SHORT REPORT

Primary Aortoenteric Fistula Due to a Swallowed Twig in a Three-year-old Child

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Abstract
Aortoenteric fistulae are infrequent causes of gastrointestinal bleeding and usually occur as a late complication of aortic aneurysm repair. Primary aortoenteric fistulae are very rare and most have an aetiopathological association with atherosclerotic aneurysmal disease.1,2 We report a primary aortoenteric fistula (PAEF) occurring in a 3 year old as a result of duodenal perforation after ingestion of a twig. To our knowledge this is the first case of a primary aortoenteric fistula reported in a child. Emergency aortic surgery in children needs certain considerations which are briefly discussed.

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Introduction

Aortoenteric fistulae present as gastrointestinal bleeding, usually complicating aortic aneurysmal disease or its repair.1,2

We report the first description of an aortoenteric fistula in a 3 year old resulting from duodenal perforation after ingestion of a twig.

Case report

A 3-year-old South Asian boy presented with a four day history of abdominal pain, rigors and loose black stool shortly after returning from a brief stay in Pakistan. WCC and CRP were raised with a reduction in haemoglobin and serum albumin. Faecal occult blood was positive but microscopy and culture were unremarkable. Blood cultures grew klebsiella, coagulase negative staphylococcus and streptococcus viridans and treatment for Gram negative septicemia was initiated with meropenem, cefotaxime and metronidazole. His symptoms persisted and an episode of haematemesis prompted transfer to the regional paediatric centre. Abdominal ultrasound was reported as normal. A further episode of massive gastrointestinal bleeding necessitated gastroscopy which revealed fresh blood in the stomach but no clear source. During the procedure he deteriorated dramatically and emergency laparotomy was undertaken. Gastroduodenotomy revealed bleeding from the distal duodenum, which was adherent to the aorta with a surrounding inflammatory mass and a diagnosis of aortoduodenal fistula was considered.

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Via a medial visceral rotation the supra-coeliac aorta was controlled. Common iliac arteries were then exposed and controlled, carefully safeguarding the clearly visible nervi erigentes. The distal duodenum was mobilised to reveal a large aortoduodenal fistula along with a 5 cm twig (Fig 1) lying within the inflammatory mass. The infra-renal aorta was freed from the surrounding dense inflammatory tissue and the proximal clamp repositioned below the visceral arteries. The fistula margins were resected back to healthy vessel resulting in a 4 cm long near-circumferential defect in the aortic wall. Superficial femoral vein, harvested from the left thigh was anastomosed as an interposition graft with interrupted 5/0 prolene. The duodenal rent was closed with interrupted 4/0 PDS and an omental pedicle was interpositioned between the duodenum and aorta to reduce risk of recurrent fistulation. The supra-coeliac and infra-renal clamp times were 45 and 80 min respectively. The operation lasted 4 h 45 min and blood loss was estimated at 4500 ml (his total blood volume was estimated at 1600 ml). He was transfused 10 units of packed RBCs, 5 units of fresh frozen plasma and 2 bags of platelets. Multi-agent anti-microbial therapy with linezolid, teicoplanin, metronidazole and fluconazole was given for four weeks post-operatively.

His post-operative course was complicated by haemorrhage from the gastroduodenal suture line. At re-operation the aortic reconstruction (Fig 2) was found to be intact. He was discharged on day 26 with a two week course of ciprofloxacin and trimethoprim. At six weeks follow up, he remains well with normal flow through the vein graft on duplex examination.

Discussion

PAEF are predominantly associated with atherosclerotic aortoiliac aneurysms.1,2 Non-aneurismal PAEF are very rare but have been reported in association with carcinoma, duodenal TB, tuberculous aortitis, following treatment with intra-vesical Bacille Calmette-Guerin, diverticular abscess, septic aortitis, radiotherapy, gallstones, duodenal ulcer, Takayasu’s arteritis, cystic medial necrosis and severe non-aneurismal aortic atherosclerosis.

An ingested foreign body has been implicated in four reported cases of PAEF.3,4 In children, one death has been reported from PAEF arising secondary to duodenitis following orthotopic liver transplantation.

To our knowledge, this is the first successfully treated PAEF in a child in literature. It appears that the ingested twig had perforated the duodenum resulting in localised retroduodenal sepsis, with contiguous aortitis and eventual aortoenteric fistulation.

This case presented several challenging aspects. Understandably PAEF would never feature in the differential diagnoses of paediatric gastrointestinal bleeding and diagnosis was only established at operation, precluding a planned surgical treatment. The initial challenge was proximal control. This was achieved with supra-coeliac clamping and despite prolonged ischaemic time of 45 min, subsequent reperfusion was well tolerated, and there was no evidence of renal dysfunction post-operatively.

Aortic reconstruction in small children has some special considerations. Synthetic conduits (Dacron, PTFE) have been used in paediatric aortic reconstruction but concern regarding recurrent infection precluded their use and extra anatomic reconstruction was rejected due to the narrow calibre of the child’s arteries, unavailability of appropriate conduit and lack of potential for growth. Autologous long saphenous vein and hypogastric artery have been used but are of narrow calibre. More recently the use of decellularised, antigen-reduced cryopreserved pulmonary artery allografts has been described.5 These grafts would be more resistant to infection and retain growth potential, but are unavailable ‘off the shelf’.

The superficial femoral vein was selected as it closely matched the calibre of the aorta (6 mm) and could be used for anatomic reconstruction in a contaminated bed while retaining potential for growth. Post-operative deep vein thrombosis and limb swelling have been reported after use of the superficial femoral vein, but was not seen in our case.

Figure 1  Twig found in the aortoduodenal fistula. Each mark on tape = 1 mm.

Figure 2  Appearance at re-operation, of aortic reconstruction using reversed common femoral vein as interposition graft. The aortic bifurcation appears dark due to use of Surgicel for haemostasis at the initial operation.
This graft’s fate is uncertain with risk of degenerative aneurysmal dilatation and conversely also stenotic disease if it lags behind the aorta as the child grows especially around puberty; long term graft surveillance is hence needed. The authors propose yearly ultrasound examination; patient compliance with CT and MR study is likely to be poor and repeated CT would entail a cumulative contrast and radiation risk.

Conflict of Interest
None declared.

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