

CASE NOTE

Intestinal duplication presenting with recurrent abdominal pain

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Duplication cysts represent one of many congenital abnormalities of the gastrointestinal tract. It is an uncommon cause of acute abdominal pain but remains a consideration in the pediatric population. To highlight aspects of the disease and the value of exploratory laparotomy in pediatric patients with recurrent abdominal pain, we present the case of a patient who was admitted to hospital several times for recurrent abdominal pain in whom we eventually diagnosed intestinal duplication.

CASE REPORT

A 13-year-old boy presented to the accident and emergency department at the University Hospital of the West Indies with a history of acute onset of cramping periumbilical pain and vomiting. His medical history included multiple admissions over a 6-year period for similar symptoms. Physical examination revealed mild abdominal tenderness but no peritonitis. Soon after admission, his acute abdominal pain resolved. A multidimensional computed tomography (CT) scan of his abdomen revealed a multiloculated cystic mass extending along the root of the mesentery, suggesting a duplication cyst (Fig. 1). The CT scan did not lead to a conclusive diagnosis; however, owing to the abnormality on the CT scan and the recurrent nature of the problem, we obtained consent for exploratory laparotomy. The procedure revealed an ileal duplication 38-cm long with a cystic dilatation that had adhesions to the transverse colon (Fig. 2). The base of the cyst was fibrotic, suggesting that

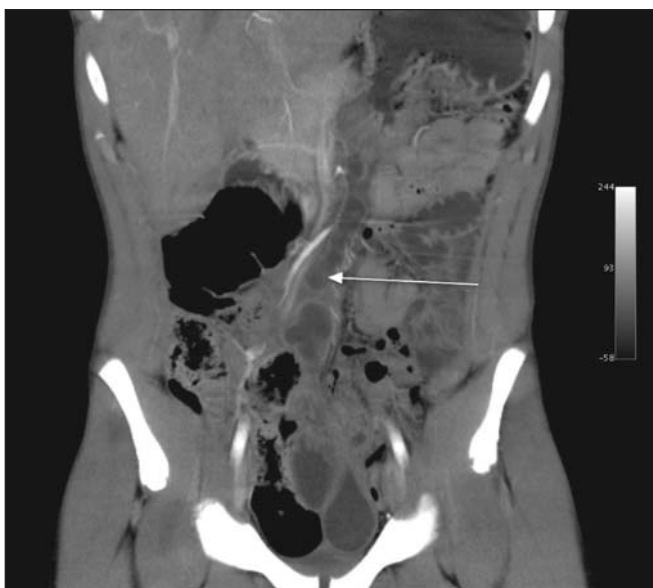


Fig. 1. Coronal volume-rendered image showing a multiloculated cystic mass extending along the mesentery (arrow).

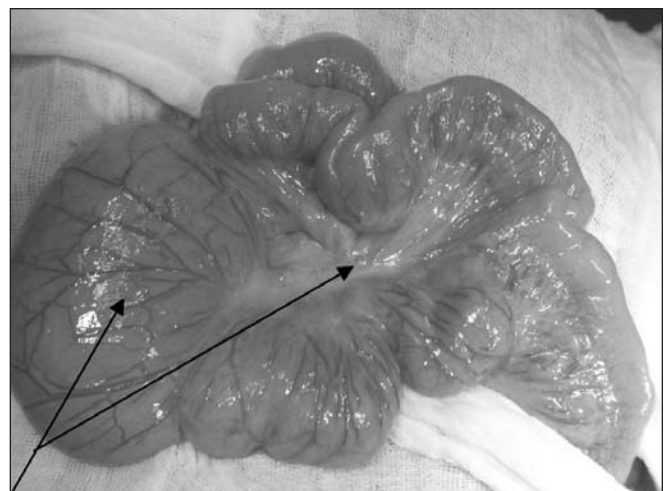


Fig. 2. Laparotomy of the duplicated ileum separating into the cystic mass with its fibrotic base (arrows).

recurrent obstruction at this level resulted in the patient's periodic episodes of abdominal pain. We resected the cyst and the parallel segment of small bowel and conducted a primary anastomosis. The patient had an uneventful post-operative period and was well at final follow-up 1 year after resection.

DISCUSSION

Gross and colleagues¹ first defined intestinal duplication as "spherical or tubular structures that possess a well developed smooth muscle layer and are lined with a mucous membrane; they are found at any level from tongue to anus and usually are intimately attached to some portion of the alimentary tube." One large study² suggests that most duplications are intra-abdominal and, of these, ileal and ileocecal duplications are the most common.

Clinical presentation is similar to that in our patient's case and includes recurrent abdominal pain, vomiting, abdominal distension, gastrointestinal hemorrhage and sometimes peritonitis if a peptic ulcer perforation occurs in the duplicated segment with ectopic gastric mucosa or in the adjacent normal bowel.^{3,4} The duplication can be the lead point in intussusception^{3,4} or the site of volvulus.² Prenatal ultrasound or ultrasound at the time of clinical presentation may lead to the diagnosis of these benign lesions. Differentiation from mesenteric cysts can sometimes be made during that initial ultrasound based on the thinner nature of the intestinal wall. Preoperative diagnosis is more commonly made with intestinal duplications of foregut than of midgut or hindgut origin.⁴

The universally accepted management of this condition is surgical intervention, and the outcome is good in patients in whom it is an isolated congenital abnormality. Recommendations for surgical management are based on the age and condition of the patient; the location, nature and extent of the lesion; and the number of anatomic locations involved. Total excision is generally preferred if possible and will usually involve resection of the involved

normal bowel,⁵ as in our patient's case. This approach is not always feasible, as in patients in whom resection will result in short bowel syndrome or the duplicated segment involves vital structures like the duodenum in close proximity to the common bile duct. In these instances, partial resection, marsupialization or internal drainage may be performed. Blunt dissection without resection or anastomosis of the normal adjacent bowel has been described.⁴ As a majority of these lesions share a common wall, the involved risks include devascularization of the adjacent bowel if the arterial supply is intimately shared and iatrogenic bowel wall injury. The presence of heterotopic gastric mucosa negates internal drainage as adjacent nongastric mucosa is at risk for peptic ulceration, hemorrhage and perforation, and mucosal stripping may be performed if resection is ill-advised.⁴

Reports of the long-term sequelae of untreated intestinal duplications are not numerous in the literature as most patients present with symptoms in childhood. Adult patients present with similar symptoms of recurrent abdominal pain, intestinal obstruction or hemorrhage, or the lesion may be found incidentally at autopsy. Rarely, cancer has been described and is more common in duplications of the large bowel; however, lesions of the ileum may also be involved.⁵

Competing interests: None declared.

References

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