bedside placement of an(A) Occlusion Balloon, covered stent was deployed in A. resection of esophagus with primary anastomosis
Coates LJ (2011) [16]	2y M	Unknown cause but patient potentially had an infective cause for the AOF	Hematemesis, melena	During operation	– SB Tube – Aorta(A) defect repaired primarily
Spiers A (2012) [17]	10m M	Button battery in the distal esophagus	Dyspnea and hematemesis	CT angiography	Aorta(A) defect repaired primarily
Takazawa S (2013) [18]	49d F	DAA+NGT	Hypovolemic shock, hematemesis	Endoscopy: esophageal ulcer at the level of the aortic arch during operation	Aorta(A) defect repaired primarily
Nicholas S (2016) [19]	6y M	Coin & A right-sided aortic arch	Hematemesis, hemorrhagic shock	– Intraoperative angiogram fail to show AEF – During operation	Autologous pericardial patch aortoplasty was performed

Table 1 continued. Literature review of surviving cases of aorto-esophageal fistula (AEF).

Ref. (Year)	Age\ Sex	Cause of AEF	Presenting symptoms	Diagnosis	Managements
Granata A (2018) [20]	3y F	20-mm battery	Hematemesis, hemorrhagic shock	Angiogram show AEF	– SB Tube – Endovascular stent was released
Bartkevics M (2019) [22]	12m F	20-mm battery	Hematemesis, melena	CT angiography	Aorta(A) defect repaired primarily
Sinclair EM (2021) [22]	6y F	21-mm battery	Hematemesis	Aortogram confirmed the AEF	Endovascular stent
Alreheili K (2020) This Report	2.5y M	20-mm battery	Hematemesis, melena	CT angiography	Vascular plug device sealing the fistula

d – days; m – month; y – years; M – Male; F – Female; DAA – double aortic arch; NGT – nasogastric tube; GI – gastrointestinal; SB tube – Sengstaken-Blakemore tube; CTA – computed tomography (CT) angiography.

infection, urinary tract infection, acute gastroenteritis, ear infection, and sepsis [5]. The age of presentation with complications secondary to BB ingestion was reported to be 1-4 years. The duration of development of these complications after ingestion ranged from 10 h to 28 days, with most patients presenting on day 9 or 10 [5].

We report the case of a child who developed AEF 9 days after BB removal and who was resuscitated aggressively. The fistula was diagnosed with CTA. The patient was managed with novel techniques using a vascular duct occluding device that perfectly closed both sides of the fistula and stopped bleeding. This technique works efficiently to save patient until the time of definitive correction. This technique has never previously been reported to treat AEF. In this article, we review the survival and fatal cases regarding demographic characteristics, presentation, mode of diagnosis, and how the surviving cases were managed, including our patient, to show that aggressive resuscitation and minimally invasive procedure are crucial in saving the patient's life until definitive correction.

Seventeen patients have been reported to have survived AEF, including our reported patient [8-22]. **Table 1** shows a literature review of cases that survived. Five of the cases were secondary to BB, one was due to an unknown cause, potentially an infective cause, one was caused secondary to ingestion of the house plant *Dieffenbachia* during suicide attempts, and 6 were secondary to a vascular ring with a FB coin, NGT, or ETT. In 3 patients, the cause was a FB (straight pin, a bolster of PEG tube, or a coin), and that in one patient was secondary to mycotic pseudoaneurysm in undiagnosed coarctation of the aorta. In the 5 patients with BB ingestion, the BB was removed 6 h after ingestion in one patient and 14 h in another one while being removed from the stomach, while in the other patient after 36 h of possible ingestion, after 18 h in our patient, and after an unknown duration in the fifth patient,

