CASE REPORT

Long-term intestinal bleeding in a child: a rare case of heterotopic gastric mucosa in the jejunum

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SUMMARY

We report a rare case of heterotopic gastric mucosa presenting in a jejunal polyp of a 9-year-old girl, diagnosed with capsule endoscopy and laparoscopic exploration, after a 15-month history of lower gastrointestinal bleeding and anaemia. During surgery, the jejunal segment containing the polyp was resected and pathological examination revealed a polyp with gastric heterotopia. Heterotopic gastric mucosa is a rare pathology characterised by the presence of gastric mucosa outside of the stomach. Heterotopic gastric mucosa usually occurs in the oesophagus, duodenum, Meckel's diverticulum and enteric duplication cysts. It can be either congenital (heterotopic) or acquired (metaplastic). Symptoms vary from mild dyspepsia to massive gastrointestinal bleeding.

BACKGROUND

Heterotopic gastric mucosa (HGM) is a rare pathology characterised by the presence of gastric mucosa throughout the gastrointestinal tract. It consists of a mucosa of specialised gastric glands made of chief and parietal cells. It is supposed that HGM originates from the primitive gut epithelium as a congenital anomaly which dissociates from the primordial stomach at the fourth week of gestation and undergoes hyperplasia over time.^{1 2} It can be classified as congenital (heterotopic) or acquired (metaplastic).^{3 4}

HGM can present itself with a wide range of symptoms depending on its size and location. Common locations include the oesophagus, duodenum and Meckel's diverticulum.⁴ Rarely, as in our case, HGM can occur in the jejunum.⁵ Common symptoms of HGM located in the small bowel are ulceration, bleeding, perforation, intussusception, pain and abdominal distension, but it can also be asymptomatic. Therefore it is very challenging to diagnose HGM preoperatively.

CASE PRESENTATION

A 9-year-old girl was followed up at our hospital for a 15 months history of gastrointestinal bleeding manifesting with melena and mild anaemia without abdominal pain or other subjective discomfort. During this period, her haemoglobin levels ranged between 9.7 and 11.1 g/dL.

The patient was reviewed by a consultant gastroenterologist, examined for suspicion of Crohn's and coeliac diseases: p-ANCA, cASCA and tTG serum antibodies resulted negative. Subsequently an upper endoscopy with biopsy was performed,

ruling out coeliac disease and the presence of *Helicobacter pylori*.

INVESTIGATIONS

As the previous investigations were inconclusive, the patient underwent capsule endoscopy, which showed a polypoid mass in the jejunum and therefore was referred for surgical evaluation.

Through a laparoscopic exploration the whole intestine could be examined and we could identify and resect the bowel loop of interest, in this case the proximal jejunum.

TREATMENT

Under general anaesthesia, the patient underwent laparoscopic exploration. We employed a 3-port technique: a 10 mm umbilical port, a 5 mm port in the left iliac fossa and a 5 mm suprapubic port. The small bowel was inspected from the terminal ileum to the first jejunal loop. A dilated segment of the jejunum at 30 cm from the Treitz ligament with an intraluminal bulk was identified and delivered through the umbilical incision (figure 1A). An enterotomy was then performed which revealed a 4 cm long intraluminal bulk (figure 1B). A 12 cm long segment was resected and an end-to-end anastomosis was performed. No other intestinal alterations were recognised. Histological examination of the specimen revealed a 4 cm polyp lined with gastric mucosa (figure 2).

OUTCOME AND FOLLOW-UP

The patient was discharged on the 8th post-operative day and at 1 month follow-up an abdominal ultrasound scan was negative.

DISCUSSION

HGM has been repeatedly reported to locate throughout the whole gastrointestinal tract. Rare

Figure 1 (A) Segment of jejunum with palpable mass delivered through the umbilical incision. (B) Single 4 cm long polyp uncovered during enterotomy.

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Figure 2 Low-power view of a typical gastric hyperplastic polyp with hyperplastic foveolae, cystic mucus-cell lined glands, oedematous lamina propria contains inflammatory cells (H&E; $\times 2$).

but reported locations include airways, umbilicus, urinary bladder and scrotum. Clinical presentation depends on its size and location. In the small bowel, the most common symptoms of HGM are: ulceration, bleeding, perforation, intussusception, pain and abdominal distension, although it can be asymptomatic. HGM presenting as a polypoid mass in the jejunum causing gastrointestinal bleeding is very uncommon, like in our case, with very few reports available.^{4 5} Diagnosis can be therefore very challenging.

Upper and lower endoscopy is still limited to the duodenum and colon, hence it is not suitable to diagnose small bowel alterations. Abdominal ultrasound scan, CT or MRI can rarely reveal an intussusception or an intraluminal polypoid mass.^{4 6} In the past, 99 m Tc-pertechnetate scintigraphy was suggested as a potential diagnostic tool because of its strong affinity for gastric mucosa, although of late it has been abandoned because of its low specificity and sensitivity. Instead, capsule endoscopy is a good diagnostic option as it can explore the whole gastrointestinal tract. It has been reported to have a high positive diagnosis rate (range from 45% to 76%) for the recognition of pathology in the small bowel compared with other methods. Although, as in our case, HGM is often discovered intraoperatively, where only pathological examination

of the resected segment could establish the definitive diagnosis.^{2 3 7}

Ultimately laparoscopy is the standard procedure for gastrointestinal bleedings that remain undiagnosed following upper and lower endoscopy because of its advantages such as small incisions, reduced pain and bleeding, smaller scars, reduced risk of postoperative infection and shorter length of hospital stay.

Learning points

- ▶ Although rare, small bowel heterotopic gastric mucosa (HGM) should be considered in the differential diagnosis of long-term gastrointestinal bleeding in infants and children.
- ▶ HGM can also occur in locations not visible with standard endoscopy. Therefore use of capsule endoscopy and laparoscopic exploration should be considered.
- ▶ A laparoscopic approach allows diagnosis and reduces postoperative complications.

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