Online Case Report

Iron-deficiency anaemia caused by an enterolith-filled jejunal duplication cyst

George S Tamvakopoulos, Virginia Sams, Peter Preston, William SL Stebbings

Department of Surgery, Norfolk & Norwich University Hospital NHS Trust, Norwich, UK

Enterogenous duplication cysts are rare, but recognised, developmental anomalies. Duplications in the midgut are the commonest of these rare anomalies with a high proportion of them being found in the jejunum. Enterolith formation within these duplication cysts is uncommon as is heterotopic mucosa giving rise to peptic ulceration. Mode of presentation depends on whether symptoms due to mass effect or gastrointestinal bleeding predominate. We present a case of iron-deficiency anaemia caused by an enterolith-filled jejunal duplication cyst diagnosed at laparotomy and treated by surgical excision and small bowel resection.

Key words: Duplication cyst – Iron-deficiency anaemia – Enterolith

A 48-year-old female school-bus driver presented with a 3-month history of loin pain in the absence of any change in bowel habit, excessive menstrual bleeding, or urinary symptoms. On examination, she was clinically anaemic, and routine blood screening revealed hypochromic and microcytic anaemia (Hb 9.8 g/dl, MCV 72.4 fl, MCH 23.4 pg). Barium enema radiography, colonoscopy and oesophago-gastro-duodenoscopy with duodenal mucosal biopsy were normal, as were an intravenous uroterogram and an ultrasound scan of the abdomen.

Barium follow-through radiography (Fig. 1) revealed oval filling defects in the distal ileum suggestive of enteroliths within a diverticulum. There was no evidence of intraluminal strictures and the initial diagnosis was presumed to be enteroliths within a Meckel's diverticulum.

Laparotomy revealed a 10-cm diameter cystic mass containing enteroliths on the mesenteric border at the level of mid-jejunum. The rest of the small bowel appeared normal. A small bowel resection including the cystic mass (Fig. 2) was carried out and an end-to-end anastomosis was performed.

Slicing the cyst revealed its contents to include multiple round, dark stones (Fig. 3). These were subsequently analysed and found to be 100% organic in nature possibly glycine. There was a single 0.3-cm diameter communication between the cystic mass and the adjacent small bowel (Fig. 4). The muscularis propria appeared in continuity between the bowel and the cyst. Microscopically, the cyst was lined with gastric epithelium. The lining of the communication showed pyloric metaplasia with adjacent mucosal erosions. Overall, these features amounted to those of an enterogenous (duplication) cyst lined with heterotopic gastric mucosa.

Postoperatively, the patient recovered well. Three months following discharge from hospital, blood parameters have returned to normal (Hb 11.6 g/dl, MCV 84.7 fl, MCH 28.7 pg).

Discussion

Enteric duplication cysts are rare developmental anomalies of the small intestine thought to arise at about 8–9 weeks of gestation when all layers of the bowel wall are 'pinched off'.¹

Correspondence to: Correspondence to: Dr George S Tamvakopoulos, Department of Surgery, Norfolk & Norwich University Hospital NHS Trust, Colney Lane, Colney, Norwich NR4 7UY, UK. Tel: +44 1603 286423; E-mail: gst@doctors.net.uk

Figure 1 Barium follow-through radiograph showing oval filling defects (arrow) presumed to be enteroliths within a diverticulum. Semicircular line is artefact within the examination paddle.

Figure 2 Resected segment of small bowel and mesentery containing a duplication cyst. The axis of the bowel lumen is on the bottom of the specimen while the duplication cyst is on the mesenteric border and is shown in the centre of this photograph.

Duplication cysts can be found anywhere from mouth to anus, the commonest site being the midgut (50% of all cases), with the foregut and hindgut trailing in distribution (36 and 12%, respectively), while 2% are extra-intestinal (retroperitoneum, spine, oropharynx).⁷

The mode of presentation of these duplication cysts depends on whether mass effect symptoms (pain, obstruction, urinary symptoms) or gastrointestinal bleeding symptoms (anaemia) predominate.^{1,4,6} Gastrointestinal bleeding is more common in duplication cysts which communicate with the bowel lumen and are lined with gastric mucosa.⁷

Time of presentation varies between 16 weeks of gestation to 12 years of age with a median of 3 months, but

presentation in adult life has also been described with a documented prevalence of malignant change.⁷

Duplication cysts are composed of all layers of the bowel wall, *i.e.* submucosa and muscularis propria with their accompanying neural plexi.²

Enterolith formation in the small bowel is thought to be a result of hypomotility or stasis. Stasis promotes bacterial overgrowth, which subsequently leads to decomposition of bile salts, precipitation and concretion. The acidic pH in the duodenum and upper jejunum encourages bile salts to precipitate and mineral salts to stay in solution; enteroliths, thus formed, are radiolucent. The alkaline pH in the distal jejunum and ileum conversely encourages calcium salts to

Figure 3 Dark enteroliths contained within the cavity of the duplication cyst.

Figure 4 Communication point between cyst and bowel lumen.

precipitate and mineralised radiopaque stones to form.⁵ Enteroliths within duplication cysts have been described⁴ and tend to occur more frequently when the duplication cyst develops in the foregut.³

Pre-operative diagnosis of duplication and other mesenteric cysts relies mainly on barium radiography, ultrasonography and CT scanning.³⁻⁵ During an acute presentation, the diagnosis is made at laparotomy.⁷

Treatment of small bowel duplications is usually by surgical excision and small bowel resection. In children, longer tubular duplications can be treated by partial excision and stripping of the mucosa within the duplication, in an attempt to preserve intestinal length.⁷

Conclusions

We present a case of iron-deficiency anaemia caused by an enterogenous duplication cyst. The duplication cyst was communicating with the bowel lumen, was lined with heterotopic gastric mucosa, and was filled with enteroliths.

References

1. Michael D, Cohen CR, Northover JM. Adenocarcinoma within a rectal duplication cyst: case report and literature review. *Ann R Coll Surg Engl* 1999; **81**: 205–6.
2. de Perot M, Bründel M-A, Tötsch M, Mentha G, Morel P. Mesenteric cysts. Toward less confusion? *Dig Surg* 2000; **17**: 323–8.
3. Paige M, Ghahremani G, Brosnan J. Laminated radiopaque enteroliths: diagnostic clues to intestinal pathology. *Am J Gastroenterol* 1987; **82**: 432–7.
4. Zamir G, Gross E, Shmushkevich A, Bar-Ziv J, Durst A, Jurim O. Duodenal duplication cyst manifested by duodeno-jejunal intussusception and hyperbilirubinaemia. *J Pediatr Surg* 1999; **34**: 1297–9.
5. Klingler P, Seelig M, Floch N, Branton S, Metzger P. Small-intestinal enteroliths; unusual cause of small intestinal obstruction. *Dis Colon Rectum* 1999; **42**: 676–9.
6. Arbell D, Lebenthal A, Blashar A, Shmushkevich A, Gross E. Duplication cyst of the duodenum as an unusual cause of massive gastrointestinal bleeding in an infant. *J Pediatr Surg* 2002; **35**: E8.
7. Stringer MD, Spitz L, Abel R, Kiely E, Drake DP, Agrawal M et al. Management of alimentary tract duplication in children. *Br J Surg* 1995; **82**: 74–8.