which was removed intraoperatively from the stomach. The BB was discovered in other cases either after laparotomy or after a patient vomited it out, or its ingestion remained unknown. The age of the patients who survived ranged from 9 days to 12.5 years. Presentation was similar to hematemesis and melena or fainting attacks secondary to hypovolemic shock; sentinel exsanguination also ranged from a few hours to a few days. Diagnosis of AEF was performed by computed tomography angiography (CTA) in 3 out of 5 patients and after laparotomy to identify the source of bleeding in 11 patients. Aortography performed in 5 patients failed to detect the fistula in 2 patients. Most patients received aggressive resuscitation with fluid and blood products before successful operation. Regarding management, an esophageal balloon catheter tamponade was performed to control bleeding in 8 out of 17 patients, of whom the Sengstaken-Blakemore tube (SBT) was used in 6, while a Foley balloon catheter was used in one patient, and bedside placement of an aortic occlusion balloon was performed in another patient. Aortic endovascular stent placement was performed in 3 patients. A novel vascular device was done in our patient. In all patients who treated surgically, thoracotomy was performed, the fistula was identified and excised, and esophageal and aortic defects were repaired primarily, except in 2 patients in whom the diseased segment of the aorta was replaced with graft.

A review of published cases was performed using PubMed and the National Capital Poison Center. Fatal outcomes were reported in 50 cases [5,6,24-42], of which 58% (29 cases) were BB-induced injuries, and 42% (21 cases) were not related to BBs. Among the BB-related cases, 27 were diagnosed with AEF and 2 cases had other vascular erosions. BB size was 20 mm or larger in all cases, except one in which it was 16 mm in size. Although our review was up to age 14 years, the age of fatal cases for button battery related cases interestingly ranged from 1 to 4 years. In 22 cases, the AEF was confirmed, and in

Table 2. High index of suspicion criteria for diagnosis AEF.

Major criteria	Minor criteria	Negative criteria
	A (Clinical)	
A. sentinel bleeding event	1 – Child age less than 4 years	Absence of portal HTN
B. Massive bright red blood with hypovolemic shock	2 – History dysphagia, food refusal, or recurrent food impaction	Absence of known esophageal Varices
	3 – Suspicion of FB ingestion	
	4 – Recurrent ER visit in last 4 weeks.	
	5 – Known vascular ring	
	6 – Hx of button battery removal endoscopically or passed spontaneously in last 4 weeks	
	7 – Prolong NGT tube especially in presence of anatomical abnormalities, eg, vascular ring or repaired TEF	
	B (Radiological/Endoscopic)	
	1 – FB in chest or abdominal Radiograph	
	2 – Stomach massive. distension in radiograph	
	3 – Rib notching in chest radiograph	
	4 – Widening of the mediastinum, displacement of the trachea to the right, displacement of a nasogastric tube to the right	
	5 – Endoscopic finding of active bleeding, ulcer, or any other odd lesion in mid-esophagus	

FB – foreign body; ER – Emergency Room. {2 Major+1 minor, Major B+1 minor A or 1 minor B, Major A+2 minor A or 1 minor A and 1 minor B} PLUS {Negative Criteria}.

5 cases, the diagnosis was made clinically and no diagnostic procedures mentioned in the literature. The BB was removed in 12 cases (11 endoscopic and 1 surgical removal), while removal was unknown in 2 cases. Fatal GI bleeding occurred 10 h to 28 days after removal. Diagnosis of AEF was confirmed postmortem in 15 cases, during the operation in 3 cases, by CTA in one case, and by unknown methods in 3 cases. Non-BB esophago-vascular injuries were reported in 21 cases, all of which were AEF. Ten cases were secondary to vascular rings, 9 of which were double aortic arch (DAA) with prolonged NGT in 9 cases. The duration was reported to be as short as 8 days in pediatric patients, and Montgomery salivary bypass tube (MSBT) was used in one patient. In 6 patients, the AEF was due to FB, and one was due to VATER syndrome with operated TEF and prolonged NGT. One post-PDA repair and iatrogenic sutures in the aorta wall led to subsequent formation of AEF. Two were due to rupture of the mycotic aortic aneurysm and one due to an unknown cause.

