Case report

Bleeding from multifocal heterotopic gastric mucosa in the colon controlled by an H₂ antagonist

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SUMMARY The sixth documented case of heterotopic gastric mucosa in the large bowel proximal to the rectum is described in a two year old girl with a neural tube defect and recurrent rectal bleeding. Unusual in itself, this case is unique in that the rectal bleeding has been controlled with an H₂ receptor antagonist.

Heterotopic gastric mucosa involving the hindgut is exceedingly rare, especially at sites other than the rectum. The management of all cases described to date has involved surgical excision of the affected bowel. A new approach to management is presented in this case report.

Case history

After an uncomplicated pregnancy, this young girl was born at full term, weight 4.5 kg, with a 3×3 cm lumbar myelomeningocele containing a small neural stalk. This was successfully repaired and moderate hydrocephalus controlled with oral isosorbide dinitrate.

An anteriorly placed anus functioned well until six months of age, when bleeding per rectum and constipation developed. An anoplasty was carried out followed by regular dilatation.

At one month, two months, four months, and 16 months rectal bleeding recurred. Haemoglobin was 5.4 g/dl. Barium meal and follow through, barium enema, coagulation screen and repeated stool examination for ova, parasites, culture and sensitivity were normal and negative respectively. Technetium scan revealed increased isotope uptake in the right iliac fossa suggestive of Meckel's diverticulum (Fig. 1).

Laparotomy revealed a 1 cm Meckel's diverticulum, lined with normal small bowel mucosa which was removed. In addition, an ulcer with surrounding induration was excised from the proximal ascending colon with histology revealing a localised ulcer in inflamed colonic mucosa.

Colonoscopy revealed a polypoid oedematous lesion in the rectum, prominent hyperaemic fold in the sigmoid region, and ulcerated swollen mucosa in the ascending colon and caecum. Biopsy of each of these lesions revealed heterotopic body type gastric mucosa (Fig. 2). Oral ranitidine 15 mg bd was started which initially controlled the bleeding. Repeat colonoscopy, four months later, showed a reduction in the size of the lesion and healing of the ulcerated areas. After a small episode of bleeding five months after initiation of therapy, the dose of ranitidine was increased to 30 mg bd (4 mg/kg/day). No further bleeding has recurred. This standard dose of ranitidine has fully controlled the bleeding to date.

Discussion

In this case report, a young girl is described with repeated episodes of rectal bleeding because of heterotopic gastric mucosa present in the rectum, sigmoid colon, ascending colon, and caecum. The widespread distribution of the lesions made conservative surgery difficult. The patient was treated successfully with an H₂ receptor antagonist, ranitidine.

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Fig. 1 Technetium scan showing increased uptake of radioisotope in the right lower abdomen suggestive of Meckel's diverticulum (MD). Also shown: thyroid (T), stomach (S), and urinary bladder (B).

dine, in an effort to reduce acid secretion and thus reduce ulceration and bleeding. This is the first described case of medical therapy controlling haemorrhage from heterotopic gastric mucosa, and suggests that the mechanism controlling secretion by heterotopic mucosa may be similar to that operating in normal gastric mucosa.

Heterotopic gastric mucosa has been reported all along the alimentary tract, including tongue, oesophagus,¹⁻³ small intestine,⁴⁻⁷ biliary tract,⁸⁻⁹ gall bladder,¹⁰ colon and rectum,¹⁵⁻³¹ the most common being in Meckel's diverticulum. In addition, heterotopic gastric mucosa has been described in the mediastinum,¹¹ face,¹² spinal cord,¹³ and umbilicus.¹⁴

Gastric heterotopia in segments of the alimentary tract derived from the fetal hindgut is extremely rare. Review of the literature has revealed 10 cases involving the rectum alone (Table 1a), four having an associated reduplication of the rectum; and only five cases involving the colon proximal to the rectum (one of whom also had rectal involvement and is included in Table 1a) – Table 1b.¹⁵⁻³¹

Five of these 19 reported cases have had anomalies of other systems: vertebral (four), digital (one), and other heterotopias. All cases, except one,²⁰ have shown fundic type gastric mucosa. Only six previous cases have been reported in children.

The majority of cases presenting with gastric heterotopia involving the rectum present with rectal bleeding (Table 1a), and a mass or rectal ulceration may be seen on sigmoidoscopy.

