CASE REPORT

Ileal duplication: an unusual cause of intestinal obstruction in adult life

L Barbosa, C Soares, A A Póvoa, J P Maciel

Department of General Surgery, Centro Hospitalar de Vila Nova de Gaia/Espinho, Porto, Portugal

Correspondence to

Dr L Barbosa,
lilitebarbosa@gmail.com

Accepted 19 May 2015

SUMMARY

Gastrointestinal duplications are rare congenital malformations seldom diagnosed in adulthood. They may vary greatly in size and location, with the small intestine being their major focus. Their clinical presentation is widely variable and unspecific, mimicking more common pathologies, thus making preoperative diagnosis very difficult. The intraoperative surgeon's experience and knowledge are crucial in recognising these lesions so that they can be correctly managed. In this report, the authors present a case of a 36-year-old man with an acute intestinal obstruction as the first presentation of ileal duplication.

BACKGROUND

Gastrointestinal or enteric duplications are rare congenital anomalies often presenting before the age of 2 years, with a minority remaining asymptomatic until adulthood.^{1–4} These duplications consist of spherical cysts or, less frequently, tubular structures that tend to be located on the mesenteric side of the alimentary tract sharing with it a common muscular wall and blood supply. Enteric duplications can occur anywhere along the gastrointestinal tract from mouth to anus, but the small intestine is the most prevalent site of occurrence. Ileal duplications are the most frequent, representing about half of the cases, followed by the oesophagus, colon and jejunum.^{3 4}

Their pathology has a wide range of clinical presentation in adults, and depends on location, size and type of duplication as well as the presence of complications.⁵ Owing to their rarity and unspecific symptoms, an accurate preoperative diagnosis is very difficult.^{1 3} Malignant transformation has been reported in ileal duplications and therefore complete surgical resection is the recommended treatment.^{6 7}

In this report, the authors present the case of a 36-year-old man with an ileal duplication revealing an acute intestinal obstruction.

CASE PRESENTATION

A 36-year-old previously healthy man was admitted to our hospital with an acute onset of persistent abdominal pain and vomiting. Physical examination revealed abdominal distension and diffuse rebound tenderness on palpation. Laboratory data showed only a slightly elevation of white cell count (17 800 with 86% of neutrophils) and the plain abdominal radiograph in erect position demonstrated intestinal obstruction with the presence of multiple air–fluid levels. An abdominopelvic CT scan was performed,

which showed a mass of adherent small bowel ileal loops along with dilated, wall-thickened proximal small bowel loops, with air–fluid content (figures 1 and 2).

In the presence of intestinal obstruction, a laparoscopic approach was not considered and the patient underwent an exploratory laparotomy, revealing a mass involving the proximal ileum with multiple adhesions to adjacent loops that conditioned the obstruction. A sac-like structure at the mesenteric side was identified after freeing the strong adhesions and an intestinal duplication was considered (figure 3). An enterectomy encompassing the lesion, with a mechanical enteroenteric laterolateral anastomosis was performed, and the resected piece was sent for histological study.

Histopathological examination was consistent with an ileal cystic duplication with no signs of malignant transformation (figure 4).

OUTCOME AND FOLLOW-UP

The postoperative period had no major complications; the patient was discharged on the 12th day. He remained asymptomatic up to 12-month follow-up.

DISCUSSION

Gastrointestinal duplications are rare congenital malformations found in 1/10 000 live births and account for 0.2% of all malformations in children.^{5 8} The true aetiology of gastrointestinal duplication remains unknown.³ Most patients become symptomatic within the first year of life,

Figure 1 Abdominal CT scan showing loops of dilated proximal small bowel, with some air–fluid content.

CrossMark

To cite: Barbosa L, Soares C, Póvoa AA, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-206638

Figure 2 Abdominal CT scan showing a mass of adherent small bowel loops at ileal level with bowel wall thickening (arrows).

and more than 80% of cases show up before the age of 2 years.^{4 5 9 10} Only 5–6.5% may remain asymptomatic until adulthood, and the diagnosis becomes mostly incidental.⁴

Duplications are consistently located on the mesenteric side of the adjacent alimentary tract, sharing a portion of its wall as well as blood supply, which separate them from mesenteric cysts and intestinal diverticula.^{1–3 10} The definition of an enteric duplication is histological with two distinct types described in the literature: cystic duplications have no communication with adjacent alimentary tract and represent 82% of the cases;

tubular duplications are less common, usually communicating with adjacent bowel, and contain ectopic gastric mucosa.^{3 4}

It has been estimated that approximately 75% of duplications are located within the abdominal cavity; intrathoracic lesions account for 20% of the cases and 5% are thoracoabdominal duplications.^{4 11}

