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Esophago-Vascular Fistulae in Children: Five Survivors, Literature Review, and Proposal for Management

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ABSTRACT

Introduction: Esophago-vascular fistulae in children are almost uniformly fatal with death occurring by exsanguination. We present a single centre series of five surviving patients, a proposal for management and literature review.

Materials and methods: Patients were identified from surgical logbooks, surgeon recollection and discharge coding data. Demographics, symptoms, co-morbidities, radiology, management and follow up details were recorded.

Results: Five patients (1M, 4F) were identified. Four were aorto-esophageal and one caroto-esophageal. Median age at initial presentation was 44 (8–177) months. Four patients had cross sectional imaging prior to surgery. Median time from presentation to combined entero-vascular surgery was 15 (0–419) days. Four patients required repair on cardio-pulmonary bypass with four undergoing staged surgical procedures. All required combined esophageal and cardio-vascular surgery.

Length of PICU stay following combined surgery was 4 (2–60) days and overall hospital stay was 53 (15–84) days. Median follow up was 51 (17–61) months.

Two patients had esophageal atresia and trachea-esophageal fistula managed as neonates. Three had no co-morbidities. Four had esophageal foreign bodies:1 esophageal stent, 2 button batteries, 1 chicken bone. One patient had a complication following colonic interposition. Four patients required an esophagostomy at the time of definitive surgery. All patients were alive and well at last follow up with one having successful reconnection surgery.

Conclusion: In this series, outcomes were favourable. Multidisciplinary discussion and surgery are mandatory. If hemorrhage is controlled at presentation, then survival to discharge is possible but the magnitude of surgical intervention is both significant and very high risk.

Level of evidence: Level 3.

1. Introduction

Esophago-vascular fistulae (EVF) in children are almost uniformly fatal due to exsanguination [1–4], but there are several reports where children have been successfully managed [5–8].

EVFs result from ingested [9–13] or iatrogenic foreign bodies (FB) for example, button batteries, cleaning fluids or stents. They can also occur post operatively [14] following surgery for structural anomalies such as esophageal atresia (EA) with or without a trache-esophageal fistula (TEF) [15] or abnormalities of the great vessels, including those repaired during cardiovascular surgery [16–18]. Some have unrecognized anatomical abnormalities which can increase the likelihood of fistula formation, such as vascular rings around the esophagus and other aberrant vasculature.

We report a single centre series of five consecutive successfully managed children, a literature review and a management proposal. This represents one of the largest reported series of successfully managed EVF in infants and children.

Abbreviations: EVF, Esophago-vascular fistula; FB, Foreign Body; EA, Esophageal atresia; TEF, Tracheo-esophageal fistula; SBT, Sengstaken-Blakemore tube; PDS, polydiaxanone; CP, Cardio-pulmonary; DCA, Digital subtraction angiography; CPB, cardiopulmonary bypass; BB, Button battery; CTA, computed tomographic angiogram; CXR, chest x-ray; MDT, Multidisciplinary team; PTFE, Polytetrafluoroethylene; BBI, Button battery ingestion; LCCA, Left common carotid artery.

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2. Materials and Methods

All patients who presented to our institution with hematemesis and were diagnosed with EVF between 01/2005 and 01/2020 were included. Patient notes, electronic records, radiology, paediatric intensive care and theatre records were reviewed. Demographics, presenting symptoms, diagnosis, co-morbidities, management, and follow-up details were recorded. Data is median (range) unless specified. Patient timelines are shown figuratively (Fig. 1).

3. Case histories

3.1. Patient 1

A previously well 15-year-old girl presented with a massive hematemesis eight days after swallowing a chicken bone and four days after its removal at her local hospital. Following resuscitation, an esophagoscopy showed an esophageal ulcer with overlying clot. Later that day, she had a further massive hematemesis, which was controlled with a Sengstaken-Blakemore tube (SBT). An emergency posterolateral thoracotomy was performed, and an aortic defect was repaired. The oesophageal perforation was repaired using polydioxanone sutures (PDS) and an intercostal muscle flap was interposed.

