CASE REPORT

Cervical diverticulitis: a novel complication of a neonatal colonic interposition graft following oesophagectomy

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SUMMARY

A 47-year-old woman presented to a tertiary emergency department with an 8-day history of odynophagia, a 4 cm swelling on her left neck and intermittent fevers. Shortly following her birth, a congenital oesophageal atresia had been managed surgically with colonic interposition graft. Contrast CT of the neck demonstrated several large diverticula within her interposition graft at the level of the cervical vertebrae. A colocutaneous fistula was identified between the colon and left neck, with an associated abscess. The patient received intravenous meropenem followed by abscess drainage. A high output fistula developed at the drainage site, and the patient required intravenous fluids and stoma placement to manage fluid discharge. She left the hospital after a 17-day stay. At 6-month follow-up, the wound was erythematous, but the patient was otherwise well. We believe that this is the first reported case of diverticular disease arising in what was originally neonatal colon interposed for oesophageal atresia at birth.

BACKGROUND

This is the first reported case of diverticular disease arising in a neonatally placed colonic interposition graft for oesophageal atresia. This may be because neonatal colon used for interposition may not be sufficiently exposed to standard causative factors for diverticular development prior to resection, therefore minimising development of diverticula within the colonic graft.

In addition to being an example of a previously unreported presentation of a common disorder, this case raises questions regarding the pathogenesis of diverticular disease and emphasises the importance of treatment by first principles in the absence of established guidelines.

CASE PRESENTATION

A 47-year-old woman presented to a tertiary hospital emergency department with an 8-day history of gradual onset odynophagia, swelling on the left lateral aspect of her neck and intermittent fevers. She had a history of congenital oesophageal atresia, which had been managed surgically with colonic interposition graft shortly following birth. Her only ongoing medication was esomeprazole for gastro-oesophageal reflux.

INVESTIGATIONS

On physical examination, the patient had a well-defined, subcutaneous mass of 4 cm diameter on

the left lateral aspect of her neck. It was erythematous, warm and fluctuant. There was minor purulent discharge from the wound opening, but no blood. She reported no difficulty in turning her head, no sensory loss and only mild tenderness at or around the site.

Blood tests revealed neutrophilia $9.5 \times 10^9/L$ (normal $4.0\text{--}7.5 \times 10^9/L$), thrombocytosis $616 \times 10^9/L$ (normal $150\text{--}400 \times 10^9/L$) and elevated C reactive protein 355.8 mg/L (normal <5 mg/L), consistent with infection and/or inflammation. Ultrasound imaging suggested local colonic wall thickening in the interposed conduit, associated with a subcutaneous collection. A contrast CT of the neck allowed visualisation of the interposed segment of colon, which was seen to run anterior to the left carotid artery before entering the superior mediastinum along the left lateral aspect of the thoracic inlet. Several large diverticula of the interposed colon were noted at the level of the cervical vertebrae. A colocutaneous fistula was identified between the colon and the left neck skin with an associated abscess. A contrast swallow examination additionally revealed an extraluminal leak of oral contrast (figure 1).

These features were consistent with colonic diverticulitis at the level of the cervical neck, complicated by colocutaneous fistula and subcutaneous abscess.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis included superficial cutaneous abscesses, lymph node pathology, branchial cysts or carotid body tumours.

TREATMENT

The patient received intravenous meropenem for 2 days to manage the systemic sepsis and then underwent abscess drainage. Postoperatively, the patient received oral co-amoxiclav, together with paracetamol and oxycodone for analgesia. A high output fistula developed at the site of operative drainage and the patient required intravenous fluid support, careful management of the surrounding skin and the use of a stoma appliance over the site of fistulation, to collect discharging fluid.

OUTCOME AND FOLLOW-UP

The patient was discharged after a 17-day hospital stay, and was requested to monitor her wound site for excoriation or increased fluid leak. When reviewed at 6-month follow-up, there was persisting mild erythema surrounding her fistula site, with

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Figure 1 Postcontrast CT of the head and neck showing diverticulitis of colonic interposition graft and colcutaneous fistula (arrows) at the level of cervical neck in coronal (A) and axial (B) views.

gas and serous fluid expressible from the opening. However, she was well, and her symptoms eventually resolved without intervention. No further imaging was undertaken.

DISCUSSION

Colonic interposition surgery has long been used to reconstruct the oesophagus, and for oesophageal atresia in neonates, traumatic caustic ingestion and oesophageal malignancy.^{1 2} The most common symptom indicating surgery is chronic, persisting dysphagia. Surgically, the left-sided colon is generally preferred over right-sided colon for interposition, as the left colon has a smaller diameter, increased length, better blood supply and better bolus-propulsion ability, which all contribute to the functionality of the neo-oesophagus.³ In addition, less gastric regurgitation is described in postcolonic interposition grafts when compared to gastric conduits; however, colonic interposition does require a longer, more technically complex operation.⁴ Early complications following interposition surgery most often involve graft ischaemia and anastomotic leak or stricture.^{5 6} Late complications of this type of interposition are rare, but include progressive graft fibrostenosis, gastrocolic reflux and colopericardial or colobronchial fistula.^{3 7}

Diverticular disease of the colon is found in approximately 60% of patients over 80 years of age.⁸ The most common location diverticula develop in is within the sigmoid colon.⁸ While the condition is asymptomatic in most affected people, complications do occur and can be severe, including infection, abscess formation, fistulation, perforation, intestinal obstruction and bleeding.⁸

We believe that this is the first reported case of diverticular disease arising in a neonatal colonic interposition graft placed at birth. This may be because the neonatal colon used for interposition may not be sufficiently exposed to the recognised causative factors for diverticular development prior to resection, which may minimise development of diverticula within the interposed graft. Furthermore, it is possible that other patients who received interposition surgery at birth may also have developed diverticular disease within their interposed colo-oesophageal grafts but remained asymptomatic, and thus have not presented for care. Patients who have undergone interposition grafting at birth are now surviving longer, thus similar cases may be described in the future.

Development of diverticular disease has rarely been described post-colo-oesophageal graft for malignancy or caustic injury, but in each case where diverticula developed, interposition grafting

occurred in the patient's 5th–7th decade, when early diverticular disease may already be underway within the patients' large intestines.²

The two most significant risk factors for diverticular disease are age over 60 years, and a low-fibre diet; however, further research continues.^{9 10} Our patient was relatively young, reported a healthy diet, and despite having a neonatally placed colonic interposition graft, developed diverticular disease several decades post-interposition. The aetiology of diverticulosis in her case is unclear; however, management of her local and systemic symptoms was not significantly altered in comparison to treatment of diverticulitis.

Learning points

- ▶ This is a previously unreported presentation of a common disorder.
- ▶ This case raises questions regarding the pathogenesis of diverticular disease.
- ▶ The importance of treatment by first principles in the absence of established guidelines is emphasised.
- ▶ A thorough medical and surgical history, blood examination and contrast CT imaging were important in confirming the correct diagnosis.
- ▶ Soft tissue imaging was particularly important as it helped to rule out similar presentations such as superficial cutaneous abscesses, lymph node pathology, branchial cysts or carotid body tumours.

Contributors VVP wrote and edited the case report, and examined the patient. EO and MWH treated the patient, supervised the writing of the report and edited the report.

Competing interests None declared.

Patient consent Obtained.

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