Diagnostic procedures included chest and abdominal radiography, esophagography, EGD, CTA, and aortography. The

importance of chest and abdominal radiography is during the initial evaluation of the patient. Its importance is to identify FBs as a possible cause of AEF. The stomach may be hugely distended as blood from the fistula accumulates in the stomach and has been reported in many cases. Other findings may indicate rupture of aortic aneurysm or missed coarctation of the aorta, which may be complicated by an aneurysm. These findings include widening of the mediastinum, displacement of the trachea to the right, displacement of the nasogastric tube to the right, and rib notching on chest radiograph. Although barium esophagography has low sensitivity, it may detect AEF in adult patients [43]. We did not find any significant benefit in pediatric patients, which may delay the diagnosis. This is because of the different aetiologies in pediatric and adult patients; the most common cause in adults is thoracic aortic aneurysm, which is rare among children. Endoscopy findings varied in reported cases, and included pulsatile or fresh bleeding [11,12,40], pulsatile submucosal hematoma [44], a non-pulsatile purple mass protruding into the esophagus in the aneurysm [36], esophageal ulcer with [18] or without bleeding [13], thickened mucosa [21], and an odd purple streak [16].

All of these findings were in the middle or upper esophagus. Endoscopy can also be effective in that it can rule out other differential diagnoses. CTA showed extravasation of contrast material from the aorta into the esophagus.

The rate of CTA for AEF in pediatric patients was 60% in our review (Table 1). Rapid diagnosis is critical; therefore, the diagnostic approach for pediatric patients presenting with massive upper gastrointestinal bleeding included upper EGD, CTA, and aortography. The first modality to be performed depended on the most likely diagnosis and the patient's hemodynamic status. If AEF is suspected, CTA is the preferred initial diagnostic test as it is easier to obtain and less invasive than EGD or aortography. This requires a high index of suspicion. Chiari's triad was used in adult patients [43]. The triad comprises midthoracic pain, sentinel arterial hemorrhage, and exsanguination after a symptom-free interval. We believe it is not applicable to the pediatric age group as almost all patients are under the age of 4 years and they are non-verbalized to indicate pain and to give a history of FB ingestion, as it was most commonly unwitnessed.

We developed criteria that aided in a high index of suspicion for the diagnosis of AEF (Table 2). These criteria were developed depending on the revision of all cases showing survival and fatalities of AEF. It includes 2 major criteria, clinical and radiological/endoscopic minor criteria, and exclusion criteria. Therefore, in the absence of portal hypertension and known cases of esophageal varices, the presence of 2 major+1 minor, major B+1 minor A or 1 minor B, major A+2 minor A or 1 minor A, and 1 minor B will determine a high probability that the patient has AEF. These criteria were applicable for most of reviewed cases and because this is not the scope of this article, we are planning to validate these criteria in the future for all possible reported new cases and do modification as necessary in separate work.

During the management of AEF in children, aggressive resuscitation is crucial to allow diagnostic tests and definitive treatment to be carried out. In many reported cases of survival, patients were supported by large volumes of fluids and blood products. Trials to stop bleeding are also important measures. The SBT has proved advantageous in controlling bleeding. It is

valuable for gaining time to allow for definitive surgical treatment. Percutaneous endovascular aortic repair (EVAR) using balloon expandable stents is used in pediatric patients. Three patients have been reported to survive [15,20], while in one patient with mycotic aneurysms, the definitive surgical treatment was delayed, leading to death of the patient 3 months after stent placement [35]. In adult studies, EVAR should be considered only as a temporizing strategy until definitive repair can be performed [45]. In addition, the use of EVAR in children is debatable because of technical considerations, and it is recommended as a lifesaving procedure. In our case, we report the novel use of a vascular plug device to close both aortic and esophageal defects and which was stable to date, almost 2 month later. Whether to consider this procedure as a definitive treatment or as a bridge to definitive surgical repair needs to be evaluated. Surgical procedures must consider the procedure of choice at the time, and must include identification and resection of the fistula and repair of esophageal and aortic defects with or without the use of a graft.

Conclusions

AEF is rare and has a fatal outcome in most cases. Diagnosis requires a high index of suspicion, and treatment should include aggressive management. The endovascular device remains a non-invasive emergency treatment option and should be considered as lifesaving and as a bridge to definitive repair.

Department and Institution Where Work Was Done

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Conflict of interest

None.

Declaration of Figures Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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