A further bleed five days later required the excision of a necrotic aortic area and patch aortoplasty with bovine pericardium. A small oesophageal leak was noted on a contrast study (Fig. 2A) and a further aortic bleed (Fig. 2B) nineteen days after re-exploration was managed successfully with an endovascular covered CP stent (Numed Inc, NY, USA).

She was discharged after 15 days and represented a month later with melaena when esophagoscopy documented a persistent esophageal ulcer. Extravasation was seen at digital subtraction angiography (DSA) (Fig. 2C) and further endovascular stenting was performed successfully. A barium swallow showed no esophageal leak.

A further episode of melaena occurred a month following stent insertion. DSA showed no leak, and a gastrostomy was formed to rest the esophagus.

Further melaena and fresh blood in the gastrostomy aspirates occurred 22 days later. A SBT was inserted, and the left chest was re-explored. The posterior esophageal perforation was sealed in dense adhesions to the aorta along a 6 cm length with a 1 cm aorto-esophageal communication.

The descending aorta was excised on cardiopulmonary bypass (CPB) and replaced with a Dacron graft (Terumo, Renfrewshire, UK). The esophagus was closed in two layers. She was discharged 39 days later and did not require further surgery.

3.2. Patient 2

A 2-year-old girl was referred following episodes of hematemesis. Nineteen days before admission, she had had an esophageal button battery (BB) removed at another hospital. She was stable on admission, and a computed tomographic angiogram (CTA) revealed a focal pseudoaneurysm of the right anterolateral wall of the descending aorta at T6/7, consistent with the level of the BB on chest x-ray (CXR).

After a multidisciplinary (MDT) meeting with pediatric surgery, pediatric cardiac surgery, and pediatric intensive care, she underwent left posterolateral thoracotomy and replacement of the severely diseased section of the descending aorta using a Polytetrafluoroethylene (PTFE) graft (W.L. Gore & Associates, AZ, USA). A postoperative upper GI contrast study showed a small esophageal leak which was managed with naso-jejunal feeding. She developed a recalcitrant esophageal stricture at the perforation site requiring dilatations and re-presented with haematemesis 11 months following initial surgery. A pseudoaneurysm was identified at the distal aortic anastomotic site and a covered CP stent (Numed Inc, NY, USA) was deployed to cover the defect. An ongoing esophageal leak caused a stent infection, and she required a Gelweave graft (Terumo, Renfrewshire, UK) replacement of the descending aorta, resection of the diseased esophagus and esophagostomy formation.

Three years later she underwent a right thoracotomy, laparotomy and jejunal interposition replacement of her esophagus with fluorescence angiography support. At last follow up, two years after esophageal replacement (she is eating and drinking normally and the gastrostomy has been removed.

