Lymphangioma of the jejunal mesentery and jejunal polyps presenting as an acute abdomen in a teenager

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ABSTRACT

Cystic lymphangioma of the small bowel mesentery is a rare clinical entity, especially after childhood. Medical literature reveals a limited number of such cases presenting as acute abdomen due to bowel obstruction, small bowel volvulus and bleeding into the tumour. We present the management experience of an 18-year-old woman who presented with rapid onset diffuse peritonism and raised inflammatory markers. Computed tomography showed a mass in the small bowel mesentery with suspicion of segmental bowel ischaemia. Emergency laparotomy revealed a mass in the mid-jejunal mesentery close to the bowel wall with no bowel ischaemia. The patient made an uncomplicated recovery after segmental bowel resection and end-to-end anastomosis. Histology confirmed the mass as a cystic lymphangioma involving the jejunal mesentery and two small jejunal polyps. Lymphangioma could be considered in the differential diagnosis of an acute abdomen in a young adult when the presentation is atypical.

KEYWORDS

Cystic lymphangioma – Mesenteric lymphangioma – Jejunal lymphangioma

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Introduction

Cystic lymphangioma of the small bowel mesentery is rare and the incidence is around 1 per 100,000 admissions in paediatric hospitals.¹ The entity is even uncommon beyond childhood and medical literature reveals less than 250 reported cases overall. The majority of adult cases have presented as acute abdomen due to small bowel volvulus, bowel obstruction or bleeding into the tumour.^{1–4} There are infrequent reports of incidentally detected mesenteric lymphangioma of the jejunum.^{4,5} We present the management experience of a female teenager who presented with diffuse peritonism of rapid onset and raised inflammatory markers.

Case history

An 18-year-old previously healthy woman, who was well and active until the onset of symptoms, was admitted as a surgical emergency with acute severe abdominal pain and vomiting for 4 hours. She had no previous abdominal surgeries and denied lower gastrointestinal symptoms. She was not pale but had a mildly elevated temperature (37.2°C). Other observations were stable except for a marginal tachycardia. Her abdomen was diffusely tender with evidence of early peritonism, which was more pronounced in the central

abdomen and left upper quadrant. There were no masses, organomegaly or ascites. Haematological investigations showed normal haemoglobin levels, leucocytosis of 13900/mm³ with predominance of neutrophils, elevated C-reactive protein of 63 nmol/L and normal serum amylase levels. Computed tomography showed a low-density mass lesion in the small bowel mesentery encasing the mesenteric vessels with a suspicion of bowel ischaemia (Fig 1).

The woman underwent an emergency laparotomy under general anaesthesia. At laparotomy, there was a well-demarcated 10 × 10 × 9 cm mass lesion with cystic and solid areas in the jejunal mesentery adjacent to the bowel wall, about 70 cm from the duodenojejunal flexure. There was no bowel ischaemia (Fig 2) and the rest of the peritoneal survey was unremarkable. The mesenteric mass was excised with about 15cm segmental jejunal resection and bowel continuity was achieved by end-to-end hand-sewn anastomosis. The post-operative period was uncomplicated and the patient was discharged on the second postoperative day. She remained well at 6 months from the surgery.

Macroscopically, there was a yellowish-white mass in the mesentery adjacent to the jejunal wall with a spongy appearance in the cut surface due to closely packed cystic spaces with no obvious involvement of the bowel wall. Two small polypoidal lesions were seen in the opened-up jejunum near

Figure 1 Low-density mass seen in the mesentery (white arrow) encasing the vessels seen on cross-sectional computed tomography.

Figure 2 Perioperative image of the mesenteric mass after being delivered to the laparotomy wound.

the mass (Fig 3). Microscopy of the mesenteric mass contained closely packed thin-walled lymphatic spaces containing amorphous eosinophilic material, neutrophils and lymphocytes, confirming a cystic lymphangioma of the jejunal mesentery. Both jejunal polyps were lymphangiomas. The surgical excision was complete.

Discussion

Lymphangiomas are childhood benign tumours and almost 90% of the reported cases have manifested before the age of 3 years.^{2,3,5} The head, neck and axillae account for more than 95% of lymphangiomas and isolated intra-abdominal lesions arise in less than 1% of the cases.³⁻⁵ The exact pathogenesis of lymphangioma formation is unknown. It is commonly believed to be caused by anomalous development of the lymphatic system, although further postulations have

Figure 3 Macroscopic appearance of the resected specimen with a prominent yellow-white mass in the mesentery adjacent to the jejunal wall. Note the closely packed cystic spaces giving a spongy appearance (white arrows); two small polyps in the jejunal mucosa near the mass can be appreciated (black arrows).

been made about the association of abdominal trauma, surgery, radiation and inflammation.^{3,5} Histologically, lymphangiomas have been classified into three types as capillary, cavernous and cystic lesions. Cystic lymphangiomas, as in the index case, would contain large lymphatic spaces containing chylous or serous fluid with no formal communication between the adjacent lymphatics.

Most cases of adulthood mesenteric lymphangioma have presented with acute abdomens from bowel obstruction, tumour rupture, anaemia with bleeding into the lesion and bowel gangrene.¹⁻⁵ As there was no obstruction, bleeding or tumour rupture, the mechanism that led to the acute abdomen in the index case is unclear. Ultrasonography and computed tomography have been generally very sensitive in providing structural details of the lesions but there have been inadequacies in establishing a preoperative diagnosis.^{1,5} The main modality of treatment for mesenteric lymphangioma is adequate surgical excision, because of the risk of recurrence and malignant transformation, particularly after radiotherapy for the primary lesion.^{3,6} These lesions are rare but this should not prevent the surgeon from including them in the differential diagnosis when considering an atypical case of acute abdomen in a young adult.

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