Table 1a Summary of reported cases of heterotopic gastric mucosa in rectum

Age/sex	Presenting symptom	Duration of symptoms	Sigmoidoscopic findings	Type of mucosa	Associated anomalies	Reference
4/F	Rectal bleeding	2 yrs	Rectal diverticulum	Fundic	Rectal duplication	15
45/M	Rectal bleeding	4 yrs	Polypoidal tumour	Fundic + pyloric	None	16
26/F	Rectal bleeding + tenesmus	16 yrs	Polypoidal tumour	Fundic	None	17
26/M	Rectal bleeding	2 yrs	Polypoidal tumour	Fundic	Rectal diverticulum Meckel's diverticulum Colonic rotation Vertebral defects	18
19/F*	Rectal bleeding + diarrhoea	7 days	Multifocal ulcers	Fundic + pyloric	Malrotation Vertebral anomalies Megacolon	19
7/M	Rectal bleeding	—	Polypoidal tumour	Fundic	None	20
51/M	Rectal bleeding/ulceration	3 mo	Ulcer	Pyloric	None	20
46/F	Rectal Mass	2½ mo	Polyp	Fundic	None	20
24/M	Rectal bleeding + pain	3 yrs	Normal	Fundic + salivary	Rectal diverticulum Vertebral anomalies	21
14/M	Rectal bleeding	12 yrs	Ulceration	Gastric	None	22
24/M	Rectal bleeding	Not stated	Polypoidal tumour	Fundic	None	23
22/M	Rectal bleeding + tenesmus	6 days	Polypoidal tumour	Fundic	None	24
6 mo/M	Rectal bleeding	4 mo	Polypoidal tumour	Fundic	None	25
4/M	Rectal bleeding	1 yr	Ulcer	Fundic	Rectal duplication	26
16/M	Rectal bleeding + pain	5 yrs	Polypoidal tumour	Fundic	None	27

*Also had proximal colonic involvement (see Table 1b)

Fig. 2 Heterotopic gastric body type mucosa (G) adjacent to normal colonic mucosa (C). H&E.

In contrast, in those patients with lesions proximal to the rectum, the clinical presentation is more varied with diarrhoea (two cases), rectal bleeding (one case), acute appendicitis (one case), and intussusception (one case). Rectal bleeding is less prominent in this group. There has only been one previous case report describing both proximal and distal large bowel involvement,¹⁹ it is noteworthy that this patient also had spina bifida.

The aetiology of colonic heterotopia is uncertain. Although gastric heterotopia in foregut structures

may be caused by dislocations of part of the embryonic structures during developmental descent of the stomach, such an explanation is not tenable in the hindgut. It has been suggested that it may arise as a result of abnormal differentiation of local tissues (heteroplasia) based on the concept that all the cells lining the primitive intestinal canal are pluripotential.³²

It has also been suggested that the condition may be acquired, and represents an abnormal regenerative process after mucosal destruction.³³ A develop-

Table 1b Summary of reported cases of heterotopic gastric mucosa in the colon, proximal to the rectum

Age/sex	Presenting features	Duration of symptoms	Segment of colon involved	Type of heterotopic tissue	Associated anomalies	Reference
4/F	Colocolic intussusception	Not stated	Transverse	Gastric duodenal appendiceal pancreatic	None	28
19/F	Not stated	Not stated	Ascending	Gastric	None	29
0/M	Watery diarrhoea	From birth	Caecum, ascending colon	Gastric + oesophageal	Rib + vertebrae	30
14/M	Acute appendicitis	1 day	Appendix	Gastric	None	31
19/F*	Diarrhoea rectal bleeding	7 days	Ascending transverse descending colon	Fundic + pyloric	Spina bifida occulta Malrotation Megacolon	19

*Also had rectal involvement (see Table 1a)

mental aetiology appears the more likely in view of the association with neural tube and other abnormalities.

All the previous cases described have been managed by surgical excision of the affected area. Because of the widespread colonic extent of the abnormality in this case, medical therapy using an H₂ receptor antagonist was first attempted. This has resulted in immediate and adequate control of haemorrhage (with one small episode of breakthrough bleeding), and healing of ulceration and decrease in size of the lesions noted at colonoscopy, allowing the child to continue without colonic resection. The mechanism of action of ranitidine in this case is open to debate. The rationale behind its use was that the heterotopic gastric mucosa, like that in the stomach, has H₂ receptors responsible for control of acid output, which could be blocked by appropriate drug therapy, decreasing acid secretion and thus colonic ulceration and bleeding. The success of this therapy suggests that this may well be the case.

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