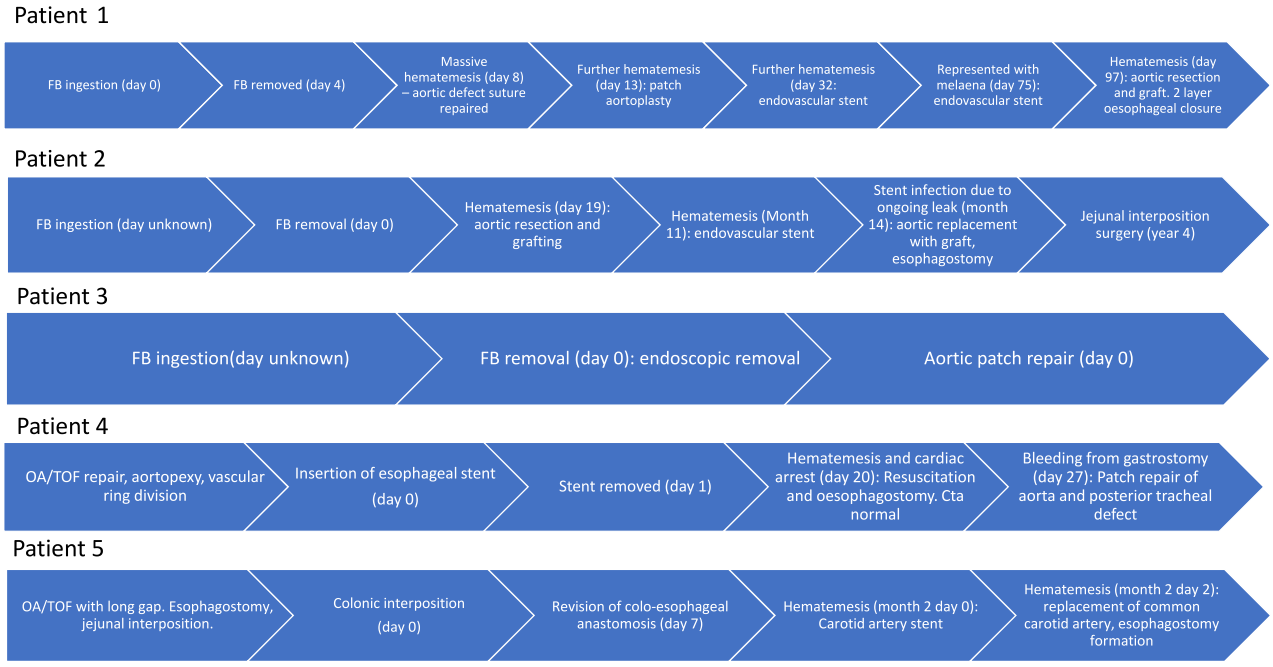

Fig. 1. Patient time lines.

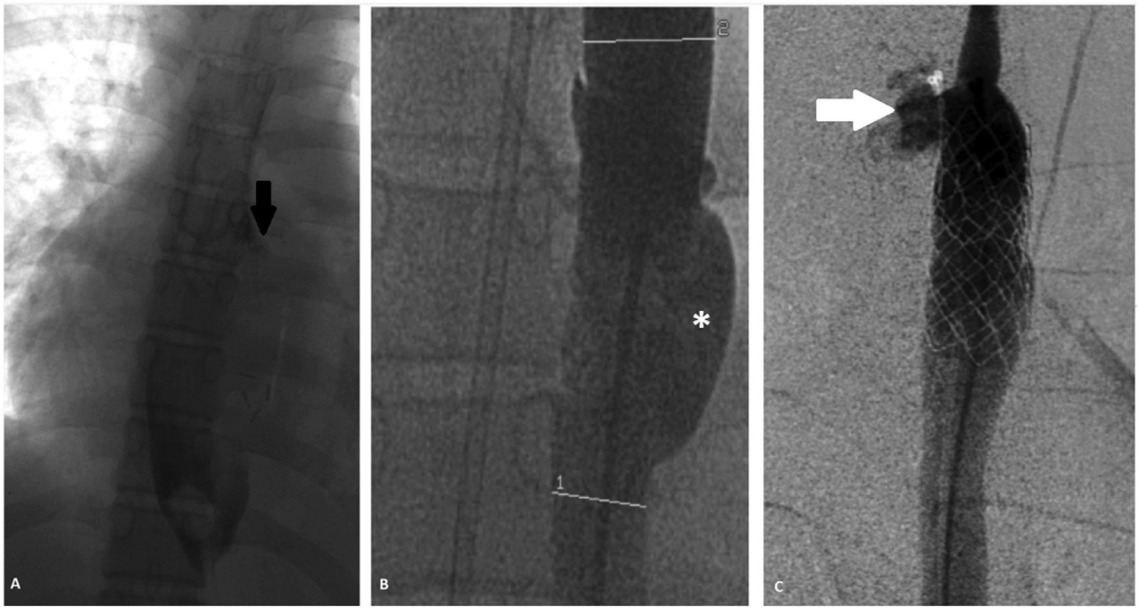

Fig. 2. Images from patient 1. 2A: UGI contrast study showing small leak (black arrow). 2B: Contrast aortogram showing aortic pseudoaneurysm (white star). 2C: Contrast aortogram showing extravasation from descending aorta at the proximal border of the endovascular stent (white arrow) (UGI: Upper gastro-intestinal).