Abdominal lesions often present pain, vomiting, gastrointestinal bleeding related to ectopic gastric mucosa and an abdominal mass that may be palpable in about half of the cases.^{3 5 10} Their first presentation might also be secondary to a complication such as intestinal obstruction, intussusception, volvulus, perforation and peptic oesophageal stricture.^{5 8 10} Malignant transformation is rarely seen in enteric duplications, however, when it does occur, ileal duplications are those most associated with malignancy, showing evidence of epithelial instability growing in long strand duplications. These tumours are often diagnosed at an advanced stage with metastatic disease.^{6 7}

Imaging studies such as ultrasonography, CT scan or even MRI, may be helpful diagnostic tools for detecting intestinal duplications; however, due to their rarity in the adult population, it becomes a challenge to consider them in the differential diagnosis of an abdominal mass.^{5 11}

Recently, some reports have suggested that laparoscope-assisted surgery could be beneficial for both making a diagnosis

Figure 3 Sac-like structure located at the mesentery side of the proximal ileum (arrows).

Figure 4 Histological section of the ileum duplication showing a shared portion of its wall with the adjacent ileum.

and performing accurate treatment of this condition.^{12 13} Despite the chosen surgical approach, the mainstay of treatment relies on complete surgical resection and the overall outcome of surgery is generally favourable.^{2 6 7 11}

Learning points

Intestinal duplication is not currently considered in the differential diagnosis of an abdominal mass when evaluating a symptomatic adult patient. Even with the advances of modern imaging techniques, an accurate preoperative diagnosis is still very difficult mainly due to the rarity and lack of specific symptoms of this condition. The intraoperative surgeon's experience and knowledge play major roles in the recognition of this pathological entity so it can be properly treated.

Contributors CS, AAP and JPM contributed to writing of the manuscript and researched the literature.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Hackam DJ, Zalev A, Burnstein M, *et al.* Enteric duplication in the adult, derived from the foregut, midgut and hindgut: presentation, patterns and literature review. *Can J Surg* 1997;40:129–33.
- 2 Ildstad ST, Tollerud DJ, Weiss RG, *et al.* Duplications of the alimentary tract. Clinical characteristics, preferred treatment, and associated malformations. *Ann Surg* 1988;208:184–9.
- 3 Abubakar AM, Troebbs RB. Gastrointestinal duplications. In: Ameh EA, Bickler SW, Lakhoo K, *et al.*, eds. *Paediatric Surgery: A Comprehensive Text For Africa*, pp. 397–400. Seattle, WA, USA: Global HELP organization. ISBN 978-1-60189-091-7.
- 4 Frering V, Velecela E, Fouque P, *et al.* Upper digestive duplications in adults. *Ann Chir* 1995;49:928–35.
- 5 Fiorani C, Scaramuzzo R, Lazzaro A, *et al.* Intestinal duplication in adulthood: a rare entity, difficult to diagnose. *World J Gastrointest Surg* 2011;3:128–30.
- 6 Blank G, Königsrainer A, Sipos B, *et al.* Adenocarcinoma arising in a cystic duplication of the small bowel: case report and review of literature. *World J Surg Oncol* 2012;10:55.
- 7 Beltrán MA, Barriá C, Contreras MA, *et al.* Adenocarcinoma and intestinal duplication of the ileum. Report of one case. *Rev Med Chil* 2009;137:1341–5.
- 8 Hicham el B, Abdelmalek O, Youssef AL, *et al.* Ileal duplication revealed by a perforation in adults. *Pan Afr Med J* 2010;5:14.
- 9 Gümüş M, Kapan M, Gümüş H, *et al.* Unusual noncommunicating isolated enteric duplication cyst in adults. *Gastroenterol Res Pract* 2011;2011:323919.
- 10 Johnson JA III, Poole GV. Ileal duplications in adults. Presentation and treatment. *Arch Surg* 1994;129:659–61.
- 11 Shah A, Du J, Sun Y, *et al.* Dynamic change of intestinal duplication in an adult patient: a case report and literature review. *Case Report Med* 2012;2012:297585.
- 12 Patiño Mayer J, Bettolli M. Alimentary tract duplications in newborns and children: diagnostic aspects and the role of laparoscopic treatment. *World J Gastroenterol* 2014;20:14263–71.
- 13 Kajiwara Y, Ueno H, Shinto E, *et al.* Enlarged ileal duplication treated by laparoscopy-assisted surgery in an adult. *J Jpn Soc Endosc Surg* 2013;18:567–73.

Copyright 2015 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow