Rare disease

Bleeding small bowel cavernous haemangioma following blunt trauma to the abdomen presenting as subacute intestinal obstruction in a child

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Summary

The authors report a case of a 6-year-old girl who developed subacute intestinal obstruction after a trivial blunt trauma to her abdomen. Her normal vital signs masked the presence of intestinal bleeding. An incidental finding at surgery of a haematomatous polypoid vascular growth of the ileum was subsequently confirmed to be cavernous haemangioma of the small bowel. Surgical resection was curative in this patient.

BACKGROUND

Vascular malformation of the small bowel is a rare disease. Most common presentation is chronic intestinal bleeding leading to anaemia. It has never been reported in any English literature of a cavernous haemangioma causing intestinal obstruction after a trivial trauma.

Surgical exploration revealed a polypoid lesion filled with haematoma causing obstruction in the ileum. This was the first time we encountered this rare disease. This appearance of this tumour fascinated us. We would also like to share the literature reviews and the definitive treatment to this rare disease.

CASE PRESENTATION

A 6-year-old girl with no known medical or surgical history suffered a mild blunt trauma to her abdomen. The handle bar of her bicycle hit the lower part of her abdomen following a fall while mounting her bicycle. There was no loss of consciousness, ear, nose or throat bleeding. She complained of slight abdominal pain following the trauma. She was admitted into a district hospital for 2 days. During her admission, her vital signs were stable and a plain abdominal x-ray was done which was normal. Her symptoms subsided and she was allowed discharge. However, at home she was not able to take orally as per normal. She vomited

Figure 1 Ultrasound abdomen.

Figure 2 The intraoperative picture of bleeding small bowel cavernous haemangioma.

after each oral feeds. The abdominal pain recurred. At post trauma day 5, she became lethargic and her abdomen was distended. She was immediately referred to our institution. On examination, she was afebrile, lethargic, dehydrated and mildly pale. Examination of the abdomen revealed a mildly distended abdomen with no sign of bruising. There were palpable, tender lumpy masses at the lower abdomen. There were no other signs of injuries elsewhere. Her symptoms worsened the next day with bilious aspirate and more frequent abdominal pain. Decision was made to do a diagnostic cum therapeutic laparoscopy.

INVESTIGATIONS

She was anaemic with haemoglobin of 8g/dl. There was no leucocytosis. Her serum amylase was normal. Plain abdominal x-ray did not show evidence of bowel obstruction, instead it showed opacity at the lower abdominal region. Urgent ultrasound of the abdomen revealed no solid organ injuries; there was also minimal fluid in the pelvic cavity. However, there were huge dilated bowels in the lower abdomen, which contained large amount of floating echogenic debris representing blood products, consistent with areas where the lumpy masses were felt. Doppler ultrasound did not show any abnormal vascularity. The ultrasound was reported as possible submucosal haematoma of the small bowel causing luminal obstruction (figure 1).

DIFFERENTIAL DIAGNOSIS

In a child with history of handle bar injury to the abdomen who presents with acute abdomen and pallor, solid organ injuries must be ruled out. Once that is excluded, delayed perforation of the viscous needs to be considered. The two common sites for perforation are duodeno-jejunal junction

and ileo-caecal area, these are sites of the root of mesentery which are fixed and immobile, and hence blow-out perforation can occur with impact. This is most likely in this child. It is possible at this point that the child suffers from a perforated ileo-caecal area with probable intramural haematoma causing bowel obstruction.

TREATMENT

A diagnostic laparoscopy via a 7 mm port supraumbilical incision was performed under general anaesthesia to assess the bowel involved. At laparoscopy, there was a huge bluish small bowel mass seen. It was not feasible to deliver the affected bowel via the umbilical wound even when it was enlarged considerably. The surgery was converted to open. An extended lantz incision was made. The huge, bluish, polypoid small bowel mass was exposed (figure 2). Fifteen cm of affected small bowel was resected. End to end anastomosis was performed with polyglactin 4-0.

OUTCOME AND FOLLOW-UP

Our patient recovered well post surgery. She was discharged home at postoperative day 7. She came for follow-up at 1 month post surgery, she was well. The histopathological examination of the resected bowel was reported as small intestine cavernous haemangioma. Patient was given another follow-up 6 months later, she did not turn up but we received phone call from the parents that she remained well and healthy.

DISCUSSION

The small intestine is the most common site of gastrointestinal haemangiomas, with the jejunum being the most commonly involved segment; haemangiomas represent approximately 7%–10% of all benign tumours of the small

intestine.¹ The colon is the second most common site of gastrointestinal haemangiomas; recto-sigmoid is the most commonly involved segment. Haemangiomas are defined as masses of capillaries, blood filled endothelial lined spaces or a combination of them. Haemangiomas of small bowel are uncommon and accounting for only 0.05% of intestinal neoplasia and 7%–10% of all benign tumours of small intestine.^{2–3} Haemangioma of intestine may be solitary or multiple and they may manifest as a segment of different syndromes; Blue rubber bleb syndrome, Maffucci syndrome and Klippel-Trenaunay-Weber syndrome.⁴ Intestinal haemangioma has also been reported in Peutz-Jeghers syndrome.

Aetiology of haemangiomas is still unknown, but it is believed that these tumours are angiogenesis dependent. This process is tightly controlled by inhibition of endothelial cell growth. Haemangioma could result from a localised diminution of normal angiogenesis inhibition or increased production of angiogenetic stimulator.⁵

Intestinal haemangioma is usually soft and polypoid. It varies from a few millimetres to a large mass projecting into the lumen. Microscopically, cavernous haemangioma is the most common type, followed by roughly equal proportion of tumours classified as capillary or mixed capillary-cavernous. Haemangiomas usually manifest in the form of overt rectal bleeding, abdominal pain, or chronic symptoms of anaemia. Other forms of presentation include intussusception, small bowel obstruction, intramural haematoma and perforation.⁶

Due to the rare nature of the condition, the diagnosis of small bowel haemangioma is only possible after a barrage of investigation especially with the presentation of obscure gastrointestinal bleeding. Traditional techniques to image the small intestine include angiography, nuclear scans, CT enteroclysis and push enteroscopy. These often lack accuracy, and the median time to diagnosis of chronic bleeding lesions can be as long as 8 years.⁷ With the advent of capsule endoscopy, the diagnosis of the above problem is very much made easier and less non-invasive.

Randomised trials have showed that the overall sensitivity is greater with capsule endoscopy than enteroscopy.⁸ Capsule endoscopy has been shown to be safe in paediatric patients, receiving clearance from the US Food and Drug Administration in October 2003.⁹ The incidence of solitary

haemangioma is much lower than that of multiple vascular tumours, with only a few cases reported in literature.¹⁰

Learning points

- Intestinal obstruction following a blunt trauma to the abdomen in a child needs proper and detail investigations.
- Diagnostic laparoscopy is a useful tool to assist management.
- Rare causes should be entertained when clinical presentations are not clear cut.
- In this case, surgical resection is curative.
- This is a rare case with unusual presentation worth sharing with as many clinicians as possible.

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Competing interests None.

Patient consent Obtained.

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