3.3. Patient 3

A previously well 3-and-a-half-year-old boy presented with a history of BB ingestion (BBI), melena and hematemesis. A BB was evident on a CXR. He required fluid resuscitation but was stable enough to undergo a CTA. There was no evidence of aortic involvement, but images were degraded by metal artefact from the BB (Fig. 3A). An emergency MDT involved pediatric surgery, pediatric cardiac surgery, pediatric intensive care, and interventional cardiology. The BB was removed endoscopically but he had slow ongoing bleeding within the esophagus requiring insertion of a SBT under the same general anaesthetic. Following blood transfusion, he remained stable, and a repeat CTA was performed to identify the site and extent of vascular injury. This revealed an aortic pseudoaneurysm at the proximal descending aorta (Fig. 3B). He underwent primary repair via median sternotomy, deep hypothermic CPB and selective antegrade cerebral perfusion, and a bovine patch repair of the aortic injury. The esophagus was mobilized, the injured tissue debrided, and the defect closed primarily using PDS. A flap of lateral pericardium was rotated in situ and interposed between the repair sites. He recovered well with no sequelae.

3.4. Patient 4

A 9-month-old girl with a Gross type C EA underwent open repair as a neonate. She had a friable upper and lower esophagus and a right-sided aortic arch. Following the resolution of a

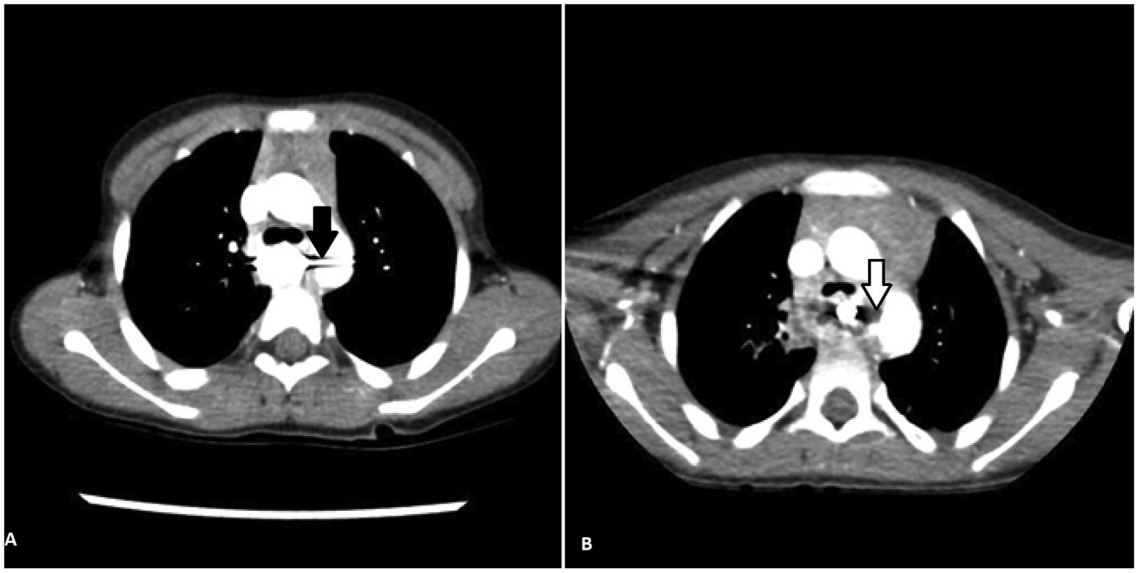

Fig. 3. Images from patient 3. 3A: Axial slice from CTa with BB insitu showing artefact obscuring EVF (black arrow). 3B: Axial slice from CTa following removal of BB and insertion of SBT with obvious EVF (white arrow with black border) (CTa: computed tomographic angiogram; BB: Button battery; EVF: Esophago-vascular fistula, SBT: Sengstaken-Blakemore tube).

significant anastomotic leak and after multiple failed extubations, a CT scan showed a vascular ring formed by the right-sided aortic arch with Kommerell diverticulum and an aberrant retro-esophageal left subclavian artery. A vascular ring division and aortopexy were performed. She recovered well but required weekly esophageal dilatations for an esophageal stricture and suffered several perforations.

