Case report

Bleeding from multifocal heterotopic gastric mucosa in the colon controlled by an $H_2$ 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 $H_2$ 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 occurred. 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 $H_2$ receptor antagonist, ranit-
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,\(^\text{13}\) small intestine,\(^\text{14}\) biliary tract,\(^\text{14}\) gall bladder,\(^\text{19}\) colon and rectum,\(^\text{18-21}\) the most common being in Meckel's diverticulum. In addition, heterotopic gastric mucosa has been described in the mediastinum,\(^\text{11}\) face,\(^\text{12}\) spinal cord,\(^\text{13}\) and umbilicus.\(^\text{14}\)

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.\(^\text{15-31}\)

Five of these 19 reported cases have had anomalies of other systems: vertebral (four), digital (one), and other heterotopias. All cases, except one,\(^\text{26}\) 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

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

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

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

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

*Also had rectal involvement (see Table 1a)
mental aetiology appears the more likely in view of the association with neural tube and other abnormali-
ties.

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 break-
through 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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