To prevent these, an esophageal stent was placed, but 24 h after insertion she required invasive ventilation. Imaging showed the stent compressing the trachea by 50% and it was removed the same day. Nineteen days following removal, she presented to the emergency department with hematemesis and suffered a pulseless electrical activity cardiac arrest. Following resuscitation, esophagoscopy failed to identify a bleeding point and an esophagostomy was formed. She recovered well but following further bleeding from her gastrostomy 1 week later, CTA (Fig. 4) revealed a pseudoaneurysm in the lesser curvature of the distal aortic arch neighbouring the residual esophagus.

Emergency surgical repair was performed on CPB with deep hypothermic circulatory arrest. The aortic defect was excised and patched with bovine pericardium. Unfortunately, the stent had also created an acquired tracheo-esophageal fistula (TEF) so the esophageal end, distal to the esophagostomy, was resected; the tracheal defect required tracheal transection and reconstruction with an autologous pericardial patch. She recovered well but suffered a bilateral vocal cord palsy requiring a tracheostomy.

3.5. Patient 5

A 6-year-old boy was born with Gross type C EA. Initial surgery was challenging, and he required an esophagostomy. A jejunal interposition in infancy failed requiring redo esophagostomy formation. At another institution gastric pull-up surgery was abandoned due to bleeding from the subclavian vessels and retrosternal colonic interposition was performed. The proximal anastomosis was noted to be immediately adjacent to the left common carotid artery (LCCA). He had a proximal anastomotic leak during his inpatient stay which required revisional surgery.

Following discharge, he presented with hematemesis and was transferred to our unit. Although stable on admission he had a further massive hematemesis the following day. The bleeding site could not be identified on endoscopy or DSA and CTA showed dilatation of the LCCA without extravasation. After another massive hematemesis, a repeat DSA showed a significant LCCA to esophageal fistula (Fig. 5A–D) A 7 × 25 mm covered Viabhan stent (W.L. Gore & Associates, AZ USA) was placed which controlled the leak, but four days later a further hematemesis required large volume transfusion.

DSA showed no active bleeding but no reliable cross-collateral flow through the circle of Willis (Fig. 5E). An MDT meeting was convened with pediatric surgery, pediatric cardiac surgery, vascular surgery, pediatric intensive care, and interventional radiology. In conjunction with his parents, the decision was made to defunction the esophagus and replace the LCCA.

A temporizing elongated arterial stent was placed and surgery the following day was performed on CPB at 25 °C with an additional perfusion cannula in the distal left internal carotid artery. The diseased LCCA was excised and replaced with an expanded PTFE graft (W.L. Gore & Associates, AZ, USA) and a contralateral esophagostomy formed.

He had minor frontal lobe changes on postoperative CT and was discharged after a period of convalescence. During this time his carotid graft thrombosed, but slowly enough for collateral circulation to develop.

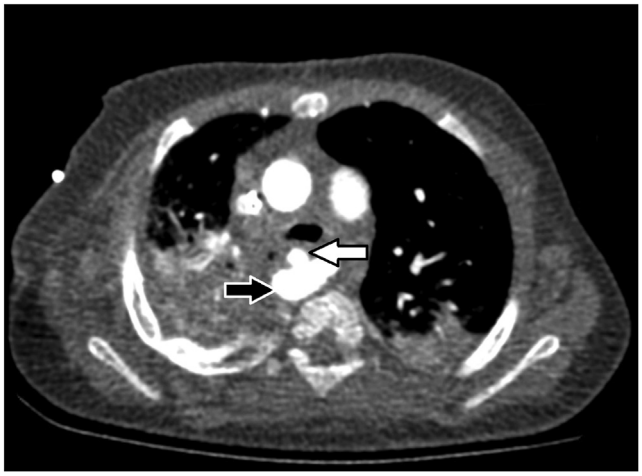

Fig. 4. Image from patient 4 Axial slice from CTa of patient 2 showing apex of aortic arch (black arrow with white border) and pseudoaneurysm (white arrow with black border).

4. Results

Five patients (1M:4F) were identified (Table 1). Median age at the initial presentation of fistulation was 44 (8–177) months. The median length of time between presentation and requirement for combined entero-vascular surgery was 16 (0–419) days. One patient had two major vascular procedures prior to combined surgery. The length of stay on PICU following combined surgery was 6 (2–60) days and overall length of hospital stay was 53 (15–84) days. The median follow-up was 51 (17–61) months.

All patients had a herald bleed.

Two patients had neonatal management of EA/TEF; the other three had no co-morbidities. In four patients, a FB caused fistulation, one of which was iatrogenic (esophageal stent), and in the other three, an ingested FB was at fault (2 BB, 1 chicken bone). One patient had an esophago-carotid fistula; all others had an esophago-aortic connection. All patients survived.

Four patients required esophagostomy formation, of whom one has undergone successful esophageal jejunal replacement at 34 months following combined surgery. In addition, one patient required a tracheostomy as part of their long-term management due to iatrogenic injury to recurrent laryngeal nerves bilaterally.

All parents required psychological input with some needing extended communication and support. In addition, the patients had age-appropriate social and psychological support.

4.1. Proposed guideline for management

We propose the following guideline for the management of EVF (Fig. 6). The main concepts are early recognition, resuscitation and stabilisation (if feasible) which allows cross-sectional imaging to be performed. A subsequent MDT approach with pediatric surgery, pediatric cardiac surgery, pediatric anaesthesia, pediatric intensive care, and interventional radiology is then required. In addition, a vascular surgeon may be needed in patients who do not have great vessel involvement.

5. Discussion

Esophago-vascular fistulae are almost always fatal. They comprise some of the most challenging and technically demanding

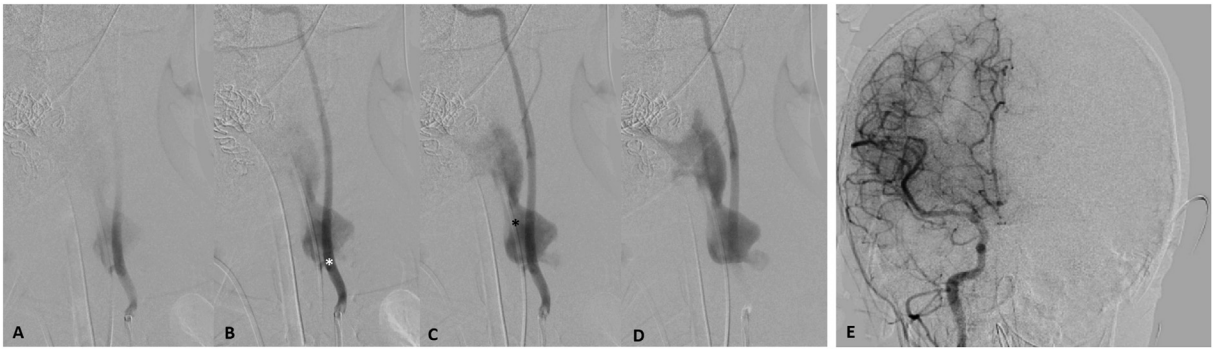

Fig. 5. Images from patient 5. 5 A–D: digital subtraction angiography showing carotid to esophageal fistula (white star) and contrast entering the pharynx (black star); 5E: neuroangiogram showing no reliable cross-over flow between. cerebral hemispheres.

cases any surgeon faces. Surgery is performed in anatomical areas with little margin for error, under extreme time pressures, with high risks of significant morbidity and intra-operative mortality. We report a cohort of five consecutive children with EVF's, who have survived to discharge from hospital. They have all had high-risk life-changing surgical interventions, and some still require major reconstructive surgery, but their outlook is much brighter than generally considered.

EVF can follow FB erosion and can be directly associated with anatomically adjacent procedures. In adults, EVF are described in case reports and short series following ingestion of fish bones [10,11]. Unfortunately, the diagnosis is often delayed as the small volume herald bleed is commonly dismissed without the knowledge that it foretells major haemorrhage.

Diagnostic difficulties may be associated with the rarity of the pathology; two cases of EVF were identified in a series of 2394 patients who had an ingested FB [19]. Within the paediatric population, FB ingestion, especially BB's requires prompt management, but the diagnosis can be challenging especially following unwitnessed ingestion and presentation may be delayed. The development of EVF has been reported to form days or weeks after BB removal [20–23]. Atlas et al. [24] reported on 65 patients over 10 years following BBI with one developing a EVF and surviving to discharge. It is useful to note that the two patients who developed life-threatening complications in this series (the other was a TEF) did not have these identified by serial inpatient screening and presented following discharge.

Akinkugbe et al. [25] reported on vascular complications following BBI between 1977 and 2021 in patients recorded on the National Capital Poison Centre BB registry. Of 361 cases, 51 involved an EVF and only nine of these patients survived to discharge giving a baseline mortality rate of 82%. Imaging and surgical techniques will have changed over this period, but the authors noted that impaction time was statistically significantly different with a median of 96 h in fatalities and 36 h in survivors. The commonest vascular injury was an aorto-esophageal fistula which occurred in 39 patients with 9 surviving, but no long-term outcome data is available. Vascular repair in these children ranged from oversewing to resection and anastomosis or insertion of a graft.

We advocate resuscitation with permissive hypotension, CTA, and DSA where feasible, temporizing with a stent if possible or operating if not. The utilization of endovascular stents as a temporizing measure before definitive surgery has been described successfully in adults [26–30] and in a child following BBI [31]; but when coupled with enteric contamination, infection can be problematic, as seen in patient 2 in our series. We found that in our series, cross-sectional imaging could be performed even in patients who had been resuscitated following cardiac arrest and DSA could be performed in patients who were actively bleeding. Imaging

Table 1
Patient demographics, aetiology of EVF, surgical intervention, follow up and current status (BB: Button battery; EA: Esophageal atresia; CPB: Cardio-pulmonary bypass).

Patient	Age (year of presentation)	Gender	Aetiology	Presenting feature	Definitive surgical procedure	Sequelae or further surgery undertaken	Follow up (months) since definitive surgery	Current status
1	15yo (2005)	F	Chicken bone	Haematemesis	CPB, Primary esophageal repair + descending aortic replacement with dacron graft	None	17	Transferred to adult services for ongoing follow up
2	2 yr (2017)	F	BB	Haematemesis	CPB, Subtotal esophagectomy, replacement of descending aorta with Gelweave graft, formation of esophagostomy	Jejunal interposition performed 2021	52	Ongoing follow up
3	3 yr (2017)	M	BB	Haematemesis	CPB, Primary esophageal repair + aortic repair (bovine patch)	None	61	Ongoing follow up
4	9 mo (2018)	F	Type C EA	Haematemesis	CPB, esophagostomy formation, aortic bovine patch repair, tracheal reconstruction with bovine patch	Bilateral recurrent laryngeal nerve injury requiring tracheostomy	51	Tracheostomy decannulation planned followed later by oesophageal replacement
5	6 yr (2018)	M	Type C EA + esophageal replacement	Haematemesis	CPB, Gortex graft replacement of left common carotid artery, division of colonic interposition with retrotracheal transfer of esophagus for esophagostomy.	Requires monthly esophagostomy dilatations	45	Awaiting reconnection

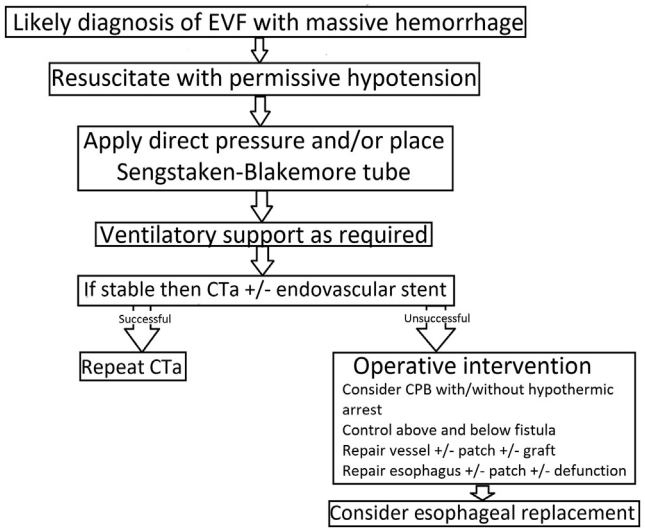

Fig. 6. Proposed guideline for management.

along with temporising stenting allows enough time for surgical MDT planning ensuring that as much anatomical knowledge as possible is gained prior to operating.

Patient 1 was our initial experience of successful management of an aorto-esophageal fistula, and we hypothesized that the reason for ongoing fistulation and bleeding was enteric fluid bathing the vessels and causing ongoing damage. Following further iterations of management, we viewed stenting as a temporizing measure because unless the leak is managed, stenting will inevitably fail as the saliva erodes the vessel wall proximal or distal to the device.

Patients 2,3 and 4 all clinically had aorto-esophageal fistula having presented with hematemesis, but we noted that on the CTa there were pseudoaneurysms at the site of the fistulation which usually, but not always abutted the esophagus. This is not an uncommon scenario as usually the patient needs to be stable enough to be put through the CT scanner. This means that the vascular side of the defect must have developed a clot and thus the bleeding settled, only to recur in time as happened in patient 4.

We form an esophagostomy at the same time as repairing the damaged vasculature if there is no other option. Unfortunately, this usually commits the patient to an esophageal replacement which should be undertaken after convalescence as an outpatient.

If a robust enteric repair is feasible, it may require interposition of muscle or pleura and should be undertaken in the knowledge that ongoing salivary leakage may predispose to graft infection or ongoing arterial damage requiring further intervention. Postoperative care in a pediatric intensive care unit is mandatory with close monitoring for re-bleeding. Preventative measures and/or treatment of mediastinal infection requires broad-spectrum antibiotics with drainage where required.

Once the repair has been facilitated and the outcome known, psychological input for parents and age-appropriate input for the patients should not be forgotten. For those parents who witnessed their child's massive hematemesis and subsequent near-fatal outcome, it was a life-changing experience. More so in those who watched their child suffer a cardiac arrest. In our series the 15-year-old girl required significant input with the other four being too young to understand the sequence and significance of events. They had play therapy and age-appropriate input. However, they may need extra support as they get older and begin to consider the long-term implications of their interventions.

There are limitations to this paper, not least due to the fortunately small number of cases. Obtaining UK wide data is a possibility but there is no national database and interrogating UK hospital episode statistics is fraught with inaccuracies. Even if data was obtained there will sadly be patients who have had EVF in the community and succumbed prior to reaching tertiary-level care who we are unable to identify. Therefore, it is and would be impossible to draw statistically relevant comparisons between these two groups in terms of prognostic factors or time of intervention.

6. Conclusion

Successful management of EVF should be individualized. It should involve a team approach with pediatric surgeons, pediatric cardiac surgeons, pediatric cardiologists, pediatric intensivists and anesthetists, vascular surgeons, and pediatric radiologists to perform the appropriate interventions safely. Following imaging, surgical aims should be to repair the vessel and prevent further damage along with diverting salivary flow or ensuring robust enteric repair where appropriate.

Psychological support in the short, medium, and long term for the patient and their parents should be offered routinely.

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CI's.

Declaration of competing interest

None.

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